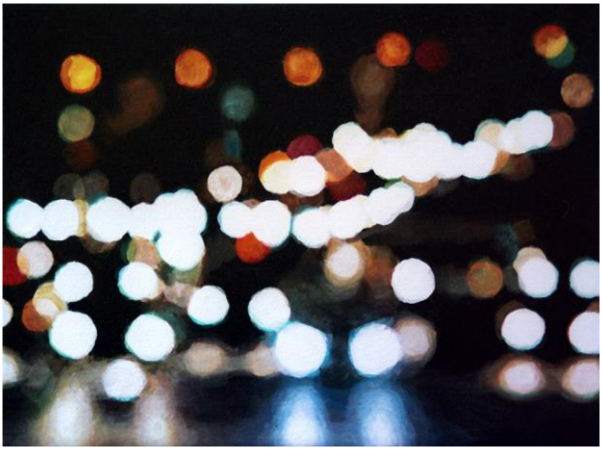
Secteur des Sciences de la Santé

Institut de Recherche Santé et Société

Spice of life
How can we explore and measure the quality of life of children facing life-limiting conditions?
Results and challenges of the MOSAIK study in Belgium

Marie Friedel

Thesis presented to obtain the degree of Doctor in Public Health Sciences
Université catholique de Louvain
Promotors : Pr. Isabelle Aujoulat and Pr. Jean-Marie Degryse



City Lights (2009) by David Russon

reproduced with permission

« On ne saisit pas ce qui nous saisit. »

P. de la Tour du Pin

This thesis is dedicated to

each **child, adolescent and parent** who shared with me, at the time I was a paediatric liaison nurse and during this research, what their spice of life is;

especially to **Luc Brilot**, who taught me the importance of providing holistic care, without ever separating the interconnected physical, psychological, social and spiritual dimensions of a human being. His gift reminded me all along this thesis to stay focused on the holistic quality of life;

my 'PhD godmother', **Evelyne d'Hoop-Jonckheere** (ǂ 2017), who revised the PhD proposal, and my friends, **David Grünberg** (ǂ 2020), **Laura Guttuso** (ǂ 2016) and **Anne-Sophie Jung** (ǂ 1985), who have sent me, from wherever they rest now, patience and energy;

Jean, **Flore** and **Timoteo**, my cherished children, who endured the burden of living with a doctoral student-mother. I hope you will understand some day why I was so often far away from you;

Gilberto, my husband, love of my life, who shares my hopes and my challenges. He accepted, with faith, to leave his beloved Italy to follow me on this PhD adventure. An unexpected adventure that has turned our family upside down. I will be ever grateful for his decision.

"Palliative care is like a dance, a dance that requires you to adapt your step to the other person's..." Dr Matthias Schell

This thesis journey was a dance, sometimes slow, jerky, hesitant, even painful, but sometimes also frenetic, joyful and exciting and certainly full of surprises. I would like to deeply thank here the many people who have entered the dance.

First, I would like to express my warmest thanks to the **Marguerite-Marie Delacroix Fund** for their confidence in this research and their financial support since its inception in 2015. Furthermore, I am sincerely grateful to **Fondation Louvain** who has contributed to the MOSAIK Fund, to **Fondation contre le Cancer**, which funded the study on the number of children with chronic complex condition and to **Haute Ecole Léonard de Vinci**, for giving me time to conduct this research. Without their support, this thesis would never have begun, nor would we have been able to obtain the results we did.

To my mentors, Prof. **Julia Downing**, nurse, PhD, chief executive at the International Children's Palliative Care Network (ICPCN), who showed me, through her inspiring professional life what the word "advocacy" meant;

Dr. Richard Hain, Consultant and Clinical Lead in Paediatric Palliative Medicine in Wales with expertise in clinical paediatric palliative care, pharmacology, professional education and clinical ethics. Thanks to his trust, I had the opportunity in 2015 to deliver my first invited presentation at Cardiff University in a very hesitant English and despite my completely inappropriate dress code for a conference in the UK.

Joan Marston, nurse and founder of the Sunflower children's hospice in South Africa and the ICPCN; who teaches me how to integrate compassion in everyday life;

Dr. Matthias Schell; paediatric oncologist, founder and coordinator of the interuniversity Diploma in acute/chronic paediatric pain and paediatric palliative care at university of Lyon, France;

Sonja Develter, paediatric nurse, who started the paediatric liaison team, Interface pédiatrique at Cliniques Universitaires saint Luc, Brussels, with so much dedication and perseverance and founded the respite care service, Villa indigo in Brussels, Belgium. She transferred in 2016, when I started this PhD, all her books and documents in paediatric palliative care to my new office at IRSS. What a legacy!

I am also grateful to **Dr Chantal Wood**, who introduced me to the world of paediatric palliative care during the first ICPCN Conference in Mumbai, India, in 2014;

to Prof. Dominique Jacquemin, nurse and PhD in Public Health who first listened to my moral distress in February 2014 and encouraged me to start a PhD; **Prof. Mylène Botbol-Baum**, professor in ethics, who accepted initially to be my supervisor when I still only had a 6-page draft;

to Jenny Hunt, incredible social worker and bereavement consultant in Zimbabwe and **Shelagh Mc Connell**, neonatal nurse PhD in Canada. These 2 persons played an important role in this PhD journey. Jenny edited many of the very first conference papers/posters and Shelagh gave me the secret key to completing a PhD, stating that it was just all about perseverance!

to Sue Boucher in South Africa, **Danai Papadatou**, professor for clinical psychology and founder of the Greek society of palliative care, **Natasha Pedersen** founder of the Norvegian Association for children's palliative care, **Joana Mendes**, neonatal nurse and PhD in Portugal, **Loris Bonetti** and **Ivan Favarin** in Italy. Your commitment in clinical practice, research or health services has inspired me a lot, especially your strength during this pandemic.

And to all my wonderful colleagues **Afi Agboli**, **Lucia Alvarez**, **Thérèse Van Durme**, **Olivier Schmitz, Maria Rodrigues Leal Moitinho de Almeida, Charlotte Fyon** at IRSS-UCLouvain and **Fabienne Liénard**, **Véronique Grégoire**, **Ingrid Mouton, Bénédicte Lejeune, Nataly Filion, Sophie Ghysselinckx, Marie des Neiges Grignard and Cédric Juliens** at HE Vinci, for their informal support, to share my joy and sometimes my fatigue; to my teachers, now retired, who instilled to me the joy of doing research: **Dominique Lemenu, Alix Fierens, Andrée Meyer, Anne-Marie Gérard, Françoise Cerexhe.**

To all the colleagues from the administrative staff at IRSS-UCLouvain who helped me out so many times in very different ways: **Nathalie Chaidron**, for her infinite open door, **Mostafa Berdii** for his endless patience with my computer issues, **Djamila Taimount**, **Sophie Jassogne**, **Valérie Van Butsele**, **Maria Cymbron** and a special thanks to **Nathalie Malevé**, who formatted this written thesis in the midst of the summer holidays!

A special thanks goes to **Frederika Roberts**, who agreed to check this thesis for English accuracy within a very short timeframe.

I would like to thank also each person who collaborated on the different projects:

Each member of the six paediatric liaison teams in Brussels, namely the team **Globul'home, Interface pédiatrique, Equipe soins continus pédiatrique CHR Liège, Equipe soins palliatifs pédiatrique CHC Liège, Koester and Kites team** for their trust and collaboration since the beginning, without which this research would not have been possible;

Saskia Van Bogaert, Flore Alix, Katrien Moens for their openness to share the data of the annual paediatric liaison team report's at Cancer Plan;

Aurélie Gilson, Dr Dominique Bouckenaere, Thomas Wojcik, Dr Christine Fonteyne, Pr. Bénédicte Brichard, Nicolas Donnez, Alaa Mahboub, Alain Guillet, Etienne De Clerck and Magali Lahaye for their collaboration within the very complex study looking at the access to paediatric liaison care teams for children with chronic complex conditions in the Brussels region;

Anne-Catherine Dubois for her contribution to the systematic review, but also for patiently listening to my stories;

Mandana Mehta, **Pauline De Becker**, and **Claire Mougenot**, who contributed to the linguistic and semantic forward/backward translation of the Children's palliative outcome scale (CPOS) into French. **Indy Machiels** for his technical help with Filemaker at the beginning.

To my successive heads of department and directors of Parnasse-ISEI, Haute Ecole Léonard de Vinci, who trusted me for this project : **Anne Melery, Sophie Breedstraet, Florence Orlandi, Annick Jacobs, Valérie Biéva**;

I am hugely grateful also to the members of my PhD steering committee:

Pr. Bénédicte Brichard, a very special gatekeeper and indefectible coach,

Pr. Stéphan Clément de Cléty, for his profoundness, his wise advices and reassuring support since the very beginning;

Pr. Stéphan Vanden Broucke, for his fineness and encouragements;

Pr. Vincent Lorant, for his sincerity and thought-provoking comments;

A special thanks go to the external members of my thesis jury: **Pr. Anne-Marie Etienne**, **Pr. Lorna Fraser** and **Dr Richard Hain**, for their commitment, availability and acceptance to read, discuss and evaluate this thesis, despite their full agenda. I feel very honoured to have you as external jury members.

And last but not least, my sincere gratitude goes to my promotors, **Prof. Isabelle Aujoulat** and **Prof. Jean-Marie Degryse,** for their trust, patience, flexibility and indefatigable support, with a rebel student such as me. I feel privileged for having benefitted from their high expertise. We, three, formed a very particular triangulation dance design, which has certainly made this research so rich, challenging and complex. Thank you for having taught me the full nuances of sciences.

Table of contents

Summary

The core goal of paediatric palliative care (PPC) is to improve the quality of life (QoL) of children and their parents but defining and assessing this construct for children with life-limiting conditions is challenging.

The objectives of this MOSAIK (Move to open shared advanced interventions for Kids with life-limiting conditions) research consisted in documenting the characteristics of children and their access to paediatric liaison teams (PLTs) in Belgium, adapting an instrument able to evaluate children's and parents' quality of life, assessing the psychometric properties of this instrument and analysing how its use was perceived among paediatric liaison teams.

A systematic review looked at instruments used in PPC. We used a mixed-methods convergent triangulation design to confront the perspectives of children, parents and PLTs on their QoL. A pilot-test assessed the face and content validity, feasibility and acceptability of the further developed children's palliative outcome scale (CPOS-2) in French. Finally, we conducted a multicentric national field-test among six PLTs, in French and Dutch, to document the reliability and concurrent validity of the CPOS-2, to provide an overview of children followed-up and to document the experiences of PLTs when using the CPOS-2.

Results showed that more than 700 children /adolescents are referred to PLTs on an annual basis, but, between 2010 and 2014, only 1.7% of children facing complex chronic conditions and admitted to hospitals in the Brussels region had access to these. We also found a scarcity of outcome measurement instruments in PPC and that existing ones do not include children's self-reports. Our study, conducted among 73 families, led to an adapted 20-item CPOS-2 able to evaluate children's and parental quality of life (QoL), showing satisfactory reliability and concurrent validity. Quality of life scores do not seem to be linked to the severity of a child's disease. Involving the main stakeholders during the whole process helped to develop a sound instrument. Beyond the metrics, the CPOS-2 is perceived by PLTs as a compass to deepen the relation with families, to address unmet needs, to engage in difficult conversations and to confirm their intuition on the level of suffering and care needs.

PPC teams in France have already manifested their interest in using the CPOS-2, leading to a larger sample size of children included which will help us to further document its validity and reliability. Evaluating the quality of life of children with serious illnesses in a family-centred approach with the CPOS-2 might improve the quality of care provided by PLTs.

Key-words: Children's Palliative Outcomes Scale version 2 (CPOS-2), Family-centred care, Implementation study, Life-limiting conditions, Mixed-methods study design, Outcome measurement instrument, Paediatric Liaison teams, Paediatric Palliative Care, Psychometrics, Quality of Life.

Synthèse

L'objectif principal des soins palliatifs pédiatriques (SPP) est d'améliorer la qualité de vie des enfants et de leurs parents, mais la définition et l'évaluation de ce concept restent difficiles. Les objectifs de cette recherche MOSAIK (Move to Open Shared Advanced Interventions for Kids with life-limiting conditions) visent à documenter les caractéristiques des enfants et leur accès aux équipes de liaison pédiatrique (ELP) en Belgique, à adapter un instrument capable d'évaluer la qualité de vie des enfants et des parents, à évaluer les propriétés psychométriques de cet instrument et à analyser comment son utilisation est perçue par les équipes de liaison pédiatrique.

Pour cela, une revue systématique a été réalisée pour identifier des instruments utilisés en SPP. La méthode générale utilisée consistait en un design de triangulation convergente à méthodes mixtes pour confronter les perspectives des enfants, des parents et des ELP. En outre, nous avons combiné des instruments standardisés et individualisés de la qualité de vie. Une étude pilote a permis d'évaluer la validité de face, la validité de contenu, la faisabilité et l'acceptabilité d'une 2ème version du children's palliative outcome scale version 2 (CPOS-2), adapté dans sa version francophone. Enfin, nous avons mené une étude nationale multicentrique auprès des 6 ELP, en français et en néerlandais, afin de documenter la fiabilité et la validité concurrentielle du CPOS-2, de fournir un aperçu des enfants suivis et de documenter les expériences des ELP lors de l'utilisation du CPOS-2 auprès des familles.

Les résultats ont montré que plus de 700 enfants/adolescents sont référés chaque année vers les ELP, mais que seulement 1,7% des enfants confrontés à une maladie chronique complexe et admis dans les hôpitaux de la région bruxelloise y ont eu accès entre 2010 et 2014. Nous avons également constaté que les instruments évaluant les résultats des SPP sont rarement utilisés chez les enfants pour évaluer eux-mêmes leur qualité de vie, et s'ils le sont, leurs propriétés psychométriques font défaut. Notre étude, menée auprès de 73 familles, a débouché sur une version révisée du CPOS-2 en 20 items, capable d'évaluer la qualité de vie des enfants et des parents, montrant une fiabilité et une une validité concurrentielle satisfaisante. Il ne semble pas avoir de liens entre les scores de qualité de vie et la sévérité de la maladie de l'enfant. L'implication des principales parties prenantes tout au long du processus a permis de développer un instrument robuste et a contribué à sa mise en œuvre dans les soins cliniques de routine. Au-delà des mesures, le CPOS-2 est perçu par les CPL comme une boussole permettant d'approfondir la relation avec les familles, de répondre aux besoins non satisfaits, d'engager des conversations difficiles et d'objectiver leur intuition sur le niveau de souffrance et le type de soins requis.

Huit équipes régionale ressources de SPP en France utilisent actuellement le CPOS-2, ce qui va permettre d'élargir la taille de l'échantillon d'enfants inclus, et permettra de mieux documenter la validité et la fiabilité de l'instrument. L'évaluation de la qualité de vie des enfants atteints de maladies graves dans une approche centrée sur la famille avec le CPOS-2 pourrait améliorer la qualité des soins fournis par les ELP.

Mots-clés: Children's Palliative Outcome Scale version 2 (CPOS-2), Conditions limitant la vie, Design méthodes mixtes, Etude d'implémentation, Equipes de liaison pédiatrique, Instrument de mesure, Psychométrie, Soins centrés sur la famille, Soins palliatifs pédiatriques, Qualité de vie.

List of abbreviations

AIDS Acquired immune deficiency syndrome

APCA-CPOS African Palliative Care Association children’s palliative outcome scale

CCC Complex chronic condition

CPOS-2 Children’s palliative outcome scale version 2

CTT Classical test theory

D-study Decision study

eHealth platform electronic platform of the Public federal institution which mission is to promote and sustain the safe exchange of electronic information’s between health care professionals

G-analysis Generalizability analysis

GT Generalizibility theory

IQR Interquartile range

KINDL Fragebogen für KINDer und Jugendliche zur Erfassung der gesundheitsbezogenen Lebensqualität

LLC/LTC Life-limiting condition/life-threatening condition

MOSAIK Move to Open Shared Advanced Interventions for Kids with Life-limiting conditions

PedsQL 4.0 Paediatric Quality of life questionnaire version 4

PLTs Paediatric liaison teams

PPC Paediatric Palliative Care

QoL Quality of Life

QOLLTI-F Quality of life in life-threatening illness-Family carer

SD Standard Deviation

SEIQoL-dw Scheduled evaluation of individual Quality of Life-direct weighted

SEM Standard error of measurement

SPSS Software package for statistical analysis

TTP Third Trusted Party

List of tables and figures

List of publications

Publications linked to the thesis

Friedel, Marie ; Brichard, Bénédicte ; Boonen, Sabine ; Tonon, Corinne ; de Terwangne, Brigitte ; Bellis, Dominique ; Mevisse, Muriel ; Fonteyne, Christine ; Jaspard, Maud ; Schruse, Marie ; Harding, Richard ; Downing, Julia ; Degryse, Jean-Marie ; Aujoulat, Isabelle. Face and content validity, acceptability and feasibility of the adapted version of the Children's Palliative Outcome Scale (CPOS-2): a qualitative pilot study. In: Journal of Palliative Medicine,(2020).Jul 15. doi: 10.1089/jpm.2019.0646. http://hdl.handle.net/2078.1/231142

Friedel, Marie ; Aujoulat, Isabelle. Quel accès aux soins palliatifs pour les enfants porteurs d'une maladie chronique complexe en Belgique?. In: Revue multidisciplinaire d'oncologie et hématologie, Vol. 14, no.1, p. 3 (2020). http://hdl.handle.net/2078.1/231170

Friedel, Marie ; Gilson, Aurélie ; Bouckenaere, Dominique ; Brichard, Bénédicte ; Fonteyne, Christine ; Wojcik, Thomas ; De Clercq, Etienne ; Guillet, Alain ; Mahboub, Alaa ; Lahaye, Magali ; Aujoulat, Isabelle. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. In: BMJ Paediatrics Open, Vol. 3, no.1, p. e000547 (2019). doi:10.1136/bmjpo-2019-000547. http://hdl.handle.net/2078.1/220225

Friedel, Marie ; Aujoulat, Isabelle ; Dubois, Anne-Catherine ; Degryse, Jean-Marie. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review.. In: Paediatrics, Vol. 143, no. 1, p. 22p. (2019). doi:10.1542/ peds.2018-2379. http://hdl.handle.net/2078.1/208885

Friedel, Marie. La qualité de vie des enfants en contexte palliatif, comment la documenter? Peut-on la mesurer? . In: Médecine Palliative, Vol. 18, no.4-5, p. 184-197 (2019). doi:10.1016/j.medpal.2019.03.004. http://hdl.handle.net/2078.1/231167 2018

Friedel, Marie ; Brichard, Bénédicte ; Fonteyne, Christine ; Renard, Marleen ; Misson, Jean-Paul ; Vandecruys, Els ; Tonon, Corinne ; Verfaillie, Françoise ; Hendrijckx, Georgette ; Andersson, Nathalie ; Ruysseveldt, Ilse ; Moens, Katrien ; Degryse, Jean-Marie ; Aujoulat, Isabelle. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014.. In: BMC Palliative Care, Vol. 17, no. 1, p. 77 [1-11] (2018). doi:10.1186/s12904-018-0324-2. http://hdl.handle.net/2078.1/198432

Conference papers and posters linked to the thesis

Friedel, Marie ; Gilson, Aurélie ; Bouckenaere, Dominique ; Brichard, Bénédicte ; Fonteyne, Christine ; Wojcik, Thomas ; Mahboub, Alaa ; Lahaye, Magali ; Aujoulat, Isabelle. Making the invisible more visible: a retrospective study identifying children and adolescents with complex chronic conditions and their access to specialised palliative care teams. 16th World Congress of the European Association of Palliative Care (EAPC) (Berlin, Germany, du 22/05/2019 au 24/05/2019). In: BMJ- Paediatrics Open, Vol. 3, no.1, p. e000547 (2019). doi:10.1136/bmjpo-2019-000547. http://hdl.handle.net/2078.1/231705 2018

Friedel, Marie ; Degryse, Jean-Marie ; Dubois, Anne-Catherine ; Aujoulat, Isabelle. Do we evaluate the outcomes of our interventions in paediatric palliative care? A systematic review.3rd International Childrens Palliative Care Network (ICPCN) Conference: Innovation, Integration, Inspiration. (Durban,South Africa, du 29/05/2018 au 31/05/2018). In: Instruments to measure Outcomes in Paediatric Palliative Care: a systematic review., Vol. 143, no.1, p. e20182379 (2019). doi:10.1542/peds.2018-2379. http://hdl.handle.net/2078.1/231704

Friedel, Marie ; Brichard, Bénédicte ; Aujoulat, Isabelle. Evaluating what we intend to reach: How should we measure children's quality of life in a palliative context? Searching for valid indicators of quality palliative care for children and their families. 10th Biennial Joanna Briggs Institute Colloquium 2018. Successful implementation of Evidence-based practice: Hard work or good luck? (Antwerp, Belgium, 04/05/2018). In: Journal of Palliative Medicine, (2018). http://hdl.handle.net/2078.1/231706

Friedel, Marie ; Brichard, Bénédicte ; Degryse, Jean-Marie ; Aujoulat, Isabelle. How can we evaluate quality of life of children with life- limiting conditions? A pilot-study at one PPC center in Brussels. 3rd ICPCN Children's Palliative Care Conference: Innovation Integration, Inspiration (Durban, South Africa, du 28/05/2018 au 31/05/2018). In: Journal of Palliative Medicine, (2018). http://hdl.handle.net/2078.1/231707

Friedel, Marie ; Gilson, Aurélie ; Aujoulat, Isabelle. Recensement d'un nombre d'enfants atteints d'une maladie chronique complexe en région de Bruxelles-Capitale (rapport final), 2018. 57p. p. http://hdl.handle.net/2078.1/214135 2017

Friedel, Marie ; Aujoulat, Isabelle. La qualité de vie des enfants en contexte palliatif: comment la documenter? Peut-on la mesurer? 8ème Congrès International du Réseau Francophone de Soins Palliatifs Pédiatriques: faim de vie, fin de vie (Palais des congrès, Liège, Belgique, du 03/10/2018 au 05/10/2018). In: Médecine Palliative, Vol. 18, no.4-5, p. 184-187 (2018). doi:10.1016/j.medpal.2019.03.004 . http://hdl.handle.net/2078.1/231708

Friedel, Marie ; Aujoulat, Isabelle ; Brichard, Bénédicte. Des soins de liaison pour accompagner l'enfant en phase palliative et sa famille : Résultats d'une étude rétrospective auprès des 5 équipes belges de liaison pédiatrique. 22ème colloque GERIMOC (Libramont, Belgium, 23/09/2017). http://hdl.handle.net/2078.1/231717

Friedel, Marie ; Brichard, Bénédicte ; Fonteyne, Christine ; Misson, Jean-Paul ; Renard, Marleen ; Vandecruys, Els ; Degryse, Jean-Marie ; Aujoulat, Isabelle. Hidden Part of the Iceberg: Making Paediatric Palliative Care in Belgium visible. 3rd International Maruzza Conference on Paediatric Palliative Care (Rome,Italy, du 16/11/2016 au 19/11/2016). http://hdl.handle.net/2078.1/231703

Friedel, Marie ; Brichard, Bénédicte ; Fonteyne, Christine ; Misson, Jean-Paul ; Renard, Marleen ; Vandecruys, Els ; Degryse, Jean-Marie ; Aujoulat, Isabelle. Integrated Paediatric Palliative Care in Belgium: The Paediatric Liaison Model. InSupC-RN Symposium EAPC (European Association of Palliative Care) Research Network (Brussels, Belgium, 30/09/2016). http://hdl.handle.net/2078.1/231701

Friedel, Marie ; Brichard, Bénédicte ; Fonteyne, Christine ; Misson, Jean-Paul ; Renard, Marleen ; Vandecruys, Els ; Aujoulat, Isabelle. The Belgian Liaison model of Paediatric Palliative Care: First findings of a retrospective study based on a national database (2010-2014). 2nd International Children's Palliative Care Network Conference (ICPCN) (Buenos Aires, Argentinia, du 18/05/2016 au 21/05/2016). http://hdl.handle.net/2078.1/231700 2015

Publications not directly related to the thesis but to palliative care

Friedel, Marie ; Hunt,Jenny ; Talawadekar, Pradnya. Anticipatory grief and bereavement support (Chapter 16). In: Julia Downing, Children's Palliative Care: an International Case-Based Manual, Ed. Springer, Springer, 2020, p. 201-210. 978-3-030-27375-0. doi:10.1007/978-3-030-27375-0_16. http://hdl.handle.net/2078.1/231146

Friedel, Marie. La liaison pédiatrique. Un modèle original de soins palliatifs pédiatriques transmuraux. In: De Berre R. (coord.), Manuel de soins palliatifs, Ed. Dunod, Dunod: Paris, 2020, p. 1231-1245. 2100712365 (Accepté/ Sous presse). http://hdl.handle.net/2078.1/231152

Friedel, Marie ; Renard, Marleen ; Devlieger, Hugo ; Verellen, Gaston. Belgian End-Of-Life Care Study Did Not Include French-speaking Units. In: Acta Paediatrica, Vol. 109, no.3, p. 636 (2020). doi:10.1111/apa.15082. http://hdl.handle.net/2078.1/231143

Friedel, Marie. Un dispositif pédagogique out of the box pour encourager la collaboration interdisciplinaire/interprofessionnelle? In: Les Politiques Sociales, Vol. 1, no.1&2, p. 128 p. (2020). http://hdl.handle.net/2078.1/231157 2019

Susam, Volkan ; Friedel, Marie ; Basile, Patrizia ; Bonetti, Loris. Efficacy of the Buzzy System for pain relief during venipuncture in children: a randomized controlled trial. In: Acta Biomed for Health Professions, Vol. 89, no. S.6, p. 6-16 (2018). doi:10.23750/abm.v89i6-S.7378. http://hdl.handle.net/2078/209247

Vandenbroeck, Kris ; Schmitz, Olivier ; Aujoulat, Isabelle ; Mistiaen, Patrick ; Friedel, Marie ; Genet,Magali ; Ricour, Céline ; Kohn, Laurence ; Wens, Johan. Appropriate care at the end of life. Report number 296 (), 2017. 211 p. p. http://hdl.handle.net/2078.1/231176

Friedel, Marie ; De Terwangne, Brigitte ; Brichard, Bénédicte ; Ruysseveldt,Ilse ; Renard, Marleen. The Belgian euthanasia law and its impact on the practises of Belgian paediatric palliative care teams. In: International Journal of Palliative Nursing, Vol. 24, no.7, p. 333-337 (2018). doi:10.12968/ijpn.2018.24.7.333. http://hdl.handle.net/2078.1/207384 2017

Friedel, Marie. Des soins palliatifs intégrés. In: Médecine Palliative, Vol. 16, no.3, p. 169-172 (2017). doi:doi.org/10.1016/j.medpal.2017.01.004. http://hdl.handle.net/2078.1/231709

Friedel, Marie. D'un passage à l'autre, analogies entre l'accouchement et la mort. In: Soins Pédiatrie - Puériculture : la revue de tous les acteurs du soin à l'enfant, Vol. 37, no.289, p. 37-41 (2016). doi:10.1016/j.spp.2016.01.010. http://hdl.handle.net/2078.1/207389

Friedel, Marie. L'art de faire dans de la dentelle. Des soins palliatifs en périnatalogie. In: Médecine Palliative, Vol. 15, no.16, p. 307-308 (2016). doi:doi.org/10.1016/j.medpal.2016.05.001. http://hdl.handle.net/2078.1/231710

Clément de Cléty, Stéphan ; Friedel, Marie ; Verhagen, A A Eduard ; Lantos, John D ; Carter, Brian S. Please Do Whatever It Takes to End Our Daughter's Suffering! In: Paediatrics, Vol. 137, no.1, p. e20153812 [1-8] (2015). doi:10.1542/peds.2015-3812. http://hdl.handle.net/2078.1/176171 2014

Friedel, Marie. Does the Belgian law legalising euthanasia for minors really address the needs of life-limited children? {commentary}. In: International Journal of Palliative Nursing, Vol. 20, no.6, p. 265-268 (2014). http://hdl.handle.net/2078/209241

Friedel, Marie. Soins palliatifs en pédiatrie, éthique et relation au patient. In: Soins Pédiatrie - Puériculture, Vol. 35, no. 281, p. 42-46 (2014). doi:10.1016/j.spp.2014.09.008. http://hdl.handle.net/2078.1/160403

Friedel, Marie. Symposium sur la démarche palliative intégrée (Bruxelles, septembre 2016). Symposium in INtegrated Palliative Care (Brussels, 01/09/2016). In: Médecine Palliative : Soins de Support - Accompagnement - Éthique, Vol. 16, no.3, p. 169-172 (2017). doi:10.1016/j.medpal.2017.01.004. http://hdl.handle.net/2078.1/209240 2016

Conference papers and posters not directly linked to the thesis but to palliative care

Friedel, Marie. La complexité des processus décisionnels partagés en soins palliatifs: l'utilité de s'appuyer sur des guides de bonnes pratiques. Soins palliatifs: la vie en transition (Namur, 03/03/2020). http://hdl.handle.net/2078.1/231173 2019

Friedel, Marie. Comment aborder les adolescents gravement maladies à domicile? Des pistes pour oser la rencontre! Annual Conference of the Association Ecole à Domicile, Brussels, Belgium (Bruxelles, 17/05/2018). http://hdl.handle.net/2078.1/231714

Friedel, Marie ; Mendes, Joana. Creating a safe environment for children and parents : how the concept of holding applied to paediatric palliative care might collid with the Belgian child euthanasia procedure ? 4th Maruzza Congress on Paediatric Palliative Care: A global gathering (Rome, Italy, du 25/10/2018 au 27/10/2018). http://hdl.handle.net/2078.1/231712

Van den Broek, Kris ; Schmitz, Olivier ; Friedel, Marie ; Aujoulat, Isabelle ; Wens, Johan. Defining appropriate end of-life care: a Belgian community study. 10th World Research Congress of the European Association of Palliative Care (Bern, Switzerland, du 24/05/2018 au 26/05/2018). http://hdl.handle.net/2078.1/231726

Friedel, Marie ; Schmitz,Olivier ; Van den Broek, Kris ; Wens, Johan ; Aujoulat, Isabelle. Definition of (in)appropriate care seen from the patient's perspective. 10th World Research Congress of the European Association of Palliative Care (Bern, Switzerland, du 24/05/2018 au 26/05/2018). http://hdl.handle.net/2078.1/231724

Van den Broek, Kris ; Schmitz, Olivier ; Friedel, Marie ; Aujoulat, Isabelle ; Wens, Johan. Opinions and attitudes about end of life care: A Belgian community study. 10th World Research Congress of the European Association of Palliative Care (Bern, Switzerland, du 24/05/2018 au 26/05/2018). http://hdl.handle.net/2078.1/231725 2017

Friedel, Marie ; Jacqmin, Nadine. Le leadership infirmier dans l'implémentation des bonnes pratiques. CONGRES ANNUEL acn/FNIB pour la journée internationale des infirmiers et infirmières: le leadership infirmier (CEME, Charleroi, Belgique, 20/09/2018). http://hdl.handle.net/2078.1/231713

Mendes,Joana ; Friedel, Marie. Non-religious arguments to discuss paediatric euthanasia : why should paediatric palliative care use them. 4th Maruzza Congress on Paediatric Palliative Care: A global gathering (Rome, Italy, du 25/10/2018 au 26/10/2018). http://hdl.handle.net/2078.1/231711

Dancot,Jacinthe ; Friedel, Marie ; Savignac, Paule. La prise de décision éthique : Concepts et exemple d'application clinique. 14th European Conference in Advanced Practice Nursing (MICX, Mons, Belgium, 16/11/2017). http://hdl.handle.net/2078.1/231715

Friedel, Marie. Il tempo dell'attesa. L'infermiere di fronte alle scelte di fine vita. XXII Congresso nazionale delle Societa Italiana di Neonatologia (Napoli, Italy, 15/10/2016). http://hdl.handle.net/2078.1/231718

Friedel, Marie. D'un passage à l'autre. Quelles analogies entre l'accouchement et la mort? 21th Conference of the Société Française d'Accompagnement et de Soins Palliatifs (SFAP) (Nantes, France, du 24/06/2015 au 27/06/2015). In: Soins Pédiatrie - Puériculture : la revue de tous les acteurs du soin à l'enfant, Vol. 37, no.289, p. 37-41 (2015). doi:10.1016/j.spp.2016.01.010. http://hdl.handle.net/2078.1/231720

Friedel, Marie ; Clément de Cléty, Stéphan ; Jacquemin, Dominique. Self-request euthanasia in minors: an act of compassion. Analyzing arguments during public debate in Belgium. 7th Cardiff Conference : Medicine and compassion. Tool for the task or dangerous distraction? (with ICPCN) (Cardiff university, United Kingdom, du 08/07/2015 au 10/07/2015). In: International Journal of Palliative Nursing, Vol. 20, no.6, p. 121-124 (2015). doi:10.12968/ijpn.2014.20.6.265. http://hdl.handle.net/2078.1/231719 2014

Friedel, Marie ; Tonon, Corinne ; D'Oultremont, Elisabeth ; Develter, Sonja. Comment les concepts de créativité et de spiritualité peuvent-ils éclairer des situations cliniques en soins palliatifs pédiatriques? 6ème Congrès du Réseau francophone de soins palliatifs pédiatrique: Savoirs et incertitudes (St Malo, France, du 01/10/2014 au 03/10/2014). http://hdl.handle.net/2078.1/231722

Friedel, Marie. How does the Belgian law about child euthanasia impact a Belgian paediatric palliative care course? 2nd Maruzza Conference: Paediatric palliative care : a global gathering. (Roma, Italy, du 19/11/2014 au 21/11/2014). http://hdl.handle.net/2078.1/231721

Friedel, Marie. What does the assessment of a paediatric palliative course tells us? 1st Conference of the International Children’s Palliative Care Network (ICPCN): From ideas to action! (Mumbai, India, du 09/02/2014 au 11/02/2014). http://hdl.handle.net/2078.1/231723

Photo credits @ Marie Friedel

PART I INTRODUCTION AND METHODS

CHAPTER 1. INTRODUCTION

I would like to introduce the theme of quality of life by illustrating it with a real situation encountered in paediatric palliative care as a former paediatric liaison nurse.

"L. is 17 years old. He suffered multiple degenerative diseases since his infancy and has had two types of cancers. His respiratory function is so weak that he is on permanent oxygen support. He is almost bed-ridden. Sometimes, when he doesn't feel too tired, he sits in a wheel chair and follows school lessons from his computer. Being a very smart young man, he has succeeded in achieving his secondary diploma, which he absolutely wanted to receive in person in school and not by post. He has an older brother and a close relationship with his father. His mother, a teacher, left her profession to dedicate herself to L's care. Each year, L. is admitted to hospital to treat infections or manage pain or digestive symptoms. As he faces sleep problems, he likes the relaxation sessions offered by a paediatric liaison nurse, who regularly visits him at home and coordinates the interdisciplinary care between hospital and home. Whenever he has visitors, he enjoys offering each of them a elaborate home-made scoubidou representing animals, ships or airplanes. Once, he gifted me a CD on which he had recorded five versions of the song called 'Little Boxes'. He explained that, unlike in the song, he felt he was being cared for as a whole person and not segmented into his component organs."

This short description of a situation gives an insight of what quality of life can represent for an adolescent facing a life-limiting condition. The situation also highlights the complex and subjective dimension of quality of life, which is why it is such a challenge to evaluate it through an outcomes measurement instrument.

Palliative care as a public health issue

Palliative care is the holistic management of physical, psychological, legal and spiritual problems faced by patients with life-threatening illness and by their families. It aims to improve the quality of life of patients, their families and their caregivers by addressing their physical, psychosocial and spiritual needs through the prevention and relief of suffering. This is achieved through early identification and impeccable assessment and treatment of pain and other symptoms (1).

The Lancet commission on Palliative Care and Pain relief study group affirms that "alleviation of the burden of pain, suffering and severe distress associated with life-threatening and life-limiting health conditions and with end of life is a global health and equity imperative."(2,3).

The Astana Declaration affirms that palliative care is part of primary health care (4) following the World Assembly of Health, which called to "develop, strengthen and implement, where appropriate, palliative care policies to support the comprehensive strengthening of health systems to integrate evidence-based, cost-effective and equitable palliative care services in the continuum of care, across all levels, with emphasis on primary care" (5). Palliative care is explicitly recognised under the human right to health. It should be provided through person-centred and integrated health services that pay special attention to the specific needs and preferences of individuals (4). Although palliative care is not a basic human right, it is a component of the right to health, and access to palliative care services should be ensured (6).

Integration of palliative care into existing health care systems can be achieved, according to the WHO, through its implementation in universal health coverage, financing it by public funds and integrating it into national insurance and social security programs. Furthermore, palliative care must be anchored in clinical guidelines and referral systems at all levels of care. It also refers to the monitoring and evaluation of legislative provisions, policies, interventions and programs. "Progress on health and on human rights can be monitored with explicit outcomes scales and benchmarks, using an appropriate set of metrics that extend beyond mortality and morbidity. " (2)

Whilst palliative care for adults is progressively integrated in a European country like Belgium, palliative care for children remains quite invisible. It distinguishes itself from adult palliative care for several reasons: “A smaller number of children die each year compared to adults.

Many rare childhood conditions will never be encountered in the adult population.

Multiple family members can be affected by the same genetic condition.

The time span of a childhood condition can extend over a number of years, which may require the patient to transition from child-centred to adult palliative care services at some stage.

Children’s palliative care embraces the whole family. Family members, especially parents and siblings, will be vulnerable as they face the changes in life that the child’s diagnosis creates, and as they anticipate bereavement.

Children’s palliative care providers need to be aware of the continuing physical, emotional and cognitive development throughout childhood and respond to each child’s changing levels of communication and their ability to understand.

Provision of education and play when a child is seriously ill is essential. This introduces an additional dimension which adds to the complexity of care provision.” (7)

Prevalence and definitions

A cross-sectional study estimates that 21 million children worldwide are facing life-limiting or life-threatening conditions (8), where the prevalence of life-limiting conditions ranges from 10 to 32 in 10.000 children (0-19 years) (9-10). Approximately 170.000 children with life-limiting conditions die every year in Europe (11).

Life-threatening conditions (LTCs) are illnesses or conditions that pose a grave threat of mortality to children or young adults, and for which medical treatment may result in a cure, but may fail. Life-limiting conditions (LLCs) are illnesses or conditions for which there is no cure, and which are extremely likely to result in death at some point in time during childhood or young adulthood. Most LTCs and LLCs cause substantial impairments or progressive deterioration, resulting in increasing dependence on parents and other caregivers (12,13).

The Association for Children’s Palliative Care (ACT) and the Royal College of Paediatrics and Child Health (RCPCH) classified LTCs and LLCs into four categories in 1997 (14). These are:

1.Life-threatening conditions for which curative treatment may be feasible but can fail. Access to palliative care services may be necessary when treatment fails or during an acute crisis, irrespective of the duration of threat to life. On reaching long-term remission or following successful curative treatment there is no longer a need for palliative care services.

Examples: cancer, irreversible organ failures of heart, liver, kidney.

2. Conditions where premature death is inevitable. There may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities.

Examples: cystic fibrosis, duchenne muscular dystrophy.

3. Progressive conditions without curative treatment options. Treatment is exclusively palliative and may commonly extend over many years.

Examples: batten disease, mucopolysaccharidoses.

4. Irreversible but non-progressive conditions causing severe disability, leading to susceptibility to health. Children can have complex health care needs, a high risk of an unpredictable life-threatening event or episode, health complications and an increased likelihood of premature death.

Examples: severe cerebral palsy, multiple disabilities, such as following brain or spinal cord injury.

More than 400 different diseases were identified and correlated to the ICD-10 (International Classification of Diseases) by Hain & Devins in 2013 (15).

Life-limiting conditions are often correlated with intense physical and psychosocial suffering, especially at end-of-life (16,17). Life-limiting or life-threatening conditions affecting children also place a serious long-term burden on all family members, including psychological and physical suffering such as exhaustion, fatigue, depression, post-traumatic stress disorders, in addition to socio-economic worries (18-19).

To alleviate this suffering, paediatric palliative care has emerged since 1970 as a subspecialty of paediatrics and palliative care (20). In 1978, M. Bluebond-Langner published her book entitled The private worlds of dying children (21). The first children’s hospice was opened in 1983 (Helen House in Oxford, United Kingdom) and the first specialist paediatrician in children’s palliative care was appointed in 1990 in the United Kingdom. In 1998, the World Health Organization defined paediatric palliative care as “ the active total care of the child's body, mind and spirit, and also involves giving support to the family. It begins when illness is diagnosed and continues regardless of whether or not a child receives treatment directed at the disease. Health providers must evaluate and alleviate a child's physical, psychological, and social distress. Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources; it can be successfully implemented even if resources are limited. It can be provided in tertiary care facilities, in community health centres and even in children's homes.” (1)

Paediatric palliative care is patient-centred and family-focused. It seeks to inform and support medical decision- making by patients and families, to provide respite for carers and to support family members in bereavement (12,22).

Standards for paediatric palliative care in Europe have been formulated in a document called IMPaCCT, an acronym that stands for International Meeting for Palliative care in Children, Trento. It was in this town in Italy that several health care professionals from Europe, Lebanon, Canada and the USA met in 2006 to define paediatric palliative care, referral criteria and quality indicators. This document led to the creation of a paediatric taskforce at the European Association of Palliative Care (12). In 2014, the Trieste Charter was elaborated by 50 experts in paediatric palliative care, proposing ten fundamental rights for children who are approaching the end of their lives. Each right has a series of duties, and the charter recommends the most appropriate manner in which to fulfil them (23). The charter contributes to advocacy for paediatric palliative care by describing how to respond to children's needs at the end of life.

The Trieste Charter can be found in the Appendix.

Impact of paediatric palliative care

Studies suggest that specific paediatric palliative care programs would improve children's quality of life by better symptom control and offering meaningful "fun" activities in children's daily lives (24-27) and that, in turn, healthcare costs would be reduced by lowering the number of emergency admissions, the number of hospitalizations and their lengths and the number of deaths in paediatric intensive care units (25,28,29).

But how are these outcomes achieved? Which are the underpinning mechanisms? A realistic review addressed those questions and suggested that the following elements may promote quality of life: when the child and the family feel respected, heard and supported, when the emphasis of care is placed on lessening suffering, emotions are shared, and the possibility of death is acknowledged (30). Those results consider the quality of relationships between the family and the healthcare professionals to be an essential component of quality paediatric palliative care.

Quality paediatric palliative care is achieved by addressing meaningful outcomes. Those outcomes should be defined by patients themselves, in our case by children and their parents. From a clinical perspective, the main objective of paediatric palliative care is to promote quality of life. Understanding what influences children's and parent's quality of life and what matters most to them is a fundamental step towards clearly defining the dimensions that might be evaluated.

What is quality of life in a paediatric palliative care context?

Paediatric palliative care aims to promote quality of life, but currently little is known on the conceptual definition of this construct for children and their parents in a palliative context.

The World Health Organization defines quality of life (QoL) as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns. It is a broad ranging concept affected in a complex way by the person's physical health, psychological state, personal beliefs, social relationships and their relationship to salient features of their environment. This definition reflects the view that quality of life refers to a subjective evaluation, which is embedded in a cultural, social and environmental context. As such, quality of life cannot be simply equated with the terms "health status", "lifestyle", "life satisfaction", "mental state", or "well-being". Rather, it is a multidimensional concept incorporating the individual's perception of these and other aspects of life." (31)

One of the implications of the WHO's focus on the subjective evaluation of QoL means that a distinction can be made between individualized and standardized measures of quality of life. The attributes definition of individual quality of life are based on Calman's theory (32). According to this theory, the gap between aspirations and real life determines the individual's perception of quality of life. Individual quality of life does not correspond to a standard but to a reference determined by the subject himself, which leads him to evaluate the difference between his expectations and the perceived reality. In contrast, standardized measures evaluate quality of life based on predetermined domains.

A systematic review conducted by Namisango et al. looked at the meaningful domains of paediatric palliative care, paving the way to identifying what influences children's quality of life (33). She found that the following five domains reflected the priority concerns: physical (e.g. symptoms), psychological (e.g. worries), psychosocial (e.g. relationships), existential (e.g. existential loss) and others (e.g. information access). Moreover, she noticed that children's perspectives were not systematically researched. This strengthens the need to develop outcome measurement instruments which systematically include the possibility for the child to self-report their quality of life.

In line with those results, a meta-summary suggested "eight themes which might be influencing children's quality of life in a palliative care context : the relationship with professional caregivers, pain and its management, "living beyond pain," the relationship between paediatric patients and their families, children's view on their treatment and service provision, meanings children give to their end-of-life situation, consequences of clinical decisions, and the relationships among children in paediatric palliative care and their peers." (34)

We notice, again, the importance of quality relations between children and their caregivers but also with their peers, as an important domain to include in an outcome measurement instrument.

Throughout this dissertation, based on our literature review, previous work and clinical expertise as a specialist nurse working in paediatric palliative care and international networks in the field of paediatric palliative care, we would like to propose the following definition of quality of life:

In a nutshell, quality of life is what makes the spice of life. It is what boosts a person to get up in the morning. It is what makes her happy, feel secure and connected to others.

It is composed of all the dimensions which contribute, impact and/or reflect the spice of life for one person, at one moment of their life. Those dimensions are not restricted to disease-related symptoms, but encompass the whole unique person -with their feelings, experiences and hopes - in a holistic, complex and dynamic family-oriented approach.

We assume that, despite a life-limiting condition and the impact it has on the life of the child and its whole family, quality of life may still occur and, more importantly, may be improved by paediatric palliative care.

This paradox, balancing between positive and challenging aspects when dealing with a life-limiting condition, is synthesised by a quote from Denis Vasse (35), French psychoanalyst: «Je veux vivre, mais je vais mourir.» (*I want to live but I will die*). Contrasts such as life and death, day and night, light and shadows, should also be reflected in the items included in quality of life measures. Avoiding the pitfalls of both over-dramatizing and over-positivizing, but instead reaching carefully into the reality and perceptions of children.

Evaluating quality of life in paediatric palliative care

Defining what quality of life is for children living with life-limiting conditions is still partially unknown and, consequently, so is how to evaluate it. This reflects the urgent need to switch from a rather empirical intuitive-guided field to an evidence-based corpus, in order to objectively document needs and outcomes, to increase the quality of care provided to families and furthermore to foster its credibility, which might also contribute to long-term funding of paediatric palliative care services (36-38).

We recognize that one standardized instrument called the 23-item Paediatric Quality of Life Inventory (PedsQL.4.0) has been widely used and published (39-40). The PedsQL Measurement Model is a modular approach to measure health-related quality of life (HRQOL) in healthy children and adolescents and those with acute and chronic health conditions from 2 to 18 years. However, some authors argue that the PedsQL 4.0 would be unreliable for measuring HRQOL within this population. In particular, convergent/discriminant validity and construct validity were found unsatisfactory in this population. Furthermore, parents reported that the content of several items was not applicable to their children, particularly related to physical and school functioning (41-42). Restrictively assessing the child’s quality of life through the evaluation of all his declining functions during palliative care is critizised. Those facts suggest that the PedsQL 4.0 is not suitable for measuring QoL among children with life-limiting conditions.

This is in line with a systematic review, which found that there is currently “no 'ideal' outcome assessment measure for use in paediatric palliative care. The domains of generic health-related quality-of-life measures are not relevant to all children receiving palliative care and some domains within disease-specific measures are only relevant for that specific population.” (43). Huang et al. (41) and Coombes et al. (43) have suggested the use of individual QoL measures. One of these measures is the Scheduled evaluation of individual quality of life-direct weighted (SEIQoL-Dw) (44), which assess the weight of individual domains, important for each child ‘s quality of life.

To our knowledge, the SEIQoL-Dw is the only instrument which permits the child to elicit a self-generated list of the five most important domains of QoL for him, rather than responding to pre-set areas, which all carry the same weight. After having identified their most important domains, children are requested to rate how well (satisfied) each domain is for them currently and, lastly, to rate how important each domain is for them. A final SEIQoL score reflects the overall QoL. It was successfully tested in a small sample of 45 patients aged 7-16 years with acute leukaemia, in parallel with the specific PedsQL 3.0 cancer module. ‘Relationships with families’ was elicited as the most important domain for children aged 7-11 years and quality of life was found to be higher when children perceived their relationship with their family as good (45). The SEIQoL was also used in a population of children with diabetes (46) and with cerebral palsy (47). A similar tool based on the SEIQoL was developed and is called Child Quality of Life Systemic Inventory for children (QLSI-C). It is a generic self-assessment questionnaire suitable for children aged 8-12 years (48). A further study assessed the psychometric properties of a technology-based QLSI-C among 80 children aged 8-12 years comparing it to the paper-based version. Findings established the reliability of the tablet-administered QLSI-C scores (49).

In recent years, patient-centred outcome measures (PCOM), which encompass both patient-reported and proxy-reported measures, have mainly been developed. Their general aim is to improve of clinical teams’ awareness of their patients’ unmet needs. A systematic review focusing on adults in palliative care shows “a strong evidence for an impact of PCOMs feedback on process of care including better symptom recognition, more discussion of quality of life and increased referrals based on PCOMs reporting” (50). Currently, we do not know if this mechanism occurs for children. It has, however, been recommended in a paediatric family-centred approach to integrate self- and proxy-report in the same PCOM. Because of discrepancies between self- and proxy-report (51-54), many studies highlight the importance of acknowledging both parent and child perspectives to better assess outcomes in a paediatric palliative population (55-57).

Quality of life is the main goal of paediatric palliative care. But how can we evaluate whether this goal has been reached? What can quality of life be an outcome of?

The concept of outcome can be understood in two ways.

The first definition of an outcome is a 'change in health status' as a consequence of health care or interventions (58). The term health status should be understood from the broad perspective of well-being or quality of life. If we refer to Donabedian's model for evaluating quality of care, we must look at the structure, the process and the outcomes of a care program. High quality paediatric palliative care is presumed to improve quality of life. Quality of life, however, is a multidimensional construct and most probably not only influenced by Paediatric Palliative Care (PPC) interventions.

The second definition of an outcome can be related to patient-reported outcomes (PROMs), which are not directly the effect of interventions, as cited by Witt et al. : "It is important to recognise that experience of care is not the same as outcomes of care. Experiences are likely to be better if outcomes are better, but they relate more closely to how individuals are respected, listened to and heard. "(59)

In our research, we will focus only on patient-reported outcomes and on adapting an outcomes measurement instrument. Therefore, we cannot expect to be able to measure quality of care, as we didn't evaluate the structure and process of care in this research.

Several conceptual, social, legal, ethical and methodological barriers make it challenging to develop a short, feasible, valid and reliable outcome measurement instrument able to evaluate quality of life for children in a palliative context and in a family-centred approach (41,57,60,61).

Despite the methodological challenges to developing an outcomes measurement instrument, a promising tool is the APCA children's palliative outcome scale (APCA CPOS), conceived by the African Palliative Care Association (APCA) (61,62). Based on the APCA palliative outcome scale for adults (63), it combines a self- and proxy-report. The original APCA CPOS was published during this thesis (61). The CPOS is currently being tested in the UK, in Turkey and in Uganda by Pr. R. Harding's team at the King's College London Cicely saunders Institute in the United Kingdom. The CPOS is a multidimensional patient-centred outcome measure, with 5-point Likert-scale response options. It contains 12 items exploring physical and psychosocial elements. Seven items are related to children's quality of life, which can be rated by the children themselves (self-report) and rated by their parents (proxy-report), and five items focus on the parents' quality of life. The CPOS includes items focusing on physical and psychological dimensions of quality of life.

For several reasons, we decided to further build on the CPOS, despite it having some limitations. Its psychometric properties were not yet assessed nor described and it was tested in an African context of care, among a population of children who suffered mainly from AIDS.

The advantages of the CPOS, however, were multiple. It was short, meaning it would not put an additional burden on children, parents and healthcare professionals if used in a routine clinical care setting. Secondly, it was elaborated for and by a paediatric palliative care population, maximising its potential face and content validity. Third, it included within the same scale a child self-report and a parent proxy-report - as recommended by standards- in order to address children's lack of cognitive/verbal capacity to self-report, and fourthly it evaluates children and parents' quality of life in a family-centred approach.

We decided to refer to the COSMIN guidance, because it is the most recent and the most adapted to clinimetrics, in other words, to outcomes measurement instruments used for clinical teams in medicine. Recommendations and guidance from the COSMIN study group (64) include steps that must be respected when adapting an outcome measurement instrument. These steps are briefly listed hereafter and will be further developed in chapter 2 on methods: Definition of the construct, the population for which the instrument is developed, the goal of measurement, the type of instrument (single or multidimensional), the items based on literature and patient's experience and the scoring options. The iterative pilot-testing and subsequent field-testing of the outcome measurement instrument used to validate or adapt the instrument are inherent to the process.

Developing an outcome measures instrument that involves collecting data among children, especially when they are considered vulnerable because of a life-limiting condition, requires the consideration of ethical principles of paediatric palliative care research.

Ethics in paediatric palliative care research

Paediatric palliative care aims to improve the quality of life (QoL) of children with life limiting conditions and the QoL of their parents (1,12). Researching children's QoL in paediatric palliative care has been acknowledged as an important issue (38), but includes legal, clinical, ethical, psychological and methodological challenges (65-68).

Methodological challenges are, for example, the fact that a heterogeneous population of children in age, diseases and care pathways are found in PPC. Often those children are non-verbal because of their age or the neurological disease they are facing, explaining why few studies report children's perspectives on QoL in a palliative context (34,69,70). Other challenges are linked to small sample sizes, problems with research ethics committees, the unpredictable nature of diseases, and gatekeeping and social perceptions on the potential burden for children and their families if they are involved in research (38,65,69,71,62).

How can the quality of life of children who are facing life-limiting conditions be explored in a respectful way? In other terms, how can we involve them, finding the right balance between our duty to protect them (in accordance with the do not harm ethics principle) and our desire to give them a voice? The following paragraphs will present principles in clinical ethics, in research ethics, and in palliative care research before focusing on recommendations to follow when involving children in palliative care research.

Clinical ethics can be summarized as follows: how should we act to do things right? One can think that what is right is what is good. However, defining what is good relies on a moral point of view. Morals include principles, values and norms, which can vary extensively according to cultures, religions and states. Another interpretation of what is good can be drawn from a legal perspective, on which laws, rules and procedures are elaborated. Alongside of these points of view stand ethics. Ethics is the search for the good, or the « least bad », in a specific situation, through reflection and discussion with others (73).

According to Hain (74) three questions should be considered when making clinical decisions. These are: What are the intentions behind an intervention? What are the interventions? What will be the presumed consequences? All 3 questions should be addressed if one aims to make ethical decisions.

Those considerations refer directly to **research ethics**, which is defined as the analysis of ethical issues that may occur when people are involved as research participants. The main goal of research ethics is to protect human participants, especially if they are vulnerable, to ensure that research is conducted in a way that serves the interests of individuals, groups and/or society and to scrutinize research activities for their ethical soundness, looking at the management of risk, the protection of confidentiality and the process of informed consent/assent.

Research ethics were developed after world war two, during which testing on human beings was conducted without any attention to fundamental human rights. Respecting the principles of research ethics was affirmed at an international level, leading to essential documents, such as the Nuremberg Code in 1946-1947 (75) , the Declaration of Helsinki in 1964, revised in 1975 (76), the Charter of Fundamental Rights of the European Union in 2000 (77), the European Convention on Human Rights in 2010 (78) and the Council for International Organizations of Medical Sciences in 2016 (79). In Belgium, principles of research ethics are found in three laws: The Law on Privacy in 1992 (80), the Law on Patients' Rights in 2002 (81) and finally the Law on Patients' Consent and Protection in 2004 (82).

Focusing now on **palliative care research**, respecting the common ethical principles (beneficence, non-maleficence, autonomy and justice) towards vulnerable patients is even more crucial (83,84). In two studies, results showed that patients accepted to participate in research because they had the desire to help other patients, to help themselves and to help researchers while contributing to scientific knowledge. Participation in research had meaning and was considered by them as a positive experience, offering an opportunity to feel engaged, validated and to express gratitude (85-86). Those findings are in contrast with the representations of clinicians and/or researchers for whom researching palliative patients' perspectives would be considered harmful. Best practices for palliative care research in adults were published in 2013 (87-88), including recommendations for ethical issues in palliative and end of life care research. This document, the Methods Of Researching End of Life Care (MORE Care) research methods guidance, promotes patient's involvement in research, advocates for ethical committees and researchers to focus on potential benefits for patients considered as moral agents, while facilitating procedures, adequacy of information provided and type of vocabulary used for informed consent sheets.

Looking more specifically to **paediatric palliative care research**, the subtle balance between protecting children and giving them a voice has been discussed and documented (89-93). Several recommendations can be found. Following those recommendations, research must benefit children (child's best interest), the child-informed assent must be age-appropriate and researched without coercion, research must be the most inclusive possible (regardless of the disease or the social conditions) and be conducted with parental consent (loyalty). Moreover, data collection must be acceptable for children (non-maleficence), in a trusted and secured environment, integrating a certain degree of flexibility regarding data collection and acknowledging expectancy. Finally, the research should also be perceived by caregivers as feasible, and overall respect the fundamental triangulation of relationships between children, their parents and the professional carers (22,67,94-100).

One issue when involving children in research is obtaining a valid informed assent (101,102). Lambert & Glacken (103) highlight three key elements: assessing the child's capacity to understand and to assent, providing age-appropriate, comprehensible verbal and written information, ensuring voluntary participation by providing enough time to reflect and searching for subtle signs of refusal. Assent sheets should be colourful, including images and cartoons, explaining with comprehensible words the aim of the study and what is expected from the child during the study.

While few studies explored how children perceived their involvement in research, one study found that parents of children with life-limiting conditions accepted to be involved in research for altruistic reasons. Parents perceived their participation as a contribution made to science. Another reason was that parents found a personal benefit in expressing their feelings and sharing their experience with researchers. Authors concluded that parents had a positive experience of being involved in research when timing, approach and interview skills were appropriate (104).

In line with recommendations presented previously, we chose methods that would reduce the burden on children, on their parents and on the paediatric liaison team. These methods are explained in chapter 2.

Paediatric palliative care and regulations on end-of-life in Belgium

Paediatric palliative care was first introduced in Belgium in the 1980s, in Ghent (105) and in Leuven (106), emerging from paediatric oncology units that wanted to create an extension of ambulatory services by offering curative care to children. As a result, families with a child whose illness was proving to be incurable repeatedly requested that the child be cared for at home until death. In January 2002, thanks to the non-profit organisation Eclaircie, an inter-university network of paediatric liaison nursing teams was set up with the aim of facilitating the return and home care of children suffering from serious chronic diseases or at the end of their lives. Five paediatric liaison teams were officially recognised by a Royal Decree in 2010 which sets the conditions that the teams must meet in order to be approved and funded (107). Their mission is to ensure 24/7 continuity of care among all care settings (hospital/ambulatory/home/institutions) for children with life-limiting or life-threatening conditions. Paediatric liaison teams, which must be attached to a university hospital, can provide either curative care in relation to a condition for which a curative treatment is possible, palliative care in connection with a condition for which there is no or no longer any curative treatment possible, or terminal care as soon as the condition allows only an accompaniment of the end of life. Those paediatric liaison teams must be interdisciplinary and include one paediatrician, paediatric nurses, one psychologist and one administrative member. All must have been trained in paediatric palliative care.

A Law on Palliative Care, not specific to children, was approved in Belgium in 2002 (108). In the same year, two other laws, one on patients' rights and one on self-requested Euthanasia for adults, were voted in (109). Fourteen years later, in 2016, the definition of palliative care was extended to all patients with serious conditions, and not only to patients at end-of-life (110).

Belgium has the particularity of being the only country in the world to extend, in 2014, its Law on euthanasia to competent minors who are experiencing unbearable physical suffering, without any age restriction (111, 112). This policy has been largely discussed in Belgium and at an international level, arguing legal, ethical, psychosocial and clinical concerns (113-116). The need for passing this bill was questioned by paediatric liaison teams, who reported having never faced, in 20 years of existence, a child's request for euthanasia (117)[1]. More importantly, paediatric palliative care affirms that personalized quality care might help children and families to live a full and meaningful life to the end, advocating for appropriate and sufficient means to achieve this goal rather than generalizing the possibility to end this life. At the same time, no published data was available on the number of or profiles of children facing life-limiting conditions in Belgium, nor on the quality of life they experienced or the access to paediatric palliative care services they had and/or the quality of care they received. Additionally, according to a recent study, paediatric palliative care education isn't mandatory in undergraduate curricula of nursing

---

[1] See full article in the Appendix (Articles and book chapters)

schools and medical faculties in Belgium (11). Furthermore, to our knowledge, no research focused on paediatric palliative care in Belgium was ever conducted by the beginning of this thesis in 2016.

Starting from these facts, we decided to research how paediatric palliative care is offered in Belgium and which children were referred to it. We also wanted to know how children's quality of life in light of life-limiting conditions could be explored and evaluated in a family-centred and holistic approach. We hypothesise that regularly evaluating children's quality of life in a simple and acceptable way might help clinical teams to assess children's and parents' needs, optimize the care provided and objectify its impact on a family's quality of life.

We called our research "MOSAIK", an acronym which stands for Move to Open Shared Advanced Interventions for Kids with life-limiting conditions. We wanted to highlight the overall dynamic, iterative and collaborative approach to advocating for children's voices by letting them self-report their quality of life. Working closely with the paediatric palliative care teams throughout the research, we hope that it will foster the implementation process of using patient-centred outcome measures among paediatric palliative care teams.

Objectives and structure of the thesis

This research pursues **five objectives**:

a) first, to document the characteristics of children followed-up by paediatric liaison care teams in Belgium and their access to those services;

b) second, to search for reliable instruments measuring outcomes in paediatric palliative care,

c) third, to further develop the CPOS able to evaluate children's quality of life and that of their parents by documenting its psychometric properties;

d) fourth, to report on the quality of life experienced by children followed-up by paediatric liaison care teams in Belgium;

e) and fifth, to better understand the paediatric liaison teams' experience of using an outcome measurement instrument.

The thesis includes the following chapters:

Chapter 2 presents the methods: the general study design, which is a mixed-methods convergent triangulation design, based on a collaborative approach with paediatric liaison teams. Additionally, we present a reflection on how ethical precautions in paediatric palliative care research were integrated into our study. It shall clarify the methodological choices that have been made in this study.

Chapters 3-6 present the results of 4 complementary studies:

Facts and figures on paediatric palliative care in Belgium (objective a) are presented in **chapter 3**. It includes a retrospective study based on annual reports of paediatric liaison teams (118), presented in subchapter 3.1;
and a second study looking at the access to paediatric liaison teams of children with chronic complex conditions from 2010 to 2014 in Brussels (119), presented in subchapter 3.2;

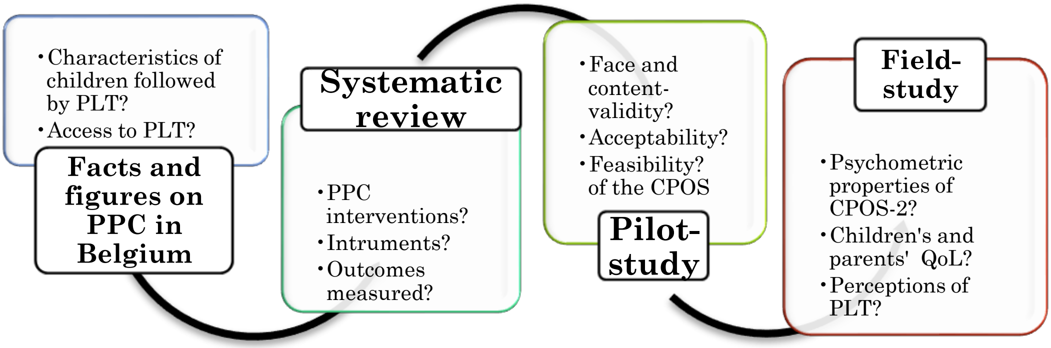
A systematic review (120) looking at the measured outcomes of paediatric palliative care interventions (objective b), presented in **chapter 4**;

A qualitative pilot-test (121) of the face/content validity, acceptability, feasibility of the further adapted CPOS-2 among children, parents and health carers (objective c) presented in **chapter 5**;

A national multicentric field-study of the CPOS-2 (objectives c, d and e), presented in **chapter 6**; including
the assessment of the reliability and concurrent validity of the CPOS-2 in subchapter 6.1;
an overview of children's and parents' quality of life, in subchapter 6.2;

and the perception of paediatric liaison teams when using the CPOS-2, in subchapter 6.3.

Figure 1. Overall design of the MOSAIK study



In **chapter 7**, we present the general discussion, highlighting the main findings and challenges faced, including the strengths and limitations of this research. Finally, perspectives, implications for practice and recommendations for further paediatric palliative care and research are suggested.

Chapter 8 presents the conclusion focusing on key messages.

The **appendix** includes the further adapted CPOS-2, and other outcome measurement instruments used in this study (KINDL, QOLLTI-F, SEIQoL); the Trieste Charter of the rights of the dying child and four additional papers/book chapters written by Marie Friedel during the thesis and linked to paediatric palliative care.

References

1. World Health Organization (WHO). Definition of palliative care. 1998. https://www.who.int/cancer/palliative/definition/en/ (consulted 15th June 2020).

2. Knaul FM, Farmer PE, Krakauer EL, De Lima L, Bhadelia A et al. on behalf of the Lancet Commission on Palliative Care and Pain Relief Study Group. Alleviating the access abyss in palliative care and pain relief—an imperative of universal health coverage: The Lancet Commission report. The Lancet, 2018;391(10128):1391-1454.

3. World Health Organization (WHO), Fact sheet N°402, Palliative Care, July 2015, http://www.who.int/mediacentre/factsheets/fs402/en/ (consulted 15th June 2020).

4. World Health Organization (WHO), Key facts, 19th February 2018. https://www.who.int/news-room/fact-sheets/detail/palliative-care (consulted 15th June 2020).

5. World Health Organization (WHO). Strengthening of palliative care as a component of comprehensive care throughout the life course. 67th World Health Assembly. 24th May 2014. https://apps.who.int/gb/ebwha/pdf_files/WHA67/A67_R19-en.pdf (consulted 15th June 2020).

6. Ezer T, Lohman D, de Luca GB. Palliative Care and Human Rights: A Decade of Evolution in Standards. J Pain Symptom Manage. 2018;55(2S):S163-S169. doi:10.1016/j.jpainsymman.2017.03.027.

7. International Childrens Palliative Care Network (ICPCN). http://www.icpcn.org/faq/ (consulted 14th July 2020).

8. Connor SR, Downing J, Marston J. Estimating the Global Need for Palliative Care for Children: A Cross-sectional Analysis. J Pain Symptom Manage. 2017;53(2):171-177. doi:10.1016/j.jpainsymman. 2016.08.020.

9. Cochrane H, Liyanage S, Nantambi R, Palliative Care Statistics for Children and Young Adults. Health and Care Partnerships Analysis. Department of Health, 2007. UK

10. Fraser et al. Rising National Prevalence of Life-Limiting Conditions in Children in England, Paediatrics 2012;129:1-7.

11. Arias-Casais N, Garralda E, Pons JJ, Marston J, Chambers L, Downing J, Ling J, Rhee JY, de Lima L, Centeno C, Mapping paediatric palliative care development in the WHO European region: Children living in low-middle income countries are less likely to access it, Journal of Pain and Symptom Management 2020. https://doi.org/10.1016/j.jpainsymman.2020.04.028.

12. Craig F, Abu-Saad Huijer H, Benini F, Kuttner L, Wood C, Feraris PC, Zernikow B. [IMPaCCT: standards of paediatric palliative care]. Schmerz. 2008;22(4):401-8. doi: 10.1007/s00482-008-0690-4.

13. Mc Namara K, Feudtner C. Chapter 1. History and epidemiology in Oxford Textbook of Palliative Care for Children. Goldman A, Hain R, Liben S (eds), 2012, p.4

14. ACT/RCPCH. A guide to the development of children’s palliative care services: report of the joint working party. Bristol, 1997.

15. Hain R, Devins M, Hastings R, Noyes J. Paediatric palliative care: development and pilot study of a 'Directory' of life-limiting conditions. BMC Palliat Care. 2013 Dec 11;12(1):43. doi: 10.1186/1472-684X-12-43. PMID: 24330676; PMCID: PMC4029745.

16. Wolfe J, Orellana L, Ullrich C, et al. Symptoms and Distress in Children With Advanced Cancer: Prospective Patient-Reported Outcomes From the PediQUEST Study. J Clin Oncol. 2015;33(17):1928-35.

17. Friedrichsdorf SJ, Bruera E. Delivering Paediatric Palliative Care: From Denial, Palliphobia, Pallilalia to Palliactive. Children (Basel). 2018;5(9). pii: E120. doi: 10.3390/children5090120.

18. Ljungman L, Boger M, Ander M, Ljótsson B, Cernvall M, vonEssen L. et al. Impressions That Last: Particularly Negative and Positive Experiences Reported by Parents Five Years after the End of a Child's Successful Cancer Treatment or Death. PLoSONE. 2016;11(6):e0157076. doi:10.1371/ journal.pone.0157076.

19. Ljungman L, Hovén E, Ljungman G, Cernvall M, von Essen L.Does time heal all wounds? A longitudinal study of the development of posttraumatic stress symptoms in parents of survivors of childhood cancer and bereaved parents. Psychooncology. 2015;24(12):1792-8. doi: 10.1002/pon.3856.

20. Sisk BA, Feudtner C, Bluebond-Langner M, Sourkes B, Hinds PS, Wolfe J. Response to Suffering of the Seriously Ill Child: A History of Palliative Care for Children. Paediatrics. 2020;145(1). pii: e20191741. doi: 10.1542/peds.2019-1741.

21. Bluebond-Langner M, The Private World of Dying Children. Princeton University Press, 1978.

22. Bluebond-Langner M, DeCicco A, Belasco J. Involving Children with Life-shortening Illnesses in Decisions about participation in clinical research: a proposal for shuttle diplomacy and negotiation, in Kodish E. (ed.) Ethics and Research with Children. A case-based approach. 2005.

23. Benini F, Vecchi R, Orzalesi M. A charter for the rights of the dying child. Lancet. 2014;383(9928):1547-8. doi: 10.1016/S0140-6736(14)60746-7.

24. Marcus KL, Santos G, Ciapponi A, Comandé D, Bilodeau M, Wolfe J, Dussel V. Impact of Specialized Paediatric Palliative Care: A Systematic Review. J Pain Symptom Manage. 2020;59(2):339-364.e10. doi: 10.1016/j.jpainsymman.2019.08.005.

25. Chong PH, De Castro Molina JA, Teo K, Tan WS. Paediatric palliative care improves patient outcomes and reduces healthcare costs: evaluation of a home-based program. BMC Palliat Care. 2018;17(1):11. doi: 10.1186/s12904-017-0267-z.

26. Vollenbroich R, Borasio GD, Duroux A, Grasser M, Brandstätter M, Führer M. Listening to Parents: The Role of Symptom Perception in Paediatric Palliative Home Care. Palliat Support Care. 2016;14(1):13-9. doi: 10.1017/S1478951515000462.

27. Friedrichsdorf SJ, Postier A, Dreyfus J, Osenga K, Sencer S, Wolfe J. Improved quality of life at end of life related to home-based palliative care in children with cancer. J Palliat Med. 2015;18(2):143-50. doi: 10.1089/jpm.2014.0285.

28. Taylor J, Booth A, Beresford B, Phillips B, Wright K, Fraser L. Specialist paediatric palliative care for children and young people with cancer: A mixed-methods systematic review. Palliat Med. 2020;34(6):731-775. doi: 10.1177/0269216320908490.

29. Postier A, Chrastek J, Nugent S, Osenga K, Friedrichsdorf SJ. Exposure to home-based paediatric palliative and hospice care and its impact on hospital and emergency care charges at a single institution. J Palliat Med. 2014;17(2):183-8. doi: 10.1089/jpm.2013.0287.

30. Mitchell S, Bennett K, Morris A, Slowther AM, Coad J, Dale J. Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review. Palliat Med 2020;34(3):387-402. doi: 10.1177/0269216319870647.

31. World Health Organization. Definition of quality of life. https://www.who.int/healthinfo/survey/whoqol-qualityoflife/en/ (consulted 15th July 2020)

32. Calman KC. Quality of Life in Cancer Patients-An Hypothesis. J Med Ethics. 1984;10(3):124-7. doi: 10.1136/jme.10.3.124.

33. Namisango E, Bristowe K, Allsop MJ, Murtagh FEM, Abas M, Higginson IJ, Downing J, Harding R. Symptoms and Concerns Among Children and Young People with Life-Limiting and Life-Threatening Conditions: A Systematic Review Highlighting Meaningful Health Outcomes. Patient. 2019;12(1):15-55. doi: 10.1007/s40271-018-0333-5.

34. Ghirotto L, Busani E, Salvati M, Di Marco V, Caldarelli V, Artioli G. Researching children's perspectives in paediatric palliative care: A systematic review and meta-summary of qualitative research. Palliative and Supportive Care. 2018;1-12. doi.org/10.1017/S1478951518000172.

35. Vasse D, Le poids du réel, la souffrance, Ed. du Seuil, 2008.

36. Fraser LK, Bluebond-Langner M, Ling J. Advances and Challenges in European Paediatric Palliative Care. Med Sci (Basel). 2020;8(2):20. doi:10.3390/medsci8020020

37. Bausewein C, Daveson B, Benalia H, Simon ST, Higginson IJ. Outcome Measurement in Palliative Care: The Essentials. 2011. PRISMA (Reflecting the Positive Diversities of European Priorities for Research and Measurement in End-of-Life Care).

38. Baker JN, Levine DR, Hinds PS, Weaver MS, Cunningham MJ, Johnson L, Anghelescu D, Mandrell B, Gibson DV, Jones B, Wolfe J, Feudtner C, Friebert S, Carter B, Kane JR. Research Priorities in Paediatric Palliative Care.J Pediatr. 2015;167(2):467-70.e3.

39. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: Reliability and validity of the Paediatric Quality of Life Inventory Version 4.0 Generic Core Scales in healthy and patient populations. Medical Care. 2001; 39, 800-812.

40.Varni JW, Limbers CA, Newman DA, Seid M. Longitudinal factorial invariance of the PedsQL™ 4.0 Generic Core Scales child self-report version: One year prospective evidence from the California State Children's Health Insurance Program (SCHIP). Quality of Life Research. 2008;17 :1153-1162.

41. Huang IC, Shenkman EA, Madden VL, Vadaparampil S, Quinn G, Knapp CA. Measuring quality of life in paediatric palliative care: challenges and potential solutions. Palliat Med. 2010;24(2):175-82. doi: 10.1177/0269216309352418.

42. Knapp C, Madden V: Conducting outcomes research in paediatric palliative care. Am J Hosp Palliat Care 2010;27(4): 277-281.

43. Coombes LH, Wiseman T, Lucas G, Sangha A, Murtagh FE. Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-949. doi:10.1177/0269216316649155

44. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33. doi: 10.1136/bmj.313.7048.29.

45. Scaon S, Chasseigne G, Giraudeau C. et al. Éléments de qualité de vie individuelle pour une amélioration de la prise en charge en psycho-oncologie pédiatrique. Psycho Oncologie. 2017;11, 69–75. https://doi.org/10.1007/s11839-017-0619-1

46. Wagner J. Acceptability of the Schedule for the Evaluation of Individual Quality of Life-Direct Weight (SEIQoL-DW) in youth with type 1 diabetes. Qual Life Res 2004;13(7): 1279-1285.

47. Vinson J, Shank L, Thomas PD et al: Self-generated Domains of Quality of Life in Children with and Without Cerebral Palsy. J Dev Phys Disabil 2010;22(5): 497-508.

48. Etienne AM, Dupuis G, Spitz E, Lemetayer E, Misotten P. The gap concept as a quality of life measure: validation study of the Child Quality of Life Systemic Inventory. Soc Ind Res. 2011; 100:241-257. doi: 10.1007/s11205-010-9614-7.

49. Touchèque M, Etienne A-M, Missotten, Dupuis G. A comparison of a tablet version of the Quality of Life Systemic Inventory for Children (QLSI-C) to the standard paper version. Psychological Assessment. 2016, 28(6), 780–785. https://doi.org/10.1037/pas0000234

50. Etkind SN, Daveson BA, Kwok W, Witt J, Bausewein C, Higginson IJ, Murtagh FE. Capture, transfer, and feedback of patient-centered outcomes data in palliative care populations: does it make a difference? A systematic review. J Pain Symptom Manage. 2015;49(3):611-24. doi: 10.1016/j.jpainsymman.2014.07.010.

51. Silva N, Crespo C, Carona C, Bullinger M, Canavarro MC. Why the (dis)agreement? Family context and child-parent perspectives on health-related quality of life and psychological problems in paediatric asthma. Child Care Health Dev. 2015;41(1):112-21.

52. Lim Y, Velozo C, Bendixen R. The level of agreement between child self-reports and parent proxy-reports of health-related quality of life in boys with Duchenne muscular dystrophy. Qual Life Res. 2014; 23(7): 1945–1952.

53. Sattoe J, van Staa A, Moll H. The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. Health Qual Life Outcomes. 2012; 10: 10.

54. Vetter TR, Bridgewater CL, McGwin G. An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? Health Qual Life Outcomes 2012; 10: 85.

55. Weaver MS, Darnall C, Bace S, Vail C, MacFadyen A, Wichman C. Trending Longitudinal Agreement between Parent and Child Perceptions of Quality of Life for Paediatric Palliative Care Patients. Children (Basel). 2017;4(8):65. doi:10.3390/children4080065.

56. Nelson KE, Feinstein JA, Gerhardt CA, Rosenberg AR, Widger K, Faerber JA, Feudtner C. Emerging Methodologies in Paediatric Palliative Care Research: Six Case Studies. Children (Basel). 2018;5(3):32. doi: 10.3390/children5030032.

57. Harding R, Chambers L, Bluebond-Langner M. Advancing the science of outcome measurement in paediatric palliative care. Int J Palliat Nurs. 2019;25(2):72-79. doi: 10.12968/ijpn.2019.25.2.72.

58. Donabedian A. Explorations in Quality Assessment and Monitoring: The Definition of Quality and Approaches to Its Assessment. Volume I. Ann Arbor, MI: Health Administration Press; 1980.

59. Witt J, Murtagh FEM, de Wolf-Linder S, Higginson IJ, Daveson BA. Introducing the Outcome Assessment and Complexity Collaborative (OACC) Suite of Measures. 2015 https://www.kcl.ac.uk/cicelysaunders/attachments/Studies-OACC-Brief-Introduction-Booklet.pdf (consulted 13th July 2020).

60. Huang IC, Revicki DA, Schwartz CE. Measuring paediatric patient-reported outcomes: good progress but a long way to go. Qual Life Res. 2014;23(3):747-50. doi: 10.1007/s11136-013-0607-2.

61. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02.

62. African Palliative Care Association. The APCA African Children's palliative outcome scale (APCA CPOS). Manual. 2012.

63. Powell RA, Downing J, Harding R, Mwangi-Powell F & Connor S on behalf of the APCA M&E Group. Development of the APCA African Palliative Outcome Scale. Journal of Pain and Symptom Management 2007, 33: 229-232.

64. De Vet HC, Terwee CB, Mokkink LB, Knol DL. Measurement in Medicine. A Practical Guide. Cambridge University Press. 2011.

65. Peake JN, Beecham E, Oostendorp LJ, et al. Research barriers in children and young people with life-limiting conditions: a survey. BMJ Supportive & Palliative Care 2018; 0:1-7.

66. Bluebond-Langner M, Beecham E, Candy B, Langner R, Jones L. Problems with preference and place of death for children too. BMJ. 2015;351:h6123. doi: 10.1136/bmj.h6123.

67. Rahimzadeh V, Bartlett G, Longo C et al. Promoting an ethic of engagement in paediatric palliative care research. BMC Palliative Care 2015; 14:50.

68. Abernethy A, Capell W, Aziz N, Ritchie C, Prince-Paul M, Bennett R, Kuttner J. Ethical conduct of Palliative Care Research: Enhancing Communication between investigators and Institutional Review Boards. Journal of Pain and Symptom Management 2014; 48,6:1211-1221.

69. Beecham E, Hudson BF, Oostendorp L, Candy B, Jones L, Vickerstaff V, Lakhanpaul M, Stone P, Chambers L, Hall D, Hall K, Ganeshamoorthy T, Comac M, Bluebond-Langner M. A call for increased paediatric palliative care research: Identifying barriers. Palliat Med. 2016;30(10):979-980. doi: 10.1177/0269216316648087.

70. Jalmsell L, Lövgren M, Kreicbergs U, Henter J-I, Frost B-M. Children with cancer share their views: tell the truth but leave room for hope. Acta Paediatrica 2016;105:1094-1099.

71. Hudson BF, Oostendorp LJ, Candy B, Vickerstaff V, Jones L, Lakhanpaul M, Bluebond-Langner M, Stone P. The under reporting of recruitment strategies in research with children with life-threatening illnesses: A systematic review. Palliat Med. 2017;31(5):419-436.

72. Crocker JC, Beecham E, Kelly P, Dinsdale AP, Hemsley J, Jones L, Bluebond-Langner M. Inviting parents to take part in paediatric palliative care research: a mixed-methods examination of selection bias. Palliat Med. 2015;29(3):231-40. doi: 10.1177/0269216314560803.

73. Beauchamp TL, Childress JF. Principles of Biomedical Ethics. 7td edition. Oxford University Press. 2013.

74. Hain RD. Ethics in Paediatric Palliative care. 1st Conference of International Children's Palliative Care Network, Mumbai. India. 13th February 2014.

75. The Nuremberg Code (1947) In: Mitscherlich A, Mielke F. Doctors of infamy: the story of the Nazi medical crimes. New York: Schuman, 1949: xxiii-xxv.

76. World Medical Association (WMA). Declaration of Helsinki. Ethical principles for medical research involving human subjects. https://www.wma.net/policies-post/wma-declaration-of-helsinki-ethical-principles-for-medical-research-involving-human-subjects/ (consulted 15 Jul 2020)

77. Charter of fundamental rights of the European Union. Official journal of the European Communities. 2000. https://www.europarl.europa.eu/charter/pdf/text_en.pdf (consulted 15 Jul 2020)

78. European convention on human rights. 2010. https://www.echr.coe.int/Documents/Convention_ENG.pdf (consulted 15 Jul 2020)

79. Council for international organizations of medical sciences. International ethical guidelines for health-related research involving humans. 2016. https://cioms.ch/publications/product/international-ethical-guidelines-for-health-related-research-involving-humans/

80. Ministère of Justice. Loi relative à la protection de la vie privée à l'égard des traitements de données à caractère personnel.
http://www.ejustice.just.fgov.be/cgi_loi/loi_a.pl?language=fr&caller=list&cn=1992120832&la=f&fromtab=loi&sql=dt=%27loi%27&tri=dd+as+rank (consulted 15 Jul 2020)

81. Ministry of Public Health in Belgium. Loi du 22 août 2002 relative aux droits du patient http://www.soinspalliatifs.be/images/pdf/loi_2002_08_22.pdf (consulted 15 Jul 2020)

82. Ministry of Justice. Loi du 7 mai 2004 relative aux expérimentations sur la personne humaine. http://www.ejustice.just.fgov.be/cgi_loi/loi_a1.pl?language=fr&la=F&cn=2004050732&table_name=loi&&caller=list&fromtab=loi&tri=dd+AS+RANK (consulted 15 Jul 2020)

83. Martins Pereira S, Hernandez-Marrero P. Research Ethics in Palliative Care: a hallmark in Palliative Medicine. Palliat Med 2019: 1-2.

84. Agar M, Ko D., Sheehan C, Chapman M, Currow D. Informed consent in palliative care Clinical trials: challenging but possible. Journal of Palliative Medicine 2013;16,5:485-491.

85. Bloomer M, Hutchinson A, Brooks L, Botti M. Dying persons' perspectives on, or experiences of, participating in research: an integrative review. Palliative Medicine 2018;32,4:851-860.

86. White C, Hardy J. What do palliative care patients and their relatives think about research in palliative care? A systematic review. Support Care Cancer 2010; 18: 905-911.

87. Higginson IJ, Evans CJ, Grande G, et al : MORECare. Evaluating complex interventions in end of life care: the MORECare statement on good practice generated by a synthesis of transparent expert consultations and systematic reviews. BMC Med 2013; 11: 111.

88. Gysels M, Evans CJ, Lewis P: MORECare research methods guidance development: Recommendations for ethical issues in palliative and end-of-life care research. Palliat Med 2013; 27;13: 908-917.

89. Rogers R, Labadie M, Pole K. Balancing voice and protection in literacy studies with young children. Journal of Early Childhood Literacy 2016; 16(1):34-59.

90. Unguru Y. Ethical challenges in early-Phase Paediatric Research for Life-Limiting Illness. Semin pediatr Neurol 2015; 22:177-186.

91. Center for children and young people, Southern Cross University, Childwatch international research network, Unicef, University of Otago. Recherche éthique impliquant des enfants. Ethical research involving children (ERIC), 2015.

92. Delfossi M-L., Parizea M.-H., Aman J-P. la recherche clinique avec les enfants: à la croisée de l'éthique et du droit . Belgique, France, Québec. Collection Bioéthique critique. 2009: 235-251.

93. Balen R, Blyth E, Calabretto H, Fraser C, Horrocks C, Manby M. Involving children in health and social research: 'Human becomings' or 'active beings'? Childhood 2006;13,1:29-48.

94. Moore TP, McArthur M, Noble-Carr D. More a marathon than a hurdle: towards children's informed consent in a study on safety. Qualitative Research 2018; 18(1):88-107.

95. Wolfe J, Siden H. Research in paediatric palliative care. Chapter 39: 441-457, in Goldman A, Hain R, Liben S (eds). Oxford Textbook of Palliative Care for Children, 2012.

96. Rapoport A. Addressing Ethical Concerns regarding Paediatric Palliative Care Research; Arch Pediatr Adolescent Med 2009; 163(8):688-691.

97. Ericsson S, Boyd S. Children's ongoing and relational negotiation of informed assent in child-researcher, child-child and child-parent interaction. Childhood 2017;24,3:300-315.

98. Schelbe L, Chanmugam A, Moses T et al. Youth participation in qualitative research: Challenges and possibilities. Qualitative Social work 2015; 14(4):504-521.

99. Mishna F, Antle BJ, Regehr C. Tapping the perspectives of Children Emerging ethical issues in qualitative research. Qualitative Social Work, sage Publications London, 2004.

100. Neill SJ. Research with children: a critical review of the guidelines. Journal of Child health care 2005;9,1:46-58.

101. Oulton K, Gibson F, Sell D et al. Assent for children's participation in research: why it matters and making it meaningful. Child: care, health and development 2016; 42(4):588-597.

102. Hadjiconstantinou M, Forbat L. Securing ethical permissions to conduct research with children. Nursing Children and Young People. 2012;24,1:22-25.

103. Lambert V, Glacken M. Engaging with children in research: Theoretical and practical implications of negotiating informed consent/assent. Nursing Ethics 2011;18(6):781-801.

104. Hynson JL, Aroni R., Bauld C., Sawyer SM. Research with bereaved parents: a question of how not why. Palliative Medicine 2006; 20,8:805-811.

105. Verlooy J, Mattheeuws K, De Clercq M J, De Porre J. Nearly twenty years experience of palliative and curative home-care for paediatric hematology/oncology patients. Pediatr Blood Cancer. 2010;55, 5:957.

106. Renard M, Van den Eynde G, Van Ruysseveldt I. Paediatric palliative care as a transmural liaison model : experience of 10 years of palliative care for all children. Oral Presentation at Maruzza Congress 2013. http://www.maruzza.org/new/wpcontent/uploads/2013/02/05-Renard.pdf

107. Royal Decree of 15th nov 2010 on paediatric liaison teams. Arrêté royal fixant les normes auxquelles la fonction liaison pédiatrique doit répondre pour être agréée http://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html (consulted 15 Jul 2020)

108. Ministry of Public Health in Belgium. Loi relative aux soins palliatifs du 14 juin 2002 http://www.soinspalliatifs.be/images/pdf/loi_2002_06_14.pdf (page consultée le 6 juin 2020)

109. Ministry of Public Health in Belgium. Loi du 28 mai 2002 relative à l'euthanasie http://www.soinspalliatifs.be/images/pdf/loi_2002_05_28.pdf (page consultée le 6 mai 2020)

110. Ministry of Public Health in Belgium. Loi du 21 juillet 2016 modifiant la loi du 14 juin 2002 relative aux soins palliatifs en vue d'élargir la définition des soins palliatifs http://www.soinspalliatifs.be/images/pdf/Loi_2002_SP_-_MOD_2016_07_21.pdf (page consultée le 6 mai 2020)

111. Ministry of Public Health in Belgium. Loi du 28 février 2014 modifiant la loi du 28 mai 2002 relative à l'euthanasie, en vue d'étendre l'euthanasie aux mineurs http://www.soinspalliatifs.be/images/pdf/LOI_MOD_LOI_2002-05-28.pdf (page consultée le 6 mai 2020)

112. Dan B, Fonteyne C, de Cléty SC. Self-requested euthanasia for children in Belgium. Lancet. 2014;383(9918):671-2.

113. Brouwer M, Kaczor C, Battin MP, Maeckelberghe E, Lantos JD, Verhagen E. Should Paediatric Euthanasia be Legalized? Paediatrics. 2018;141(2):e20171343.

114. Cuman G, Gastmans C. Minors and euthanasia: a systematic review of argument-based ethics literature. Eur J Pediatr. 2017;176(7):837-847.

115. Hain RD, Euthanasia: 10 Myths. Arch Dis Child 2014;99(9):798-9.

116. Friedel M. Does the Belgian law legalising euthanasia for minors really address the needs of life-limited children? Int J Palliat Nurs. 2014;20(6):265-7.

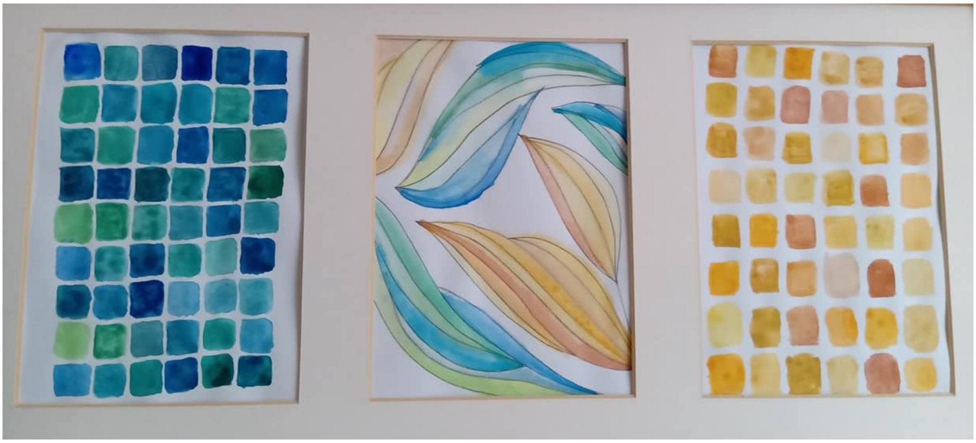
117. Friedel M, Terwangne B, Brichard B, Ruysseveldt I, Renard M. The Belgian euthanasia law and its impact on the practises of Belgian paediatric palliative care teams. Int J Palliat Nurs. 2018;24(7):333-337. doi: 10.12968/ijpn.2018.24.7.333.

118. Friedel M, Brichard B, Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care. 2018;17(1):77. doi:10.1186/s12904-018-0324-2

119. Friedel M, Gilson A, Bouckenaere D, Brichard B, Fonteyne C, Wojcik T, De Clercq E, Guillet A, Mahboub A, Lahaye M, Aujoulat I. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. BMJ Paediatr Open. 2019;3(1):e000547. doi:10.1136/bmjpo-2019-000547

120. Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi:10.1542/peds.2018-2379

121. Friedel M, Brichard B, Boonen S, Tonon C, De Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse JM, Aujoulat I. Face and Content Validity, Acceptability, and Feasibility of the Adapted Version of the Children's Palliative Outcome Scale: A Qualitative Pilot Study [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646



Mixed-methods design

by F. Castorini and M. Friedel

Photo credits @ Marie Friedel

CHAPTER 2. METHODOLOGY

This chapter focusses on the paradigms, the definition and different types of mixed method study designs that inspired our research. The detailed methods of each sub study are described in chapter 3.

Paradigms in research

In science, different approaches are classified depending on their underpinning philosophical paradigms which are summarized in table 1 (1-3). The positivist approach affirms only one objective reality, which can be observed and for which a hypothesis is verified through quantitative methods. Studies relying on a realist approach acknowledge the multiple influences of the environment on reality and the interconnection between the researcher and their subjects. In this approach, qualitative methods are often used to document and explore the mechanisms underlying a reality. In a constructivist approach, reality is socially constructed, subjectivity highlighted and methods focusing on a hermeneutical approach are privileged.

Table 1. Different approaches in research characterized by philosophical paradigms based on Creswell 2003, Greenhalgh 2010, Klein & Meyers 1999

	Positivism	**Realism**	**Constructivism**
Ontology (Theory of being)	Only what is objective is real	Reality shaped by social, political, cultural, economic values	Reality is socially constructed
Epistemology (Theory of knowledge)	Objective scientific findings are true	Subjectivity, value-mediated findings. Findings are probably true	Subjectivity, created findings
Methodology	Dualist, verification of hypothesis, quantitative methods	Dialogic, dialectic, mechanism-based,	Hermeneutical, interpretative, meaning-making, qualitative methods

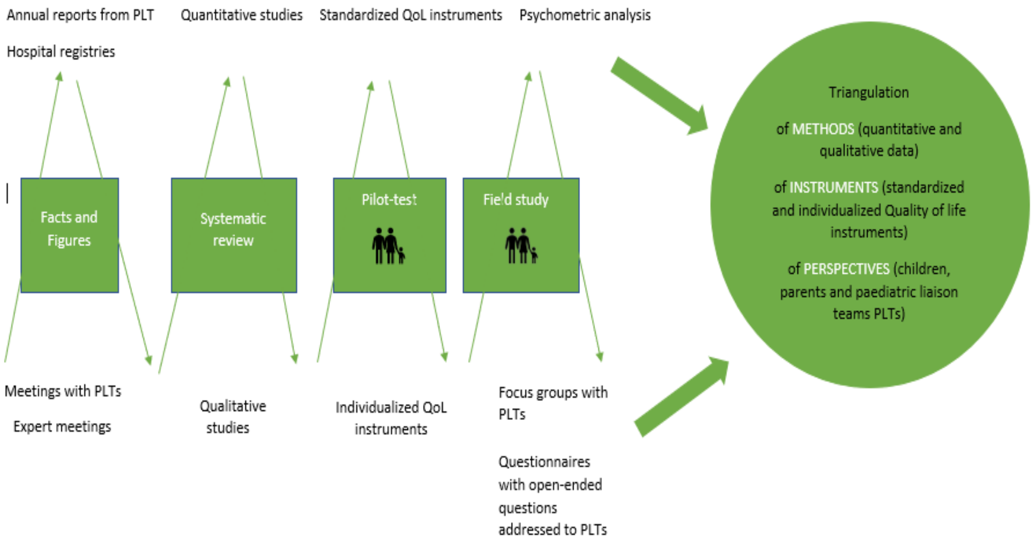
Our study has navigated between a realist, collaborative and meaning-making approach, leaving space for discussions of different perspectives among stakeholders, and a positivist approach concretized in the psychometric analysis we conducted. Just like Howe (4), we do not think that qualitative and quantitative methods should be considered as antagonistic, but rather be acknowledged as complementary, especially when developing an outcome measurement instrument. Such an instrument had to be acceptable and meaningful for children and their parents, feasible for paediatric liaison teams and robust for science. Therefore, a convergent triangulation mixed-method study design was chosen.

Mixed method study design

Mixed method studies, combining quantitative and qualitative methods, are commonly used to examine phenomena from multiple perspectives and approaches. The complementary strength of each approach helps to address one or multiple research questions. Three different types of mixed methods designs were outlined by Curry & Nunez-Smith (5) and Creswell and Plano Clark (6) and are: convergent, exploratory sequential and explanatory sequential study design. The differences are based on how the data are integrated (merged, embedded or connected), the sequence in which the data were collected on a timeline (concurrent, or qualitative before - or after - quantitative data collection) and which method was prioritized (quantitative or qualitative). In our study we used a triangulation study design. A concurrent quantitative and qualitative data collection and a convergent data analysis were used to combine findings (embedded integration) from each type of analysis (1,7).

The triangulation of perspectives (see Figure 2) between children, parents and professionals helped us to consider the complexity of the concept of quality of life (QoL) but also to foster the implementation process of the measurement instrument and the adoption of it by paediatric liaison care teams. Furthermore, we combined quantitative and qualitative methods to explore quality of life and further develop an outcome measurement instrument. Finally, to identify meaningful dimensions of QoL for children, we used an interview guide, the Scheduled evaluation of individual quality of Life-direct weighted (SEIQoL-DW) (8). With the SEIQoL, which evaluates individual QoL, meaningful QoL domains were elicited from the children themselves. These domains were then integrated as items into the standardized further developed children's palliative outcome scale, version 2 (CPOS-2).

Figure 2. Convergent triangulation design of the MOSAIK study, based on Creswell 2003, Curry &Nunez-Smith 2015 and Creswell & Plano Clark 2018, (1,5,6)



Legend: PLTs - paediatric liaison teams, QoL - Quality of Life

👪 Data collection among children and their parents

Overview of the methods used for each sub-study

Methods used to present facts and figures of paediatric palliative care in Belgium[2]

Two different studies were conducted to present facts and figures on paediatric palliative care in Belgium. They referred to the same timeframe: 2010-2014.

The first study (9) collected the data from annual reports of all paediatric liaison care teams (PLTs) officially recognised in Belgium. Those annual reports had been mandatory since 2010, the year at which the PLT's activities were structurally funded. Thematic analysis of open-ended questions was performed, and descriptive statistics of aggregated data issued

---

[2] See chapter 3

from annual reports, collected by the Belgian Ministry of Public Health through the Cancer Plan, was used. A review panel of PLT members discussed the results and contributed to their interpretation.

The second study (10) collected data from hospital registries compared to paediatric liaison teams' registries in order to explore access to those teams. To align with Belgian hospital registration systems in use from 2010 to 2014 (period considered for our study), which used International Classification of Disease codes (ICD-9), we had to rely on lists of complex chronic conditions linked to ICD-9. We used therefore a list of complex chronic conditions described by Feudtner et al. (11). All children's complex chronic condition , classified in ICD-9 and linked to a national registration numbers were extracted from the databases of all hospitals (n=8) and PLTs (n=2) based in the Brussels region. Aggregated data and pseudonymised national registration numbers were transmitted to the research team by a Trusted Third Party (eHealth). Ages and diagnostic categories were calculated using descriptive statistics.

Methods used for the systematic review[3]

To search for instruments used to assess the impact of PPC interventions, we conducted a systematic review (12) based on PRISMA reporting guidelines (13). Five databases (Embase, Scopus, The Cochrane Library, PsychInfo, Medline) were searched. Inclusion criteria were as follows: definition of PPC used; patients aged 0 to 18 years; diseases listed in the directory of life-limiting diseases; results based on empirical data; and combined descriptions of a PPC intervention, its outcomes, and a measurement instrument. Full-text articles were assessed, and data were extracted by two independent researchers, and each discrepancy was resolved through consensus.

Methods used for the pilot-test[4]

The detailed methods were described in a published article (14).

The original 12-item CPOS in English is composed of five questions exploring children's outcomes and addressed directly to the child (or substituted by a proxy-report in cases where the child is not able to respond) and seven questions related to parents' outcomes.

After producing a cross-cultural translation of the English version of the original 12-items CPOS into French using an appropriate methodology (forward translation, backward translation, review by experts as recommended by Antunes et al. (15), a pilot-test was started in 2017 at one PLT center in Belgium. An iterative and collaborative approach with

---

[3] See chapter 4

[4] See chapter 5

PLTs was chosen throughout the study. Semi-structured interviews were conducted among children and their parents, with a paediatric liaison nurse as an observer. Families could choose the place (home or hospital) they wanted to be interviewed. During the interviews, the CPOS, the interview guide of the SEIQOL-dw (16) and the quality of life in life threatening Illness-family caregiver (QOLLTI-F) (17), were used. By asking the child through the SEIQoL-dw to self-elicit the domains that mattered most to them and using the QOLLTI-F questionnaire, we verified the relevance of dimensions covered by the original CPOS. Three expert review panels composed of paediatric palliative care health professionals assessed the relevance and feasibility of the further developed CPOS-2, consisting of 22-items.

Methods used for the field-test[5]

We followed the guidance on the development of outcome measurement instruments issued by the COSMIN study group (18). See Figure 3. The COSMIN study aimed to reach consensus among experts with a background in psychometrics, epidemiology, statistics and clinical medicine, about which measurement properties are considered to be important, terms and definitions used and how they should be assessed in study designs (19).

Psychometrics refers to a methodological discipline rooted in psychological research, but increasingly used also in medicine and health.

As we aimed to assess the psychometric properties of a measurement instrument, we followed the steps recommended by the COSMIN study group, which are listed below:

First, the construct to be measured (quality of life) must be defined. If this construct has different aspects, then the measurement instrument should be multidimensional and cover all these aspects.

Second, the measurement instrument should be tailored to the target population. Age, gender and severity of diseases of the population for which the instrument is developed must be defined.

Third, the purpose of measurement should be defined. Will the instrument help the diagnosis, the evaluation of therapy or predict the future course? For our purpose it is to assess children's quality of life and to gather meaningful information, to guide the caregiving process and to assess the impact of interventions and therefore to improve quality of care.

Fourth, the type of measurement instrument should correspond closely to the construct to be measured. Do we want to measure what children can do or what they perceive, what they feel? Furthermore, a choice must be made between a single item or a multi item

---

[5] See chapter 6

instrument, which implies a discussion on reliability and definition of the construct. General opinion states that complex constructs, such as quality of life, are best measured with multi-item instruments, but in some cases, such as to reduce the administrative burden, single-item instruments might be preferable (20). Regarding the definition of the construct (quality of life), if there are many different items in an instrument, it maximises the chance to cover all dimensions considered to be relevant for a specific construct. This makes it easier for the patient to understand those items. Furthermore, multi-items instruments might also help in the interpretation and validation of the measurement instrument (19).

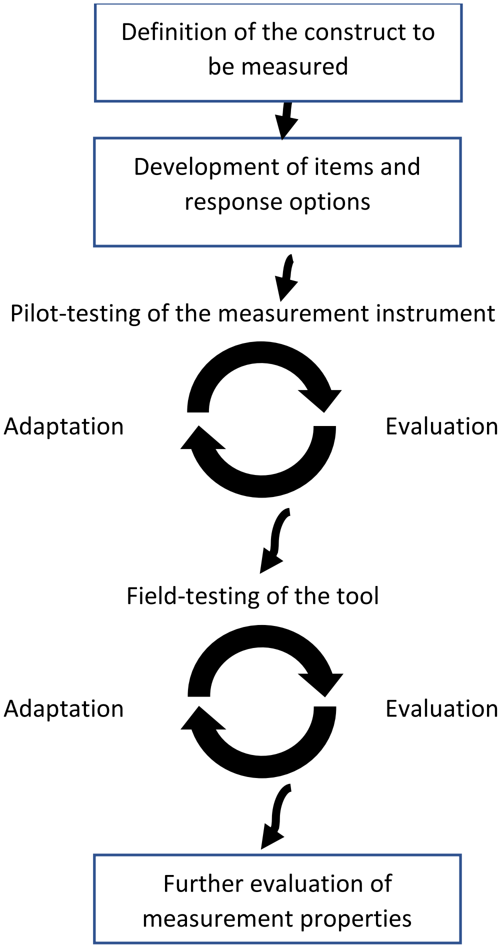
Fifth, items to integrate in the instruments should be selected based on literature, experts (such as the patients themselves, their parents and caregivers). Those items are often found through qualitative studies. Those items should be then formulated to be comprehensive, specific and clear, avoiding negative wording or multiple meaning. At this stage, it is important to decide whether the measurement instrument will follow a reflective or a formative model. Instruments based on reflective models include items which influence the constructs (e.g. a broken leg will affect physical functioning), whereas formative models include items which reflect the constructs (e.g. crying reflects the pain). Often, for questionnaires assessing QoL, which is a multidimensional construct, mixed models integrating reflective and formative items are used.

Sixth, scoring options must be defined beforehand. The response to a single item can be expressed at nominal level (number of classes without an order), at ordinal level (number of classes with an order, such as Likert scales) and at interval (scores are expressed in numbers, such as Temperature) or ratio level (with an absolute zero point, such as tumour sizes). Decide how summarizing scores for formative and reflective models must be defined.

Seventh, a pilot-test should be conducted to test the comprehensibility, relevance and completeness of the measurement instrument among the target population in order to adapt the items if necessary.

And finally, a field-testing of the measurement instrument should be conducted in an iterative way to assess the psychometric properties of the tool and further adapt the items (item reduction and data structure).

Figure 3. Steps in the development and evaluation of a measurement instrument based on de Vet et al 2011 (19)



Despite the methodological challenges to develop an outcome measurement instrument, a promising tool, developed by the African Palliative Care Association (APCA) - called the APCA children's palliative outcome scale (APCA C-POS) - was developed in an African context of care (21). Based on the APCA palliative outcome scale for adults (22), it combines a self- and proxy-report. The original C-POS was published during this thesis (23). The C-POS is currently being tested in the UK, in Turkey and in Uganda by the team of Pr. R. Harding at Cicely Saunders Institute from King's College London, United Kingdom (24). The C-POS is a multidimensional patient-centred outcome measure, with 5-point Likert-scale response options. It contains 12 items exploring physical and psychosocial elements. Seven items are related to children's quality of life.

We decided to further build on this instrument, although its psychometric properties were not yet assessed nor described, for several reasons. It was short, meaning it would not put an additional burden on children, parents and healthcare professionals if used in a routine clinical care setting. Secondly, it was elaborated for and by a paediatric palliative care population, maximising its potential face and content validity. Thirdly, it included in the same scale a child self-report and a parent proxy-report, as recommended by standards in order to address the lack of cognitive/verbal child's capacity, and fourthly, it evaluates children and parents' quality of life in a family-centred approach.

Nevertheless, the APCA CPOS was previously pilot-tested in an African context (Uganda, Kenya, South Africa), with a different organization of care and among a population of children mostly suffering from HIV (23). Furthermore, the scale was not available in French nor tested for its psychometric properties. For all these reasons, we decided to follow the steps mentioned above to adapt the CPOS to in a Belgian context of paediatric palliative care, explore its reliability and validity, and to understand PLT experiences when using it.

Research protocol of the field-study

Context of the study

The protocol of this field-test is part of the MOSAIK study (Move to Open Shared Advanced Interventions for Kids with life-limiting conditions) which has the overall aim to further develop a measure instrument to explore and measure quality of life of children facing life-limiting / life-threatening conditions.

The MOSAIK study is conducted as a collaborative project with the six national paediatric PLTs, who ensure continuity of care for children with life-limiting conditions.

We decided to explore the psychometric properties of the French version of the APCA-Children's Palliative Outcome Scale (CPOS-2) through triangulation, by combining the perspectives of children facing life-limiting conditions, their parents and health care professionals. Taking into account the perspectives and experiences of health care professionals, the main stakeholders, while developing an instrument is not only important for the identification of the core dimensions of those instruments but also for the assessment of the feasibility and acceptability of using those instruments in daily practice. Involving professionals during the whole process and documenting their difficulties and resources in an iterative way, is an essential key to a further successful implementation of an instrument (25-27).

Objectives of the study

i) To describe and analyse the characteristics of a representative sample of children facing life-limiting or life-threatening conditions;
ii) To explore the applicability of the CPOS-2 in French and in Dutch in a heterogeneous (different ages, different types and stages of diseases) population of children facing life-limiting or life-threatening conditions;
iii) To document the psychometric properties of the modified CPOS-2;
iv) To document the experiences of PLTs when using the CPOS-2 (challenges and resources perceived by health care professionals and impact on care provided)

Methods

DESIGN : multicentric cross-sectional study

TIME FRAME : from 15th February 2019 to 15th February 2020

Before starting, ethical committee approval in each PLT must be collected and some instruments (CPOS, QOLLTI-F) still need to be translated into Dutch (forward, backward, semantic expert review meeting). This explains why the four French PLTs will start to include children and their families in October 2018, whereas the two Dutch PLTs will start in July 2019.

Methods used for objectives i), ii), iii)

INCLUSION CRITERIA : children and adolescents (1-18 years) facing any life-limiting or life threatening conditions and cared for by one of the six paediatric liaison teams in Belgium (Globul'home at Hôpital des enfants Reine Fabiola, Interface pédiatrique at Cliniques universitaires st Luc, CHR Citadelle Liège, CHC Espérance Liège, Kites Leuven, Koester Gent)

EXCLUSION CRITERIA: neonates cared for in a neonatology unit and who have never left hospital since their birth. Children and parents who are not able to understand and/or to speak French or Dutch. Children or adolescents at a very close end-of -life stage will not be invited to participate in the study (expected last week of life).

SAMPLE SIZE: 100-150 children and their parents

In this study, the estimation for a required sample size can be made from different perspectives.

In response to the first research question (to describe the characteristics of a representative sample of children facing life-limiting or life-threatening conditions) and from a *survey perspective,* the sample size can be calculated using a confidence level of 95% , a confidence interval of 7% and a total population of 700.This yields a sample size of 153.

From the perspective of the third research question (a study of the psychometric characteristics), no consensus exists to define sample size in studies aiming to document the psychometric characteristics of newly-developed patient-reported outcome measurement tools (28).

Two options, however, seem to be reasonable:

1. With regard to concurrent validity where correlations coefficients are calculated, the desired precision of the correlation can be used as a point of departure to calculate the sample size, which would be 194.

2. With regard to exploratory factor analysis this would mean including a minimum of 110 children in the sample.

DATA COLLECTION and ethical considerations:

Recruitment of children and parents will be conducted through the paediatric liaison teams during their routine visits to children and their families at home.

Children and parents will be informed of the study by the paediatric liaison nurses and invited by them to participate.

Signed consent to participate themselves and to let their child participate in the study will be requested from each parent, as required by the Belgian Law on privacy passed on 8th December 1992 and the law related to patients' rights passed on 22nd August 2002. Additionally, even if it is not requested by law, but with the aim of respecting each child and assessing his willingness to participate, developmentally adapted and gentle assent processes will be used.

If a parent and child refuse to participate in the study, medical and socio-demographic characteristics of the child will be reported by PLT nurses in order to document potential selection bias.

Recruitment will be undertaken with clear safety protocols to minimise risk to participants. A distress protocol is developed in case any interviewee becomes distressed, fatigued or unable to continue for other reasons, and a psychologist in each paediatric liaison team will be available to address any adverse events. Interviews will be conducted wherever possible at the child's home, considered as the usual and safe environment for the child.

Following the same reasoning, each child will be asked if he prefers to be interviewed alone or in presence of his parents, because the risk of having desirability bias in the results is considered less important that the risk for the child of feeling insecure or worried.

In order to guarantee a maximum level of standardization in data collection in each paediatric liaison team, a video will be provided to each team explaining how the data should be gathered (how to approach children and parents, how to conduct the interview with children and parents and how to fill in the questionnaires).

Instruments used

Children's palliative outcome scale CPOS-2 in a French version and in a Dutch version

CPOS-2 include 12 questions exploring children's QoL (self- or proxy-reported depending on child's age or cognitive capacity) and 10 questions assessing parental QoL. Dimensions include physical, psychological symptoms, family and social interactions. Responses are given through a 5-point Likert scale by asking children to show with their fingers how many points they want to give to each question. This permits the inclusion of children with low verbal capacities. The face and content validity of the original CPOS in English was tested among 302 children (23).

The Quality of life in life-threatening illness - family carer version (QOLLTI-F) instrument

This is a self-report questionnaire presented as a rating scale exploring, through 7 dimensions and 17 questions, the caregiver's quality of life. Dimensions include: the overall perceived QoL of the caregiver, the patient's environment and condition, the caregiver's physical, emotional and cognitive condition, his perceived meaning in life, the quality of care, quality of relationships and financial worries. Responses are provided through a 10-points Likert scale (17). The QOLLTI-F shows excellent internal consistency with a Cronbach α of 0.857 and was already successfully tested within a PPC context (29, 30).

KINDL

KINDL is a generic quality of life instrument to be used with children with a wide range of diseases and conditions. It contains a child report and a proxy report to assess quality of life through 7 domains and 30 questions and is available in 3 versions, depending of the child's age: 4-6 years, 7-13 years, 14-17 years. Responses are given through a 3-item or 5-item Likert-scale. High concurrent validity was found with the Kidscreen-52 and discriminant validity when used among healthy children (31,32).

Background variables

-On data collection process: Children interviewed alone or in presence of his parents, place where interview took place (house, hospital, institution), duration of the interview (in minutes).

-On personal information of the child: age, day-activity (school, institute for disabled children, at home), postal code of residency.

-On family information: marital status (in couple, separated, alone), number and age of siblings, any other family member diseases, socio-economic status, education level and profession of parents.

-On medical information completed by the paediatrician in charge of the child: name of disease (according international classification of disease ICD-10), date of diagnosis, date of starting follow-up by PLT, list of pharmacological treatments and dosage at time of interview.

-On specific items considered as criteria to refer children to a paediatric liaison care team and to evaluate the palliative stage (33-35): life expectancy estimated, impact of disease on daily activities, burden of curative treatment on quality of life as perceived by physician, parental suffering/distress , types of medical devices/equipment necessary to the child, degree of parental acceptance of the child’s condition as perceived by physician.

Frequency of assessment: All child/parent data will be collected once by PLT nurses during their routine visit to the child at home or in hospital.

All the data collected by PLT nurses and specialized hospital-based physician will be sent, anonymised, to the principal investigator. A unique code will be created for each child. The link between the code and the identity of the child and its parents will only be known by the personal professional caregivers. E.g.:

A (for the name of paediatric liaison team 1) 01 (for the first patient) they included in the study.

B (for the name of paediatric liaison team 2) 01 (for the first patient they included in the study.

A central database will be created in which all collected data will be stored using a unique identifier. Concretely, each paediatric liaison care team will be provided with 30 investigator’s handbooks, based on a carbon sheet-paper, including all boxes with predefined questionnaires and background variables.

The first page of the investigator’s handbook containing child’s name will be kept by each paediatric liaison care team, whereas the second copied page including a unique numerical identifier per child (and without the name of the child) will be send to the researcher team for analysis.

DATA ANALYSIS

Methods used for objectives i) ii) and iii),

Descriptive statistics will be used in order to explore the distribution of the scores on the different scales.

Bi- and multivariate analyses will be used in order to explore the associations between scores on the different scales and the background variables.

Several analyses of the psychometric characteristics of the CPOS-2 will be conducted:

1. Reliability analysis. The internal consistency will be evaluated by calculating Cronbach's alpha, and to calculate a Standard Error of Measurement (SEM). A generalizability analysis will be performed in order to identify the different sources of measurement error (G-study) and to assess the reliability of decisions taken on the basis of different cut-off scores (D-study).The generalizability analysis will also be used to investigate reliability in function of test length.
2. An exploratory and confirmatory factor analysis will be performed in order to explore the dimensionality of the construct measured by the CPOS-2scale.
3. Item response models will be used in order to identify redundant items and to explore differential item functioning.
4. The concurrent validity of the CPOS-2 scale will be explored by comparing the correlation of scores of the CPOS-f with scores on the other scales used.

Methods used for objective iv) to document the experiences of PLTs when using the CPOS-2 (challenges and resources perceived by health care professionals and impact on care provided) of the CPOS-2 into routinely clinical care.

Data collection

Focus groups with each team (two focus groups in the 2 Dutch PLT and four focus-groups in the 4 French-speaking PLT in Belgium) will be conducted in order to assess the resources, difficulties and challenges experienced by PLT nurses during the interviews and how they perceive the impact of using the CPOS-2 on the individualized care plan.

Data analysis

Methodologies using collaborative approaches (36,37) and qualitative thematic analysis will be used to document the experiences of PLT (38).

In the next paragraph, we will explain how the ethical issues raised in the introduction section regarding PPC research informed some of our methodological choices.

Ethics in Paediatric Palliative Care Research: Recommendations which guided our methodological choices

Principles of ethics in paediatric palliative care research have already been described earlier in this thesis. We would like to illustrate how -with which methods - we integrated those into our study.

First, we decided that the study should serve children's best interests (beneficence. The interview between team members and family during which data would be collected should be an opportunity for paediatric liaison teams to better understand children's views and needs and accordingly offer individualized care. Exploring quality of life and what matters most to a child should lead to consequent suggestions made by the team. For example, if pain appears to be important, teams would be immediately easing the pain with accurate interventions, or if a teenager was expressing his sadness seat not seeing his peers, PLTs could offer them the opportunity to join social activities with peers.

Secondly, we conceived three different types of informed assent sheets, according to the different age groups (5-8 years, 9-15 years and 16-18 years), adapting the vocabulary used and leaving enough time for the child to read and understand the information.

Third, invitation, recruitment and interviews of children and parents were conducted through the child's usual carer, to provide a sense of security. Security was also provided by offering to contact a psychologist if needed and finally by letting the child decide where and with whom he preferred to be interviewed. This choice would give them some control over the interview. Some adolescents possibly don't want their parents to see which score they gave to each item. The intimate relationship between children and parents, made of loyalty and mutual protection, should be acknowledged and protected (39).

Fourth, flexibility was reached by letting teams adapt the interview to children's and parents' preferences (place and time of interviews, ideally during a routine home visit, to leave the child choosing to be interviewed alone or beside his parents).

Fifth, short and age appropriate outcome measures and interview guide were chosen, which allowed self-report, inviting children to express themselves about how they felt and what was important to them.

Sixth, triangulation, as the basics of paediatrics, was acknowledged by exploring children's and parents' perspectives on children's quality of life (self- and proxy-reported), parents' experience of their own quality of life and team's point of view on the feasibility of using an outcome measures instrument.

Seventh, we chose a study design based overall on a collaborative approach with paediatric liaison teams, recognized as experts in the field. This was reflected through a needs assessment with paediatric liaison teams highlighting an interest in developing and using a measure outcome instrument to assess children's quality of life, their active involvement in the pilot-testing and the field-testing of this instrument, in addition to several meetings to fine-tune the tool and to understand the implementation process of it.

References

1.Creswell, JW (2003). Research design: Qualitative, quantitative and mixed methods approaches. London: Sage Publications.

2. Greenhalgh T. Why do evaluation of e-health programs fail? An alternative set of guiding principles. PLoS Med. 2010;7(11):e1000360. doi: 10.1371/journal.pmed.1000360.

3. Klein HK, Myers MD (1999) A set of principles for conducting and evaluating interpretive field studies in information systems. Mis Quarterly 23: 67-93.

4. Howe K R. Against the Quantitative-Qualitative Incompatibility Thesis or Dogmas Die Hard. Educational Researcher. 1988, 17(8), 10–16. https://doi.org/10.3102/0013189X017008010.

5. Curry L & Nunez-Smith M (2015). Mixed methods in health sciences research: a practical primer. Thousand Oaks, CA: Sage in Kahwati LC, Kane HL. (2015). Qualitative comparative analysis in Mixed methods research and Evaluation. Sage Publications.

6. Creswell J, Plano Clark V (2018). Designing and conducting mixed-methods research (3rd ed), Thousand Oaks, CA: Sage.

7. Razum O, Gerhardus A. Methodological Triangulation in Public Health Research - Advancement or Mirage? Trop Med Int Health 1999 (4):243-4. doi: 10.1046/j.1365-3156.1999.00399.x.

8. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33. doi: 10.1136/bmj.313.7048.29.

9. Friedel M, Brichard B, Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care. 2018;17(1):77. Published 2018 May 22. doi:10.1186/s12904-018-0324-2

10. Friedel M, Gilson A, Bouckenaere D, Brichard B, Fonteyne C, Wojcik T, De Clercq E, Guillet A, Mahboub A, Lahaye M, Aujoulat I. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. BMJ Paediatr Open. 2019;3(1):e000547. doi:10.1136/bmjpo-2019-000547

11. Feudtner C, Hays RM, Haynes G, et al. Deaths attributed to paediatric complex chronic conditions: national trends and implications for supportive care services. Paediatrics 2001;107:E99.

12. Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi:10.1542/peds.2018-2379

13. Moher D, Liberati A, Tetzlaff J, Altman DG; PRISMA Group. Preferred Reporting Items for Systematic Reviews and Meta-analyses: the PRISMA statement. PLoS Med. 2009;6(7):e1000097.

14. Friedel M, Brichard B, Boonen S, Tonon C, De Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse JM, Aujoulat I. Face and Content Validity, Acceptability, and Feasibility of the Adapted Version of the Children's Palliative Outcome Scale: A Qualitative Pilot Study [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646

15. Antunes B, Daveson B, Ramsenthaler C, Benalia H, Ferreira P, Bausewein C, Higginson I. The palliative care outcome Scale. Manual for cross-cultural adaptation and psychometric validation. Kings College London, Cicely

Saunders Institute, Department of Palliative Care, Policy and Rehabilitation, London, March 2012 , http://pos-pal.org/Resources.php

16. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33. doi: 10.1136/bmj.313.7048.29.

17. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006 Dec;20(8):755-67. doi: 10.1177/0269216306072764.

18. Mokkink LB, Terwee CB, Patrick DL et al. (2010). International consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes: results of the COSMIN study. Journal of Clinical Epidemiology, 63, 737-745.

19. De Vet HCW, Terwee CB, Mokkink LB, Knol DL. (2011) Measurement in Medicine. Cambridge. Cambridge University Press.

20. Sloan JA, Cella D, Hays R. Clinical significance of patient-reported questionnaire data: another step toward consensus. Journal of Clinical Epidemiology 2005,58, 1217-9.

21. African Palliative Care Association. Manual of the children’s palliative outcome scale (APCA-CPOS). 2012.

22. Powell RA, Downing J, Harding R, Mwangi-Powell F & Connor S on behalf of the APCA M&E Group. Development of the APCA African Palliative Outcome Scale. Journal of Pain and Symptom Management 2007, 33: 229-232.

23. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02.

24. Harding R. ERC Research grant on the C-POS. https://www.kcl.ac.uk/news/measuring-palliative-care-needs-of-children-and-young-people (consulted 15th June 2020)

25. Peters DH. Implementation research: what it is and how to do it. BMJ 2013;347:f6753

26. Luig T, Asselin J, Sharma AM, Campbell-Sherer DL. Understanding Implementation of Complex Interventions in primary care teams. JABFIM 2018; 31(3):431-444.

27. International Society for Quality of Life Research (prepared by Aaronson N, Elliott T, Greenhalgh J, Halyard M, Hess R, Miller D, Reeve B, Santana M, Snyder C). User’s Guide to Implementing Patient-Reported Outcomes Assessment in Clinical Practice, Version: October 2014.

28. Anthoine E, Moret L, Regnault A, Sébille V, JB. Sample size used to validate a scale: a review of publications on newly-developed patient reported outcomes measures. Health Qual Life Outcomes. 2014; 12: 2.

29. Bradford N, Young J, Armfield NR,et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. J Telemed Telecare 2012;18(8):438-442.

30. Groh G, Borasio GD, Nickolay C, et al. Specialized paediatric palliative home care: a prospective evaluation. J Palliat Med. 2013;16(12):1588-1594. doi: 10.1089/jpm.2013.0129.

31. Erhart M, Ellert U, Kurth B, Ravens-Sieberer U, Measuring adolescents’ HRQoL via self and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument. Health and Quality of Life Outcomes. 2009; 7:77.

32. Bullinger M, Brütt A, Erhart M, Ravens-Sieberer U and the BELLA study group. Psychometric properties of the KINDL-R questionnaire: results of the BELLA study. Eur Child Adolesc Psychiatry (suppl 1) 2008 ; 17 :125-132.

33. Bergstraesser E, Hain RD, Pereira J. The development of an instrument that can identify children with palliative care needs: the Paediatric Palliative Screening Scale (PaPaS Scale): a qualitative study approach. BMC Palliative Care 2013; 12:20

34. Bergstraesser E, Paul M, Rufibach K, Hain RD, Held L. The Paediatric Palliative Screening Scale: Further validity testing. Palliat Med. 2014 Jun;28(6):530-533.

35. Matraka N. Critères d'identification des enfants nécessitant l'intervention de l'équipe de liaison en soins palliatifs pédiatriques selon les médecins des Cliniques Universitaires Saint-Luc : analyse qualitative et test d'un outil d'aide à la décision : la Paediatric Palliative Screening Scale. Faculté de santé publique, Université catholique de Louvain, 2018. Prom. : Aujoulat, Isabelle ; Friedel, Marie.

36. Abma TA, Pittens CAC, Visse M, Elberse JA, Broerse JE Patient involvement in research programming and implementation: A responsive evaluation of the Dialogue Model for research agenda setting *Health Expect*. 2015 Dec; 18(6): 2449–2464.

37. Abma T. Evaluating palliative care: Facilitating reflexive dialogues about an ambiguous Concept. Medicine, Health Care and Philosophy 4: 261–276, 2001

38. Paillé P, Muchielli A. 2008. L'analyse qualitative en sciences humaines et sociales. 3ème édition. Armand Colin. Paris.

39. Bluebond-Langner M, DeCicco A, Belasco J. Involving Children with Life-shortening Illnesses in Decisions about participation in clinical research: a proposal for shuttle diplomacy and negotiation, in Kodish E. (ed.) Ethics and Research with Children. A case-based approach. 2005.

Photo credits: Marie Friedel

This sculpture, entitled « Les arbres protégés » from the artist Nathalie Joiris is located in the Woluwe parc in Brussels. It can be seen as a metaphor for children, who are living and growing (tree) despite the serious diseases or life-limiting or complex chronic conditions they face (stones).

PART II RESULTS

In this chapter we will present the results of each studies who are included in the MOSAIK study (**M**ove to **O**pen **S**hared **A**dvanced **I**nterventions for **K**ids with life limiting conditions), namely:

the 2 studies describing the **facts and figures** on paediatric palliative care services in Belgium; one based on paediatric liaison annual reports from 2010 to 2014 and the second one on hospital registries in the region of Brussels from 2010 to 2014 (CHAPTER 3)

the **systematic literature review** on instruments used to measure outcomes in paediatric palliative care (CHAPTER 4);

the **pilot-study** (CHAPTER 5) on further developing the children's palliative outcome scale (CPOS);

and the **national multicentric field-study** (CHAPTER 6) on children's and parents' quality of life, with the presentation of the study protocol (CHAPTER 7), the psychometric properties of the CPOS-2, the overview of children's and parent's quality of life and the qualitative study among paediatric liaison teams.

As most of those chapters were published or are in the process of being published, we kept the original structure (Abstract-Introduction-Objectives-Methods-Results-Discussion).

CHAPTER 3. FACTS AND FIGURES ON PAEDIATRIC PALLIATIVE CARE IN BELGIUM

3.1. ACTIVITIES OF PAEDIATRIC LIAISON TEAMS IN BELGIUM AND PROFILE OF CHILDREN FOLLOWED-UP

This chapter was published as an article under the title:

Friedel M, Brichard B, Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care. 2018;17(1):77. doi: 10.1186/s12904-018-0324-2.

ABSTRACT

Background and objectives: Although continuity of care in paediatric palliative care (PPC) is an essential element of quality of care, its implementation is challenging. In Belgium, five paediatric liaison teams (PLTs) deliver palliative care. A Royal Decree issued in 2010 provides the legal framework that defines the PLTs' missions, as ensuring continuity of curative and palliative care between the hospital and home for children diagnosed with life-limiting conditions. This national study describes how PLTs ensure continuity of care by describing their activities and the characteristics of the children they cared for from 2010 to 2014.

Methods: Thematic analysis of open-ended questions was performed, and descriptive statistics of aggregated data issued from annual reports, collected by the Belgian Ministry of Public Health through the Cancer Plan was used. A review panel of PLT members discussed the results and contributed to their interpretation.

Results: Between 2010 and 2014, 3607 children and young adults (0-21 years) were cared for by the 5 Belgian PLTs (mean of 721/per year). Of these children, 50% were diagnosed with an oncological disease, 27% with a neurological or metabolic disease. Four hundred and twenty-eight (428) children had died. For 51% of them, death took place at home. PLT activities include coordination; communication; curative and palliative care; education; research and fundraising. Different perceptions of what constitutes a palliative stage, heterogeneity in reporting diagnosis and the current lack of specific valid indicators to report PPC activities were found.

Conclusion: PLTs are offering highly individualised, flexible and integrated care from diagnosis to bereavement in all care settings. Improvements in data registration and implementation of outcome measures are foreseen.

Introduction

Children with life-limiting or life-threatening conditions require paediatric palliative care (PPC) to various degrees and intensity. The World Health Organization and the European Association of Palliative Care define activities of PPC as “the active total care of the child's body, mind and spirit, (as well as) giving support to the family. It begins at diagnosis and continues regardless of whether or not a child receives treatment directed at the disease. Healthcare providers must evaluate and alleviate a child’s physical, psychological and social distress. Effective palliative care requires a broad multidisciplinary approach that includes the family and makes use of available community resources.” (1)

A recent cross-sectional analysis conducted in 23 countries revealed that 11 million children warrant a PPC approach, with 8 million of them needing access to specialised PPC services. (2)

Various types of PPC services have developed over the past 20 years in Europe. These can be classified into three main categories: hospital-based; freestanding facilities or home-based services. To date, it is not known which one of these services best ensures continuity of care. (3)

Continuity of care is a fundamental part of PPC because of the high number of health professionals the family is in contact with, the various care settings involved (in and out of hospital), and the significant distress experienced by the child and his/her family at different stages (illness, death, bereavement). From the perspectives of families, continuity of care is seen as a “seamless” organisation of care (4) and is a major element of quality palliative care. What remains unclear is how continuity is defined and what are the enablers and barriers to continuity of care in PPC.

Haggerty defines 3 types of continuity: informational continuity, management continuity and relational continuity (5). Informational continuity involves the delivery of relevant information in a timely manner. Management continuity involves sharing consistent and adaptable goals across care teams and between patients and their carers. Relational continuity refers to a therapeutic relationship with one or more health professionals over time.

Further definitions of continuity of care can be found in the multicomponent model described by Freeman (6,7) and reported as follows by Parker : “Continuity was defined as the experience of a co-ordinated and smooth progression of care from the service user’s perspective (experienced continuity), dependent on services that had: excellent information transfer (continuity of information), effective communication between professionals and services, and with patients (cross-boundary and team continuity), the ability to be flexible and adjust to the needs of the individual over time (flexible continuity), one or more named individual professionals with whom the patient could establish and maintain a therapeutic relationship (relational or personal continuity).” (8)

Saultz calls relational continuity the interpersonal continuity of care (9). This is characterised by a trusted relationship between care providers and patients. Similarly the concept of continuing bonds is found to be critical in a PPC context (10), where patients need to be cared for long periods and are in contact with many different health professionals in several care settings. Continuity of care can be achieved through nurse-led care coordination or a nurse case manager offering individualised, family-centred care (11,12).

Inherent to these definitions and according to international recommendations is that PPC should start at the time of diagnosis of a life-limiting disease (1,13). More recently, the World Health Assembly called for governments to integrate PPC within a continuum of care promoting quality of life. (14)

Early referral to PPC services is challenging (15). Cultural barriers and organizational hurdles must be identified and overcome. Whilst there is a lack of descriptions of and comparisons of models in PPC, indicators of continuity of care have been established. (16-21)

These include the following characteristics: family-centred; accessible 24 hours/day care; coordinated between in-and out-patient settings; delivered by an effectively communicating interdisciplinary team and including the possibility to access respite care and bereavement support.

Context of Paediatric Palliative Care in Belgium

The first paediatric home care service was initiated in 1989 in Gent (Flanders) as a mobile team originating from the Oncology Department of a university hospital with the objective of providing curative care to children suffering from cancer. The aim was to reduce visits to the oncology day hospital. This team accepted the challenge to ensure continuity of care for children requiring palliative care who wished to return home.

The Belgian Act on Palliative Care (2002) protects the rights of all patients to receive comfort care in case of an incurable disease (22), after disease-oriented therapies have failed.

In Belgium, PPC is offered through 5 specialised paediatric liaison teams (PLTs) affiliated to 5 university hospitals. Two are located in Flanders, 2 in Brussels (capital) and 1 in Wallonia. In 2010 a Royal Decree (23) stated that continuity of care should be available for in- and out-patients with serious illnesses (according to the 4 groups defined by the Royal College of Paediatrics and Child Health in 1997) (24) and categorised into 3 different groups of care: curative care, palliative care and terminal care at end-of-life stage.

Standards for accreditation requirements include the affiliation to a hospital which treats a minimum of 50 new patients < 16 years of age per year suffering from a haemato-oncological or haematological non-oncological illness which requires complex care, such as a stem cell transplantation. Furthermore, the Royal Decree foresees an interdisciplinary team with 4,0 Full-time equivalent (FTE) nurses, 0,5 FTE psycho-social care worker, 0,5 FTE physician and 0,5 FTE administrative assistant. Tandem work is required with the so-called first line, considered a primary health care team including a GP and home care nurses.

The liaison teams were first financed by “Kom op tegen Kanker” (a cancer charity) and by private funding. Since 2006, they have been predominantly funded by the Belgian Federal Government of Health with funds that were initiated by action point 23 of the Cancer Plan (25). Since 2012 this action point is used to further develop and expand the function of a paediatric liaison team and its accompanied tasks. Financial funding is allocated according to the number of children cared for.

Based on the data collected through annual reports and addressed to the Belgian Federal Government of Health, this study aims to (i) describe the characteristics of children cared for by PLT, (ii) the different activities provided by PLT in order to document how continuity of care is ensured in Belgium. To the best of our knowledge, it is the first publication describing the characteristics of children cared for by the paediatric liaison care teams and the activities they deliver to families, offering important insights on the number and the characteristics of children accessing paediatric palliative care services in Belgium.

Methods

Study design

This study is a secondary analysis of the data provided by the 5 PLTs in the form of their annual reports and data collected by the federal government. Preliminary findings were presented to the PLTs and thereafter enriched by their nuanced interpretation of the results.

Data collection

Aggregated quantitative data and narrative responses to open-ended questions issued from the annual reports 2010-2014 were transmitted to the first author. See Table 2. The data had been extracted from an existing database and is collected by the Cancer Plan then stored at the Belgian Federal Government of Public Health which holds the results of standardised annual PLT activity reports from 2010 to 2014. Annual reports have been mandatory since 2010, the year of the Royal Decree pertaining to the role of PLTs. The head nurse of each PLT completed the annual activity reports and sent them to the Cancer Plan.

The variables required for annual reports are presented in Table 3 and include the number of children cared for, their pathologies, the length of follow-up, as well as the number and places of deaths (hospital or home).

Table 2. Variables for aggregated quantitative data and open-ended questions included in the annual activity reports of the paediatric liaison teams

Age range classification	0-1 Y, >1-10 Y, 11-20 Y, > 20 Y
Disease classification	Haematology/Oncology Neurology/Degenerative Genetic/Metabolic Cardiovascular Gastroenterology Nephrology Congenital abnormalities Immunological Respiratory Others (Neonatology, Transplantation)
Origin of referral	Origin of referral for new patients (same hospital/other hospital)
Classification of patients	Curative/palliative/end-of-life patients
Number of patients	Total patients followed-up per year
Death	Place of death (home, hospital, other) Total number of deaths Duration of follow-up Frequencies of admission in hospital Mean average duration of admission Number of contacts with families' post-death
Objectives and needs	What objectives did you have? Which objectives did you achieve? Which objectives did you partially achieve? Why? What are your needs? Which objective(s) did you not achieve? Why? What are your objectives for next year? Did your team change during this year?
Strengths and weaknesses	What are the strengths of your team What are the weaknesses of your team?
Needs assessment	What are the requests of the target group? What are the requests of the PLT? What are the requests of services/institutions/hospitals?

Data analysis

Firstly, the characteristics of the children cared for by the 5 PLT over a five-year period, were summarised, using descriptive statistics.

Secondly, in order to identify and describe the various activities carried out by the PLT as part of their work, responses to the open-ended questions included in the annual reports were analysed by the first author inductively, ie. without any preconceived idea or hypothesis, to identify the themes that would best describe the range of activities undertaken by the PLTs (26).

Finally, the relevance and accuracy of the themes derived from the analysis performed by the first author were discussed with the representatives of the PLTs, as well as the Cancer Plan. The latter was consulted during a formal expert review panel meeting, held on 12th April 2016, which included 4 paediatric liaison nurses (CT,GH,FV,NA); 2 specialist physicians supervising the liaison teams (MR, CF); 1 social worker (IR) acting as a coordinator of a PLT and 2 researchers (IA, MF).

During this meeting, six themes were validated as describing the range of activities undertaken by the PLTs: coordination; communication; care; fundraising; training; quality improvement and research.

Moreover, several challenges encountered by PLT emerged in the annual reports and were discussed during the meeting. Those specific challenges will be presented in the results section.

The results presented hereafter, are therefore the expression of a collaborative approach of data analysis and interpretation, which involved the main stakeholders with the aim to collectively engage in a process of reflecting on past, present and future challenges to improve continuity of care in PPC.

Results

What are the characteristics of the children followed?

Based on the aggregated data from the annual reports for the period 2010-2014, a total of 3,607 children in Belgium benefitted from a follow-up of one of the five PLT (average of 721 children per year). Among all the children followed, the proportion of palliative patients was 25% (n= 910). The mean duration of follow-up was 226 days per child. Of the 3607 children, 47% were >1 and <10 of age, 25% were < 1 year, 25% were > 11 and <20 years and 3 % were >20 years.

Fifty percent suffered from an oncological pathology, 27% from neurological, genetic abnormalities, metabolic or degenerative diseases and 23% consisted of other diseases (including cardiovascular, immunological, gastroenterological, respiratory, nephrological or perinatal diseases).

Four hundred and twenty-eight children (12%) died during the study period. 51% percent of these children died at home, 47 % died in hospital and 2 % died in respite care services or institutions for disabled children. Each year, more than 40 home deaths were coordinated by PLTs across Belgium.

The characteristics of the children followed from 2010 to 2014 by the PLT are presented in Table 3.

Table 3. Characteristics of the children followed from 2010 to 2014 by the paediatric liaison teams (n = 3607)

Characteristics of children	**Number of children (%)**	**Mean per year/ per team** (min/max)	**Median per year/ per team** (P25/P75)
Age of children			
0-1 y	885 (25%)		
1-10 y	1689 (47%)		
11-20 y	879 (24%)		
>20 y	116 (3%)		
	Total: 3607 (100%)	144 (43-240)	144 (103-196)
Pathologies			
Haem-Onc disease	1816 (50%)		
Neuro-degenerativ/metabolic	989 (27%)		
Other (neonat/gastro/resp)	849 (23%)		
	Total: 3654 (100%)		
Curative/Palliative Patients			
Curative patients	2540 (70%)	102 (1-181)	105 (82-135)
Palliative patients	910 (25%)	36 (10-86)	23 (20-58)
Undefined	157 (5%)		
	Total: 3607 (100%)		
Referral of new patients from originating hospital	1848 (51%)		
External hospital	108 (3%)		
Total new patients	2056 (54%)		
	Total: 3607 (100%)		
Length follow-up **(children who died)**		226 days/child (1-4612 days)	32 days/child (21-201 days)
<120 days	272 (64%)		
121-240 days	65 (15%)		
241-400 days	31 (7%)		
>401 days	60 (14%)		
	Total: 428 (100%)		
Location of death			
Hospital	200 (47%)	8 (2-22)	6 (6-10)
Home	221 (51%)	9 (1-22)	9 (3-16)
Others (respite care home, others)	7 (2%)		
	Total: 428 (100%)	18 (3-32)	19 (12-26)

What are the main activities of PLT?

The thematic analysis of the answers given by each PLT to the open questions in the annual activity reports, enriched through discussions with the review panel, resulted in the identification of 6 main types of activities, which are presented hereafter with quotes from the annual reports (AR) or from the review panel meeting (RP).

Coordination

Coordination between home care services, hospital services, schools and respite care services was reported as an important activity. Early referral to a PLT allows the team to build relationships with the families. The referring hospital-based-specialist physician continues to be responsible for the care of the patient until death. Interdisciplinary meetings within hospital units and out-patient services are initiated and coordinated by PLTs.

In order to enhance coordination reports of home-visits and interdisciplinary meetings are stored as electronic records. The phone calls made are recorded in "liaison notebooks" which are kept at the home. These notebooks can be used by the child, family members and caregivers to write down their observations.

Several difficulties in coordination were identified by PLT. These are linked to the absence of a social worker or a permanent coordinator, but also sometimes to the lack of a general practitioner identified by the family, which is a condition to the initiation of PPC home care.

PLTs express "that the first line is established better in Flanders (northern Flemish speaking-part of Belgium) facilitating therefore the identification of a GP and home care nurses, which seems occasionally, to be a problem in Wallonia (south French-speaking part of Belgium)." (RP)

One team has created the role of a "coordinator of information", who is based permanently in the hospital to share the information among all health professionals, both for in-hospital and out-patients, and with families. "This new function represents a valuable support for PLT." (RP)

Some PLT developed not only PPC home-based care, but also in-hospital PPC consultations, depending on institutional support and sometimes person-related factors.

"It depends on certain individuals in the hospital whether or not your PLT is recognised. This has consequences for early referral. But I must say; now it's better than before. The other in-hospital services know who we are and what we do." (RP)

Communication

Raising awareness among hospital teams, home care services and general society is achieved through brochures, conferences and fundraising, because PPC is still perceived as being limited to end-of-life care. The term "palliative" frightens the families and may represent an obstacle to accessing palliative care services.

“Sometimes the families do not even know that we are the paediatric palliative care team. We are called liaison teams and introduced sometimes as a supportive team by the physicians.(…)The word ‘palliative’ frightens families.(…) Some families consider us as ‘the angel of death’, even though we know very well that palliative care is not restricted to end-of life care.(…) “Right from the beginning, we should explain clearly, what the term palliative care means and repeat it as often as necessary!”(RP)

Teams express the urgent need to represent and advocate for PPC through active membership at federal commissions and associations for palliative care.

“I have been involved in the Palliative Care Federation of Flanders as an administrator since 2015 and am also the delegate in the Cell of Federal Assessment of Palliative Care (first meeting in 2015).” (AR)

Care

PLTs provide complex individualised, patient-centred curative and palliative care at home or in hospital settings, when home based care teams are not available or not trained enough. Families can call and receive home visits at any time (available 7 days/24 hours). PLTs have observed progressively more complex chronic conditions in children resulting in increasingly higher burdens for the family, who in turn require additional social assistance, financial resources, support for administrative procedures and in-home respite care services.

Coordination for complex procedures such as chemotherapy, blood transfusion, assisted ventilation, feeding through gastrostomy, tracheotomy care, patient controlled analgesia and intravenous sedation, requires constant professional training.

« Liaison teams are small teams where patient care is the priority; the patient population is very diverse often with very complex issues. Delivering care takes time and is very intensive.» (AR)

Care continues even after a child’s death. For each family, who wished it, bereavement care was offered via telephone, home visits or letters up to one year after the child’s death. In 2014, for example 672 contacts were established for 99 families who had lost their children (mean of 7 contacts/family).

Fundraising

Concerns regarding lack of resources were repeatedly expressed by the PLTs. Lack of trained PPC nurses in first-line home care services is also reported. One team states they do not even have an office. For other teams, a full complement of staff members to be able to ensure bereavement care, psychological support for siblings or a supervision of PLT is only possible thanks to private donations.

Monthly, mandatory supervision sessions are offered via an external psychologist. All team members can suggest some themes they want to discuss. Usually, debriefing is provided for challenging clinical situations. The main objectives of these sessions are to promote quality communication, to prevent compassion fatigue and assist team members to process vicarious trauma they may experience.

Lack of adequate governmental financing was repeatedly mentioned as obliging teams in time-consuming activities to find additional private funding to cover the total operating costs of the team. Including a social worker and a coordinator in the team was particularly emphasised.

“Strengthening the team by the presence of a social worker (halftime) would free up time spent on many serious social tasks.” (AR)

“A special fundraising was undertaken for the provision of a vehicle.” (AR)

“We would like to offer regular bereavement care for siblings, but we lack funding. It’s a pity, because siblings are really overlooked.” (RP)

“We continue to seek additional help (when lack of personnel) through our own members’ networks.” (AR)

Training

Continuous training for the team itself regarding pain control, paediatric palliative care, and complex care techniques such as intravenous chemotherapy or blood transfusion is carried out. When a new nurse enters the PLT, a long training period is offered for peer training; integration of care delivery; organisational elements of services; awareness and knowledge of key contacts. Thus acquiring competences in PPC needs time due to the complexity of the interventions provided.

Teams also ensure specific child and family counselling, the education of first line home care services who are not always specialised in paediatrics (for e.g. handling a gastrostomy, patient controlled analgesia pump, insertion of a nasogastric tube) as well as various professionals in hospital settings. Tailored support for schools around issues related to bereavement is also provided. Many PLT members are offering PPC education in nursing schools, faculties of medicine or hospitals.

“We should implement or reinforce training in paediatric palliative care for physicians and nurses of the 1st line.” (AR)

Quality improvement and research

Teams are aware of the importance of quality improvement, and are keen to establish workload indicators and patient-reported outcome measures (PROMS). Caring for patients limits the teams’ ability to engage in quality improvement and research activities. This was a source of frustration for health professionals.

"These projects are carried out with a minimum of resources and staff and rely heavily on volunteers although our Government imposes these indicators and record keeping. The same sector attaches great importance to the quality indicators and recording because it improves the professionalisation of care and the development of care is factual.” (AR)

Protocols on several themes are elaborated in different ways in each PLT. Some create flow-charts to determine feasibility of PPC home care, others create protocols on how to respond to a euthanasia request, should it occur, or how to implement Advance Care Planning tools.

“Palliative care, but especially early advanced care planning should be implemented in a better and more structurally sound way in paediatric services. Advance care planning is very important in the care pathways for children with chronic conditions. This would fight therapeutic obstinacy; place care on a different and broader perspective, and allow parents / families to participate in the implementation of care goals. In paediatric palliative care, advanced care planning is an essential part of the care for children with chronic complex diseases. This still must be implemented. “(AR)

What are the specific challenges faced by PLTs?

In addition, during the review panel held in April 2016, the representatives of the 5 PLTs asserted that the workload of their teams was not truly reflected through the data reported in the annual reports. PLT’s highlighted 3 difficulties. These involved i) the way of reporting on cases, ii) the criteria to define when a palliative stage starts and iii) when to end a follow-up.

Firstly, with regards to reporting on cases, children followed by the PLTs present, over time, more complex chronic conditions, defined as medical conditions that can last at least 12 months and involve either several different organ systems or one organ system severely enough to require specialty paediatric care and probably a period of hospitalisation in a tertiary care center. (27) The PLTs expressed their difficulty to identify the principal disease of the child and in which category or categories of illness it should be reported.

Secondly, PLTS expressed difficulty in distinguishing patients requiring palliative care: “When curative treatment is no longer an option», «when death approaches”, “when the illness isn’t curable anymore and will lead to a premature death. But this does not mean that the patient doesn’t receive an illness-directed therapy. It has already happened that a patient labelled “palliative” shifted to “curative” because an experimental therapy succeeded. But for patients with neurometabolic illnesses, with a slow disease progression, the patient arrives at a palliative stage later on, when his quality of life deteriorates. It’s very different.”

This difficulty expressed by PLT to define when palliative care starts and actually stated in the annual reports, is significant and could be explained by the principle role played by PLT in being a seamless service starting at the time of diagnosis and offering a progressive step by step provision of PPC. Further, a distinction between patients receiving palliative versus curative care did not affect service provision. This discussion led to the reflection on whether a formal distinction between those two stages is required.

Finally, the review panel expressed difficulty in determining the criteria by which a PLT should decide to end a follow-up. The duration of follow-up for patients can be difficult to determine, particularly when children become more stable and is variable depending on type and severity of disease, and family specific needs.

Discussion

The results obtained in this national study showed that PLTs ensure continuity of care through a number of complex and complementary activities. More specifically, several results demonstrate how continuity of care is ensured, namely the long follow-up period (average of 7 months); starting at the time of diagnosis, often at a curative stage; the high number of home deaths; and the capacity to offer bereavement care to the family. Moreover, the difficulty for PLTs, to distinguish between curative and palliative stages, might indicate that those conceptual distinctions are not relevant to ensure continuity of care, whatever the child's care pathway might be.

While other studies have reported a higher proportion of neurological diseases (28,29,30), the large quantity of oncology cases in our study can be explained by several reasons. First of all, this is linked to the historical roots of the PLTs in Belgium that originated in oncology wards. Secondly the accreditation criteria defined by the Royal Decree, focusses on specialised oncology centres. Finally, the origin of funding for this programme, namely the Cancer Plan, likely influences the target population.

Regarding activities of the PLTs, our findings revealed a high number of home deaths, which requires an optimal degree of coordination among home care workers and hospital-based professionals. It is commonly assumed that home would be the best place to care for a child at end-of-life (31), but a study from Bluebond-Langner questions this assumption (32). According to her, better outcome measures would be to assess the possibility for children and parents to have their preferences considered. This is the case for PLTs who offer families not only the possibility to express the location to have their child cared for at end-of-life (at home or at hospital), but also to rapidly adapt the place if preferences change over time.

Continuity of care such as provided by PLTs might be positively influenced through policies. In Belgium, considered among other European countries as offering optimal PPC provision, a bill voted in 2016 has enhanced the smooth integration of PPC within the national health system by enlarging the definition of palliative care beyond end-of-life care.

This is in line with the results of a systematic review conducted by Knapp et al. on PPC provision worldwide. Of the 43 European countries surveyed, 24 had some services in PPC. Belgium was one of only 5 countries providing the highest level of PPC services (level 4: measure of integration with mainstream services provided). (34)

Nevertheless, Noyes et al. demonstrate several potential barriers to accessing PPC services. Among these, cultural beliefs are highlighted as lowering the use of specialised PPC services. (35)

Social acceptance of palliative care might influence optimal early access to specialised palliative care services and therefore continuity of care. One example is the terms used in the team's name, which seems to matter (36). Fear of the term 'palliative' can be explained by common social representation's associating palliative care with end-of-life care, giving the impression that death is near. It has been reported that families call PPC teams "the angel of death".

Therefore, the term “paediatric liaison teams”, instead of PPC teams, adopted by the Federal Government through the Royal Decree (23) seems to be more appropriate and efficacious to ensure acceptability of those services among families. Some solutions around this include for the team better communicating to the family what palliative care is and the use of the term Liaison as an alternative.

PLTs provide humane and professional support to families throughout the journey of a child’s illness and death. Quality of relationship is a core element of care, whether for curative or palliative care. Characteristics of this relationship, essential to ensure continuity of care, were found in several theoretical frameworks presented hereafter and linked to the PLT’s activities.

Although Harding et al. highlight the lack of an ideal PPC model (37), the pop-up model (38) suggests a shared care model of palliative care, where primary health services and specialist PPC providers collaborate. This is the case for the Belgian PLTs, considered mostly as a second line service, which aim is to assist and support the primary health care team (GP and home care nurses). The pop-up model underlines the necessity to provide flexible care.

This coordination between specialists and primary health services can be found in community-based PPC such as reported by Kaye et al. (16). According to Goldhagen et al. this model was even found to improve health-related quality of life and to reduce hospitalisation utilisation and costs (39).

The elements of continuity of care described by Haggerty (5), can be achieved through nurse-driven care activities (40-42), which are demonstrated by the PLTs studied here. Moreover, continuity of care can be rooted in the fundamentals of humanistic nursing theories applied to palliative care (43-45), which are focused on individualised, holistic and family-centred care.

Those components can by summarised in the paradigm of integrated care suggested by Milstein (46), which advocates the development of professional attitudes of "being with" families throughout the child's disease trajectory, both in curative and palliative stages. This constant presence of the team alongside the family can be seen as a form of companionship, and is an important resource of healing. PLTs report that parents express the valuable presence of teams being there for them “day after day”.

This is supported by Weaver et al. (47) and Moonley et al. (48), who strengthen the role of relationships between parents and health care professionals in order to help families to cope with the uncertainty and adversity of their child’s illness, leading to mixed, sometimes dichotomous feelings such as joy and sadness, hope and despair as reported by Rallison (49).

Furthermore, Carter introduces the concept of liminality which is an expression used in anthropology. Liminality refers to the intimate parental experience being nowhere, in a place where usual landmarks are becoming evanescent (50). This can be the case for parents experiencing isolation when caring for their ill child. Through home visits, active listening and coordination of care, PLTs aim to reach parents where they are, in order for them to feel less isolated and help them reconnect to social life.

Several authors (51-52) have extrapolated the concept of holding developed by the psychoanalyst D.W. Winnicott (53) as the essence of palliative care, arguing the importance of offering a secure and compassionate environment. At the same time paternalistic or judgmental attitudes should be avoided. Such support is essential for families who may be both vulnerable and isolated. Transposing Winnicott's concept of the 'good enough mother' to the 'good enough paediatric liaison team' could offer new perspectives of developing clinical guidelines which best ensure continuity of care.

Following on from the results of and challenges highlighted in our study, several developments of PPC in Belgium are presented hereafter.

Perspectives

Further discussions and workgroups on how to implement valid indicators of activity, identify children's palliative stages, and record diseases according to ICD-10 classification are in preparation among the PLT, the Cancer Plan and several authors of this manuscript. This could improve the quality of reporting, align the data collected to international research (54,55) and strengthen the legitimacy and sustainability of those specialised paediatric liaison teams.

In conjunction with this, constructive national guidelines on the organisation of PPC are currently being prepared by the Belgian Paediatric Palliative Care Group (BPPC) (56). Started in 2014, the BPPC assembles about 70 actors of PPC in Belgium from different professional backgrounds. The BPPC aims to elaborate guidelines on 5 themes: pain and symptom control, definition of paediatric palliative care, shared decision-making, organization of care, ethics and end-of life care.

It is presumed that better reporting of activities and developing guidelines might partially improve the quality of PPC. Promoting quality in PPC is a potential area of research (57). This is in line with the EAPC recommendation on outcome measures in palliative care and quality indicators which are currently restricted to studies on adults (58,59).

Belgian PLTs express strong interest for implementing tools to identify eligible children for PPC, to explore the individual quality of life of children benefitting from PPC, and to measure the impact of care provided. The relevance and feasibility of using several instruments, such as the Paediatric Palliative Screening Scale (PaPas Scale) (60), the direct weighted Scheduled Evaluation of Individual Quality of Life (SEI-QoL-dw) (61) or the Children's Palliative Outcome Scale (c-POS) was discussed (62) with paediatric liaison teams.

Limitations

To our knowledge, this is the first study describing the paediatric patients followed in Belgium by the specialised PPC teams, called liaison teams. The activities of these teams, including collaborative practice, are summarised to give a national snapshot of activity of PPC over a 5 year period.

However, the main limitation of the study lies in the fact that access to exclusively aggregated data did not permit further statistical analysis. Furthermore, some definitions in relation to curative and palliative care, reporting of patients with more than one diagnosis, and criteria for when follow-up might be concluded may assist in reducing disparities in reporting between different teams.

Conclusion

This secondary data analysis shows that Belgian PLTs ensure continuity of care through personalised, integrated care which starts at the time of diagnosis and continues, after a child's death, through bereavement care. The provision of contact with a PLT in all care settings gives the opportunity to build trusted relationship, which might reduce the important burden of a life-limiting condition on the entire family's life.

Nevertheless, improvements in data collection, development of national guidelines in PPC and implementation of specific outcome measures in routine clinical care are areas that could be further developed and described in future years.

Ethics approval

Since this study used secondary aggregated data with no personal information that could be identified, a request for ethical approval was not required. However, the necessary permission from the Belgian Federal Government of Public Health who provided the data for this study was obtained. A collaborative approach throughout the study gave the opportunity to the main stakeholders to discuss and interpret the data collected.

Acknowledgments

We wish to thank Dr Saskia Van den Bogaert and Mrs Flore Alix from the Cancer Plan at the Federal government of Public Health for the trust they showed me which was manifested in extraction and delivery of the aggregated data issued from the PLT annual reports; Charline Martens de Noordhout and Tharcisse Nkunzimana for their help in statistics; Shelagh Mc Connell, Jenny Hunt and Claudine Pauwels for their long-lasting moral support; Alison Parnell, Shaheen Shaw, Elizabeth Mc Donald, Mandana Mehta and Hannelore Andries for the proofreading of this article; Pr. Yves Benoît for his courage to initiate the first PPC home care team in Gent; Sonja Develter, for her perseverance to promote PPC in Belgium and Anne-Catherine Dubois for her engagement in developing paediatric respite care.

References

1.Abu-Saad Huijer H, Benini F, Craig F, H. Kuttner L, Wood C, Feraris P, Zernikow B. IMPaCCT. Standards for paediatric palliative care in Europe. Eur J Palliat Care. 2007; 14:2–7.

2. Connor SR, Downing J, Marston J. Estimating the Global Need for Palliative Care for Children: A Cross-sectional Analysis. J Pain Symptom Manage. 2017 Feb;53(2):171-177. doi: 10.1016/j.jpainsymman.2016.08.020. Epub 2016 Oct 17.

3. Gethins M. Paediatric Palliative Care in Europe Expands. JNCI, 104(1) 2012: 10-11.

4.Miller A, Condin C, Mc Kellin W, Shaw N, Klasse A, Sheps S. Continuity of care for children with complex chronic health conditions: parent's perspectives. BMC Health Serv Res. 2009; 21; 9:242.

5.Haggerty J, Reid R, Freeman G, Starfield B, Adair C, McKendry R. Continuity of care: a multidisciplinary review. BMJ. 2003;327(7425):1219-21.

6.Freeman G, Shepperd S, Robinson I, Ehrich K, Richards S. Continuity of Care: Report of a scoping Exercise for the NCCSDO. National coordinating Centre for Service Delivery and Organisation, London. 2001.

7.Freeman G, Woloshynowych M, Baker R, Boulton M, Guthrie B, Car J, Haggerty J, Tarrant C et al. Continuity of care 2006: What have we learned since 2000 and what are policy Imperatives now? National Coordinating Centre for Service Delivery and Organization, London. 2007.

8.Parker G, Corden A, Heaton J. Experiences of and influences on continuity of care for service users and carers: synthesis of evidence from a research programme. Health Soc Care Community. 2011;19(6):577.

9.Saultz JW. Defining and Measuring Interpersonal Continuity of Care. Ann Fam Med. 2003;1(3): 134–43.

10.Foster T, Gilmer M. Continuing bonds: a human response within paediatric palliative care. Int J Palliat Nurs. 2008;14(2):85-91.

11.Mastro K, Johnson J, McElvery N, Preuster C. The Benefits of a Nurse-Driven, Patient- and Family-Centered Paediatric Palliative Care Program. J Nurs Adm. 2015 Sep;45(9):423-8.

12.Johnson S, Giesie P, Ireland A, Rice R, Thomson B. On the Scene: Developing a Nurse Care Coordinator Role at City of Hope. Nurs Adm Q. 2016 Jan-Mar;40(1):39-50.

13.World Health Organization. WHO Definition of Palliative Care for Children. www.who.int/cancer/palliative/definition/en. Accessed 10 Feb 2018.

14.World Health Assembly (WHA), Strengthening of palliative care as a component of integrated treatment within the continuum of care. 134th session EB134.R7.

15.Kaye E, Friebert S, Baker J. Early Integration of Palliative Care for Children with High-Risk Cancer and Their Families. Pediatr Blood Cancer. 2016;63(4):593-7.

16.Kaye E, Rubenstein J, Levine D, Baker J, Dabbs D, Friebert S. Paediatric Palliative care in the community. CA Cancer J Clin. 2015;65(4):316-33.

17.Miller E, Levy C, Linebarger J, Klick J, Carter B. Paediatric Palliative Care : Current evidence and evidence gaps. J Pediatr. 2015;166(6):1536-40.

18.Lichtenthal W, Sweeney C, Roberts K, Corner G, Donovan L, Prigerson H, Wiener L. Bereavement follow-up after the death of a child as a standard of care in paediatric oncology. Pediatr Blood Cancer. 2015;62(5):S834-69.

19.Mastro K, Johnson J, McElvery N, Preuster C. The benefits of a nurse-driven, patient- and family-centered Paediatric Palliative Care Program. J Nurs Adm. 2015;45(9):423-8.

20. Weaver M, Heinze K, Kelly K, Wiener L, Casey R, Bell C, Wolfe J, Garee A, Watson A, Hinds P. Palliative care as standard in Paediatric Oncology, Pediatr Blood Cancer. 2015;62(5): S829-33.

21. Niswander L, Cromwell P, Chirico J, Gupton A, Korones D. End-of-life Care for children enrolled in a community-based Paediatric Palliative Care Program. J Palliat Med. 2014;17(5):589-91.

22. Ministry of Justice. Loi du 14 juin 2002 relative aux soins palliatifs in http://www.ejustice.just.fgov.be/cgi_loi/change_lg.pl?language=fr&la=F&cn=2002061446&table_name=loi. Accessed 10 Feb 2018.

23. Royal Decree of the 15th November 2010 which defines the norms on which the function of the paediatric liaison must respond in order to be accredited in http://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html. Accessed 10 Feb 2018.

24. Association for Children with Life Threatening or Terminal conditions and their families (ACT) and the Royal College of Paediatrics and Child Health (RCPCH). A guide to the development of children’s palliative care services. UK. 1997.

25. Action 23 of Cancer Plan in http://www.health.belgium.be/sites/default/files/uploads/fields/fpshealth_theme_file/actions.pdf. Accessed 10 Feb 2018.

26. Marks D, Yardley L. Content and thematic analysis, in: Research Methods for Clinical and Health Psychology, SAGE, 2004.

27. Feudtner C. Christakis DA, Connell FA. Paediatrics. 2000.106(1):205-9.

28. Palliative care for infants, children and young people. The Facts. EAPC Taskforce for Palliative Care in Children and Maruzza Lefebvre d’Ovidio Onlus Foundation. 2009. In http://www.palliative.lv/wp-content/uploads/2013/01/The_Fact.pdf. Accessed 10 Feb 2018.

29. Fraser LK, Miller M, Hain R, Norman P, Aldridge J, McKinney PA, Parslow RC. Rising national prevalence of life-limiting conditions in children in England. Paediatrics. 2012 Apr;129(4):e923-9. doi: 10.1542/peds.2011-2846. Epub 2012 Mar 12.

30. Fraser LK, Lidstone V, Miller M, Aldridge J, Norman P, McKinney PA, Parslow RC. Patterns of diagnoses among children and young adults with life-limiting conditions: A secondary analysis of a national dataset. Palliat Med. 2014 Jun;28(6):513-520. doi: 10.1177/0269216314528743.

31. Kassam A, Skiadaresis J, Alexander S, Wolfe J. Parent and clinician preferences for location of end-of-life care: home, hospital or freestanding hospice? Pediatr Blood Cancer. 2014;61(5):859-64.

32. Bluebond-Langner M, Beecham E, Candy B, Langner R, Jones L. Preferred place of death for children and young people with life-limiting and life-threatening conditions: a systematic review of the literature and recommendations for future inquiry and policy. Pall Med. 2013;27:705-13.

33. Ministry of Justice. Loi du 21 juillet 2016 modifiant la loi du 14 juin 2002 relative aux soins

palliatifs en vue d’élargir la définition des soins palliatifs in

http://www.ejustice.just.fgov.be/cgi_loi/loi_a.pl. Accessed 10 Feb 2018.

34. Knapp C, Woodworth L, Wright M, Downing J, Drake R, Fowler-Kerry S, Hain R, Marston J. Paediatric palliative care provision around the world: a systematic review. Pediatr Blood Cancer. 2011 Sep;57(3):361-8. doi: 10.1002/pbc.23100.

35. Noyes J, Edwards RT, Hastings RP, Hain R, Totsika V, Bennett V, Hobson L, Davies GR, Humphreys C, Devins M, Spencer LH, Lewis M. Evidence-based planning and costing palliative care services for children: novel multi-method epidemiological and economic exemplar. BMC Palliat Care. 2013 Apr 25;12(1):18. doi: 10.1186/1472-684X-12-18.

36. Maciasz R, Arnold R, Chu E. Does it matter what you call it? A randomized trial of language used to describe palliative care services. Support Care Cancer. 2013;21:3411.

37. Harding R, Albertyn R, Sherr L, Gwyther L. Paediatric palliative care in sub-saharan Africa: a systematic review of the evidence for care models, interventions, and outcomes. J Pain Symptom Manage. 2014;47(3):642-51.

38. Mherekumombe MF, Frost J, Hanson S, Shepherd E, Collins J. Pop Up: A new model of paediatric palliative care. J Paediatr Child Health. 2016 Sep 9. doi: 10.1111/jpc.13276.

39. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based paediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. BMC Palliat Care. 2016 Aug 3;15:73. doi: 10.1186/s12904-016-0138-z.

40. Pontin D, Lewis M. Maintaining the continuity of care in community children's nursing caseloads in a service for children with life-limiting , life-threatening or chronic health conditions : a qualitative analysis. J Clin Nurs. 2009;18(8):1199-206.

41. Johnson S, Giesie P, Ireland A, Rice R, Thomson B. On the scene a Nurse care Coordinator Role at City of Hope. Nurs Adm Q 2016;40(1):39-50.

42. Jankowski J. Professional boundaries issues in Paediatric Palliative Care. Am J Hosp Palliat Care. 2014;31(2):161-5.

43. Wu HL, Volker DL. Humanistic Nursing Theory: application to hospice and palliative care. J Adv Nurs. 2012;68(2):471-9.

44.Dobrina R, Tenze M, Palese A. An overview of hospice and palliative care nursing models and theories. Int J Palliat Nurs. 2014;20(2):75-81.

45. Magante MC. Developing a care model for hospice and palliative nurses. Int J Palliat Nurs. 2015;21(2):96.

46. Milstein J. A paradigm of integrative care: healing with curing throughout life, "being with" and "doing to". J Perinatol. 2005;25(9):563-8.

47. Weaver MS, Heinze KE, Bell CJ, Wiener L, Garee AM, Kelly KP, Casey RL, Watson A, Hinds PS. Paediatric Palliative Care Special Interest Group at Children's National Health System. Establishing psychosocial palliative care standards for children and adolescents with cancer and their families: An integrative review. Palliat Med. 2016 Mar;30(3):212-23.

48. Mooney-Doyle K, Dos Santos MR, Szylit R, Deatrick JA. Parental expectations of support from healthcare providers during paediatric life-threatening illness: A secondary, qualitative analysis. J Pediatr Nurs. 2017;36:163-172.

49. Rallison LB, Raffin-Bouchal S. Living in the in-between: families caring for a child with a progressive neurodegenerative illness. Qual Health Res. 2013;23(2):194-206.

50. Carter B. Liminality in Paediatric Palliative Care. Am J Hosp Palliat Care. 2017;34(4):297-300.

51. Bioy A, Van Lander A, Mallet D, Belloir M-N (dir.) Soins palliatifs. 9th part. 40th Chapt. Dunod 2017:265.

52. Duflot C. L'art thérapie en soins palliatifs. Eres. 2016.

53. Winnicott DW, Winnicott on the Child. Cambridge 2002:238.

54. Feudtner C, Feinstein J, Zhong W, Hall M, Dai D. Paediatric complex chronic conditions (CCC) classification system version 2 : updated for ICD-10 and complex medical technology dependence and transplantation. BMC Pediatr 2014;14:199.

55. Hain R, Devins M, Hastings R, Noyes J. Paediatric palliative care: Development and pilot study of a "Directory of life-limiting conditions". BMC Palliat Care 2013;11;12(1):43

56.Renard M, Brichard B, Francotte N, Fonteyne C, Misson J-P, Faingnaert M, Mattheeuws K, Ruysseveldt I. Belgian Paediatric Palliative Care: Spreading the Ideas of Paediatric Palliative Care in the Different Fields of Paediatric Medicine. P050 3rd Congress on Paediatric Palliative Care. A global Gathering. Rome, 16-19 Nov 2016.

57. Kassam A, Skiadaresis J, Habib S, Alexander S, Wolfe J. Moving toward quality palliative cancer care: parent and clinician perspectives on gaps between what matters and what is accessible. J Clin Oncol. 2013;31(7):910-5.

58. Bausewein C, Daveson B, Currow D, Downing J, Deliens L et al. EAPC White Paper on outcome measurement in palliative care: Improving practice, attaining outcomes and delivering quality services - Recommendations from the European Association for Palliative Care (EAPC) Task Force on Outcome Measurement. Palliat Med. 2016;30(1):6-22.

59. Leemans K, Deliens L, Van den Block L, Vander Stichele R, Francke A, Cohen J. Systematic Quality Monitoring For Specialized Palliative Care Services: Development of a Minimal Set of Quality Indicators for Palliative Care Study (QPAC). Am J Hosp Palliat Care 2016 Apr 7. pii: 1049909116642174.

60. Bergstraesser E, Hain R, Pereira J. The development of an instrument that can identify children with palliative care needs: the Paediatric Palliative Screening Scale (PaPaS Scale): a qualitative study approach. BMC Palliat Care 2013; 12:20.

61. O'Boyle C, Browne J. Schedule for the Evaluation of Individual Quality of Life. A direct weighting procedure for quality of life domains. Administration Manual. Department of Psychology. Royal College of Surgeons in Ireland, 1995.

62. Harding R. Outcome measures for children: current state of science and views of professional stakeholders. Conference. August 2015. United Kingdom. In http://www.togetherforshortlives.org.uk/assets/0001/0536/Richard_Harding-Outcome_measures_for_children.pdf. Accessed 10 Feb 2018.

3.2. ACCESS TO PAEDIATRIC LIAISON CARE TEAMS MIGHT BE INSUFFICIENT IN BELGIUM

This chapter was published as an article under the title:

Friedel M, Gilson A, Bouckenaere D, Brichard B, Fonteyne C, Wojcik T, De Clercq E, Guillet A, Mahboub A, Lahaye M, Aujoulat I. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. BMJ Paediatr Open. 2019 Sep 29;3(1):e000547. doi: 10.1136/bmjpo-2019-000547. eCollection 2019.

ABSTRACT

Background: Paediatric complex chronic conditions (CCC) are life-limiting conditions requiring paediatric palliative care, which, in Belgium, is provided through paediatric liaison teams (PLTs). Like the number of children and adolescents with these conditions in Belgium, their referral to PLTs is unknown.

Objectives: The aim of the study was to identify, over a 5-year period (2010-2014) the number of children and adolescents (0-19 years) living with a CCC, and also their referral to PLTs.

Methods: International Classification of Disease codes (ICD-9) corresponding to a CCC, as described by Feudtner et al. (2001) and national registration numbers were extracted from the databases of all hospitals (n=8) and PLTs (n=2) based in the Brussels region. Aggregated data and pseudonymised national registration number were transmitted to the research team by a trusted Third Party (eHealth). Ages and diagnostic categories were calculated using descriptive statistics.

Results: Over 5 years (2010-2014), 22 721 children/adolescents aged 0-19 years were diagnosed with a CCC. Of this number, 22 533 were admitted to hospital and 572 were followed-up by a PLT. Of the 22 533 children/adolescents admitted to hospital, only 384 (1.7%) were referred to a PLT.

Conclusion: In Belgium there may be too few referrals of children and adolescents with CCC to PLTS that ensure continuity of care.

Introduction

A complex chronic condition (CCC) "can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either different organ systems or 1 organ system severely enough to require specialty paediatric care and probably some period of hospitalisation in a tertiary care centre." (1) A complex chronic condition might be life-limiting or life-threatening and might require paediatric palliative care at different stages (2). It is not easy to establish the number of children living with a life-limiting condition. (3) In the United Kingdom, Fraser et al. (4) found a prevalence of 3.2 % of children (0-19 years) living with such a condition.

Many other studies relied on death registries to estimate the number of children with CCCs. A Canadian study found that 26.8 % of the 4 199 children aged 0-19 years who died between 1997 and 2001 had had a CCC, and that 48% had died in the first 6 months of life (5). A quarter of the 11 194 children aged 1-19 years who died in France between 2005 et 2008 had a CCC. [6] A European multicenter study showed that a third of the 299 children aged 1-17 years who died in Belgium in 2008 (35.8%, n= 107) had had a CCC. (7)

Paediatric Palliative Care for children/adolescents (CA) in Belgium is provided through 5 paediatric liaison teams (PLTs), each of which is attached to a university hospital. The mission per centre is to ensure continuity of care through all care settings (in and out hospital) for children with life-limiting or life-threatening conditions, including CCCs. Funding of these PLTs has been integrated into the Belgian Health coverage since 2010. (8) In 2016, the legislation was modified to state that palliative care should not be restricted to end-of-life care but should be provided progressively according to patients' needs and wishes, independently of life expectancy. (9)

It is not known how many children and adolescents live with CCCs in Belgium, or what access they have to paediatric liaison care teams. Hypothesising that many children living with CCCs are not referred to a PLT, we wished to establish 1.) the number of CA (0-19 years) with CCCs and, 2.) the percentage referred to PLTs in the Belgium's Brussels Region, which has 8 hospitals with a paediatric unit, and 2 PLTs.

Methods

Design

This retrospective hospital-based study used existing health data collected from all the hospitals in Brussels with paediatric units (n=8) and from all the PLTs based in Brussels (n=2).

There were 3 reasons for choosing the 5-year study period, which started in 2010 and ended in 2014. First, when the official funding of paediatric liaison care teams started in 2010, data registration became mandatory. Second, in 2015, the ICD-9 classification changed in Belgium to ICD-10. Third, the only published study on the number of children referred to PLTs in Belgium was based on the same 5-year period. (10)

Patient Selection

For this study, we selected all CA (0-19 years) diagnosed with a CCC and admitted between 1 January 2010 and 31 December 2014 to one of the 8 hospitals in Brussels with a paediatric unit. Independently of their diagnosis, we also selected all children (0-19 years) referred to the two PLTs based in Brussels.

Data collection

We used the methods described by Feudtner et al (11) based on the International Classification of Disease ICD-9 codes (12) linked to CCC.

All ICD-9 codes corresponding to a CCC and linked to a national register number were extracted from existent hospital databases issued from a mandatory “minimal hospital synthesis” (*Résumé Hospitalier Minimum*; RHM), an anonymised registration system for administrative, medical and nursing data. (13) We created a standardised electronic file in Excel that included the variables and formulae requested, thereby maximising the homogeneity of the data collected by the data managers. These files were sent to the data managers of each of the 8 participating hospitals and to the 2 PLTs.

Anonymously, as dictated by the Sectoral Committee for Social Security and Health, the data managers at the 8 hospitals and 2 PLTs sent national registration numbers and aggregated data to a Trusted Third party (eHealth), (14) which pseudonymised the national registration numbers, and sent the aggregated data to the research team for analysis. Knowing the pseudonymised national registration numbers of each CA, we were able to compare databases from the hospitals with those from the PLTs. See Figure 4 and Table 4.

Figure 4. Data-flow process used for this study.

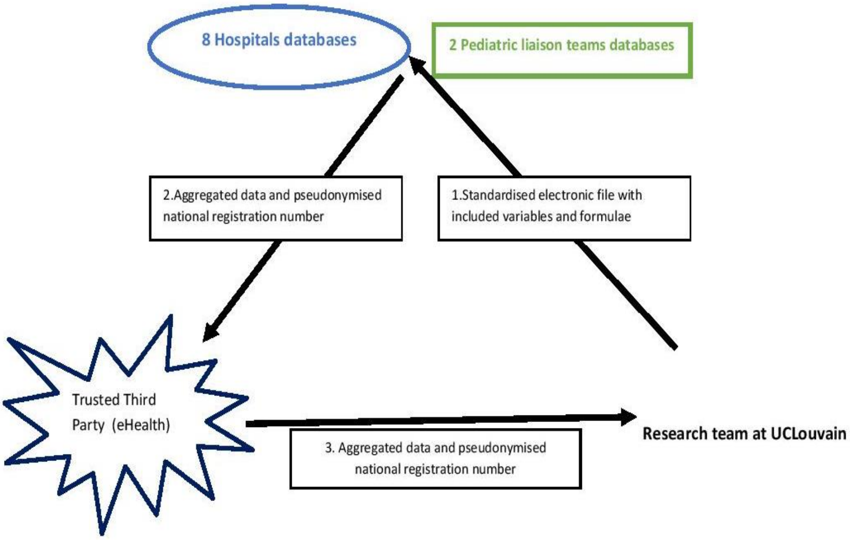


Table 4. Overview of the variables requested, received and the limitations imposed by the sectoral Committee for Social security and Health (CSSSS)

Variables requested of data managers	**Limitation imposed by CSSSS**	**Variables received by the research team**
National registration number	Pseudonymisation	Pseudonymised code
Principal and secondary diagnosis of a complex chronic condition	Principal diagnosis aggregated to 1 of the 9 categories; secondary diagnosis aggregated to 1 of the 9 categories	Cancer; digestive disease; cardiovascular disease; haematological/immunological disease; neurological disease; respiratory disease; urinary disease; congenital/genetic disease; metabolic disease
Dates of admission to and discharge from hospital	Day/month/year	day/month/year of admission day/month/year of discharge
Date of birth (day/month/year)	Aggregated to 6 categories	0-11 months, 1-4 years, 5-9 years, 10-15 years, 16-19 years, > 20 years
Gender	Male/female/unknown/changed	male/female
Postal code of residence	Aggregated to one of the 13 regions in Belgium	13 regions in Belgium; unknown
Nationality	Aggregated to one of the 6 regions defined by the WHO	6 regions
Insurance status	insured or not	insured or not

Data analysis

Figure 5. Flowchart of the process leading to the definitive database

Ages and diagnostic categories were calculated using descriptive statistics with program R software (version 3.3.2). Results were discussed in three meetings with the steering committee of the *Brussels Pluralist Federation for Palliative and Continuing Care,* consisted of DB, TW, BB, CF. (15)

Patient and Public Involvement

This study was commissioned by the Brussels Regional Platform for Palliative Care, called the Brussels Pluralist Federation for Palliative and Continuing Care (FBSP), who wanted to estimate the number of children and adolescents with complex chronic conditions and their referral to specialised paediatric palliative care teams called paediatric liaison teams. The FBSP (DB, TW, BB, CF) were involved in the design of the study and in the interpretation of the results. Dissemination of these results was initiated and coordinated by the FBSP, through a public conference addressed to paediatricians and a special public seminar on paediatric palliative care hold at the Brussels Francophone Parliament on the 6th of December 2018, at the invitation of Julie de Groote, president.

Results

Over the 5 year-period, a total of 22 721 children and adolescents aged 0-19 years with a CCC were identified through the registries of the hospitals (n=8) and PLTs (n=2) based in Brussels, Belgium. See Figure 5.

Of those, 22 149 children and adolescents aged 0-19 years with a CCC were admitted in 1 of the 8 hospitals but were not referred to a PLT, and only 384 (1.7%) children and adolescents were both admitted to a hospital and referred to a PLT.

Regarding geographical factors, we found that 40.3% (n= 9 081) of the CA with CCC admitted to one of the eight hospitals were resident in the Brussels region, whereas 45.2% (n=10 195) lived elsewhere. Data was unknown for 14.6% (n=3 257). Only 1.05% (n=95) of all children residing in Brussels and admitted to a hospital in Brussels were referred to one of the PLTs based in Brussels.

When we looked at the type of diseases found in the hospital databases (n=22 533), labelled with a CCC and over a 5-year period, our results showed that cardiovascular disease was the most common underlying condition (28%; n=6 310), followed by neurological conditions (18%; n= 4 125) and cancer (14%; n=3 011). These figures contrast with the type of diseases found in the PLT databases (n = 572), where diseases of the nervous system predominated (21%, n = 121), followed by cancer (17%, n = 98) and cardiovascular disease (10%, n = 60).

We also found the following: that nearly one third of the children were aged under one year (30% n = 6 692); and that 71% (n=276) of all registered hospital deaths (n=390; 0-19 years) involved children with cardiovascular diseases who had died before the age of one.

Of all children admitted to hospital (n=22 533) in this period, 87% had a European nationality (n=19 613); 7.5% an African nationality (n= 1 690) and 1.3% an Asian nationality (n=298). The nationality of 2.7% (n=600) was unknown.

Of all the children admitted to hospital with a CCC in this period, 6.3% (n=1 428) had no insurance and/or received public assistance (CPAS).

Table 5 shows the number of children/adolescents identified in hospital and paediatric liaison team databases from 2010 to 2014 according to gender, categories of age and disease, frequency of hospital admissions, and length of stay.

Table 5. Number of children/adolescents identified in the databased of hospitals and liaison teams

Gender	**Hospital % (n)**	**Paed liaison % (n)**
Female	44% (9 984)	46% (263)
Male	56% (12 549)	54% (309)
Total	**100% (22 533)**	**100% (572)**

Age categories (years)	**Hospital % (n)**	**Paed liaison % (n)**
0<1	30 % (6 692)	31% (177)
1-4	28 % (6 243)	31% (176)
5-9	18 % (3 978)	16% (94)
10-14	16 % (3 715)	17% (97)
15-19	8 % (1 905)	5% (28)
Total	**100% (22 533)**	**100% (572)**

Diseases	**Hospital % (n)**	**Paed liaison % (n)**
Cancer	14% (3 011)	17% (98)
Digestive disease	4% (848)	4% (24)
Cardiovascular disease	28% (6 310)	10% (60)
Haematological/immunological disease	6% (1 391)	5% (29)
Neurological disease	18% (4 125)	21% (121)
Respiratory disease	3% (752)	5% (26)
Urinary disease	7% (1 553)	2% (11)
Congenital/genetic disease	8% (1 810)	5% (28)
Metabolic disease	4% (972)	5% (27)
Unknown	8% (1 761)	26% (148)
Total	**100% (22 533)**	**100% (572)**

Deaths	**Hospital (n)**	**Hospital and Paed liaison (n)**
0-19 years	390	13
0-11 months	276	unknown
Total	**100% (22 533)**	**100% (384)**

Hospital admissions	**Hospital % (n)**	**Hospital and Paed liaison % (n)**
Frequency of hospital admissions (median, range, mean)	Once, 1-82 days, twice	3 times, 1-131 days, 6 times
Mean annual length of stay (median, range, mean)	5 days, 1-306 days, 12 days	20 days, 1-278 days, 33 days
	100% (22 533)	**100% (384)**

Discussion

We identified a large number of children with CCC who had been admitted to hospitals in Brussels during the study period (n=22 533). However, only a very small percentage (1.7%; n=384) of those had been referred to a PLT in Brussels during the same period.

The high proportion of children with CCC who are not referred to PLTs may be partially explained by the fact that many of these children die before they are discharged from hospital and that few of them are referred to a PLT. Furthermore, a previous study indicated that from all children followed-up annually by PLTs at a nation-wide level (n=721), a high proportion of home deaths (51%) occurred for all children who died (n=85) and therefore were neither registered in hospital databases. (10) Finally, all chronic complex conditions are not life-limiting conditions, which means that some of them will unlikely lead to death in childhood.

While the number of children with cardiovascular diseases is higher than in other studies (4,16), the other studies in question used a different definition of life-limiting and life-threatening conditions, and therefore different ICD-10 codes. The aggregation into diagnostic groups in the UK-based studies also featured many cardiovascular conditions in the congenital anomalies group. Moreover, 2 of the 8 hospitals included in our study are university hospitals known for their high number of highly specialised paediatric cardiological activities which could explain the high number of children suffering from cardiovascular conditions found in our results.

This study has three main limitations. First, according to ICD-9, which was published in 2001 on the basis of paediatric deaths, the rapid progress of medical therapeutics may be causing certain CCCs to evolve, which may therefore require fewer hospital admissions. Second, using the more recent Directory of life-limiting conditions (16) instead of the list of CCC may have provided more accurate results. Unfortunately, this Directory relies on ICD-10 codes, which was uncompatible with the registration system in use during the study period and based on ICD-9. Third, due to the obligation imposed on us to receive aggregated data – i.e. only the categories of diagnostics and not the accurate disease codes – we did not receive detailed information on the types of disease in the children included in this study. Number of deaths were inaccurately reported in the hospital registries. Finally, as many people were involved in data handling, human errors could not be ruled out.

Nevertheless, regarding the conceptual, methodological, logistic, ethical and legal complexities inherent to this study, our results provide useful data on the extent of the problem regarding the number of children with CCCs in Belgium. To our knowledge, this is the first multicentre study in Belgium to estimate the number of children and adolescents with a CCC and to identify the proportion who are followed up by a PLT. Having included all the 8 hospitals in Brussels (2 of which were university hospitals with over 900 beds) and 2 of the 5 national PLTs in Belgium, we believe that it will be possible to extrapolate our results to the national level.

Conclusion

Even though it is the mission of the 2 PLTs to ensure continuity of care between the hospital and the homes of children with a serious illness, we found that only a very small percentage of the 22 533 children with a chronic complex disease who were identified over a 5-year period (1.7%, n=384) admitted to a hospital were followed up by one of the city's 2 paediatric liaison teams. Our results suggest that the number of referrals of such children and adolescents to a paediatric liaison team in Belgium is probably insufficient or is reserved for the most complex situations.

Footnotes

Acknowledgments: We would like to thank the medical directors of the 8 hospitals who agreed to participate in this study: Centre hospitalier Interrégional Edith Cavell, Centre hospitalier universitaire Saint-Pierre, Clinique Saint-Jean, Cliniques universitaires Saint-Luc, Hôpital Érasme, Hôpitaux Iris Sud, Hôpital universitaire des Enfants Reine Fabiola, and Universitair Ziekenhuis Brussel. We are especially grateful to all the data managers at these hospitals for their support in extracting the data: G. Bouhy, K. Pien, M. Anthonis, G. Bockstal, N. Mbengo, C. Beguin, A. Kanfaoui, P. Kirkove. Similarly, we are very grateful for the collaboration not only of the chief nurses at the 2 paediatric liaison teams – G. Hendrijckx and N. Andersson at Globul'home; and C. Tonon at Interface pédiatrique – but also to N. Donnez at eHealth and A. Audibert at IREC-UCLouvain. Finally, we sincerely thank Prof. S. Moniotte for his discussion of the preliminary results of a number of children with cardiovascular conditions; Dr R. Hain for his comments on the interpretation of the results; and Dr Lorna Fraser for her constructive comments on the final manuscript.

Funding: We would like to thank Fondation Contre le Cancer, which funded this study through a grant to Fédération Bruxelloise pluraliste de Soins palliatifs et Continus.

Ethics approval: The study was conducted in accordance with Belgium's legal and ethical requirements for the collection of personal data. Approval was obtained on 29.2.2016 from the Data Protection Authority, and on 20.9.2016 from the Sectoral Committee for Social Security and Health (n° CSSSS/16/193). Additionally, the Sectoral Committee for Social Security and Health requested that minor cell-risk analysis be conducted by the Intermutualistic Agency; this took place on 17.8.2016. Finally, approval was granted by the ethical committees at each of the 8 participating hospitals.

References

1. Feudtner C, Christakis DA, Connell FA. Paediatric deaths attributable to complex chronic conditions: a population-based study of Washington state, 1980-1997. *Paediatrics* 2000; 106: 205-209.

2. Abu-Saad Huijer H, Benini F, Craig F, et al. Steering Committee of the EAPC task force on palliative care for children and adolescents. IMPaCCT. Standards for paediatric palliative care in Europe. *Eur J Palliat Care* 2017;14(3):109-114.

3. Connor SR, Downing J, Marston J. Estimating the Global Need for Palliative Care for Children: A Cross-sectional Analysis. *J Pain Symptom Manage* 2017; 53(2): 171-177.

4. Fraser LK, Miller M, Hain R et al. Rising national prevalence of life-limiting conditions in children in England. *Paediatrics* 2012; 129(4): e923-929.

5. Institut national de santé publique (INdSP). Soins palliatifs de fin de vie au Québec: Définition et mesure d'indicateurs. Partie 2: population pédiatrique (moins de 20 ans) https://www.inspq.qc.ca/pdf/publications/896_soinsPalPopPedFinal.pdf2008 (2008, accessed 5 Feb 2019)

6. Centre d'épidémiologie sur les causes médicales de décès CépiDC. INSERM 2008– issu du rapport 2011 de l'Observatoire national de la fin de vie : Fin de vie, un premier état des lieux. Chapitre 6: les soins palliatifs pédiatriques. http://www.onfv.org/wpcontent/uploads/2014/10/Rapport_ONFV_2011.pdf (2011, accessed 5 Feb 2019)

7. Håkanson C, Öhlén J, Kreicbergs U, et al. Place of death of children with complex chronic conditions: cross-national study of 11 countries. *Eur J Pediatr* 2017; 176(3): 327-335.

8. Belgian Ministry of Justice. Arrêté royal fixant les normes auxquelles la fonction "liaison pédiatrique" doit répondre pour être agréée. http://www.ejustice.just.fgov.be/cgi/article_body.pl?language=fr&caller=summary&pub_date=10-12-30&numac=20100244592010 (2010, accessed 5 Feb 2019)

9. Belgian Ministry of Justice. Loi du 21 juillet 2016 modifiant la loi du 14 juin 2002 relative aux soins palliatifs en vue d'élargir la définition des soins palliatifs. http://www.ejustice.just.fgov.be/cgi_loi/loi_a.pl?=&sql=(text+contains+(%27%27))&rech=1&language=fr&tri=dd+AS+RANK&numero=1&table_name=loi&F=&cn=2002061446&caller=image_a1&fromtab=loi&la=F&pdf_page=8&pdf_file=http://www.ejustice.just.fgov.be/mopdf/2016/08/29_1.pdf (2016, accessed 5 Feb 2019)

10. Friedel M, Brichard B, Fonteyne C, et al. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care 2018; 17(1): 77.

11.Feudtner C, Hays RM, Haynes G, et al. Deaths attributed to paediatric complex chronic conditions: national trends and implications for supportive care services. *Paediatrics* 2001 Jun;107(6): E99.

12. National Center for Health Statistics, International Classification of Diseases, Ninth Revision, Clinical Modification (ICD-9-CM) https://www.cdc.gov/nchs/icd/icd9cm.htm (accessed 5 Feb 2019)

13. Service Public Fédéral, Santé Publique, Sécurité de la chaîne alimentaire et environnement. https://www.health.belgium.be/fr/sante/organisation-des-soins-de-sante/hopitaux/systemes-denregistrement/rhm (accessed 5 February 2019)

14. Service Public Fédéral, Santé Publique, Sécurité de la chaîne alimentaire et environnement. Platform eHealth. https://www.ehealth.fgov.be/ehealthplatform/fr/mission (accessed 5 February 2019)

15. Brussels Pluralist Federation for Palliative and Continuing Care. https://www.fbsp-bfpz.org/ (accessed 5 February 2019)

16. Hain R, Devins M, Hastings R, et al. Paediatric palliative care: development and pilot study of a 'Directory' of life-limiting conditions. *BMC Palliat Care* 2013;11;12(1):43.

Key messages of Chapter 3

This third chapter described the activities of paediatric liaison teams and the profile of children followed-up by them. The original liaison model ensures continuity of care for children facing life-limiting condition, from curative to palliative stages and across all health care settings. Access to those teams is probably insufficient for children with chronic complex conditions. Further research is needed to more clearly define the referal criteria to paediatric liaison teams.

Children’s quality of life in a family-centred approach is the main goal to reach for paediatric liaison teams, but this goal is evaluated by feel.

Paediatric liaison teams show their interest in discovering and using outcome measurement instruments.

Which, though, are the instruments used in paediatric palliative care to measure outcomes?

CHAPTER 4. RELIABLE INSTRUMENTS TO MEASURE OUTCOMES OF PAEDIATRIC PALLIATIVE CARE ARE LACKING AND SELDOM INCLUDE CHILD'S SELF-REPORTING: A SYSTEMATIC LITERATURE REVIEW

This chapter was published as an article under the title: Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi: 10.1542/peds.2018-2379.

Abstract

Background and objectives: Paediatric palliative care (PPC) intends to promote children's quality of life using a family-centered approach. However, measurement of this multidimensional outcome remains challenging. To review the instruments used to assess the impact of PPC interventions.

Methods: Five databases (Embase, Scopus, Cochrane Library, Psychinfo, Medline) were searched.Study Selection: Inclusion criteria: definition of PPC; age 0-18 years; diseases listed in the Directory of life-limiting diseases; results based on empirical data; combined descriptions of a PPC intervention, its outcomes and a measurement instrument. Full-text articles were assessed and data extracted by 2 independent researchers, and each discrepancy was resolved through consensus. The quality of the studies was assessed using a checklist, Standard Quality Assessment Criteria for Evaluating Primary Research papers from a Variety of Fields.

Results: Nineteen out of 2150 articles met the eligibility criteria. Fifteen of them used quantitative methods, and 9 were of moderate quality. Multidimensional outcomes included Health-related Quality of Life, spiritual well-being, and satisfaction with care/communication, perceived social support and family involvement in treatment or place of care preferences. PPC interventions ranged from home-based, hospital and respite care. Only 15 instruments (of 23 reported) indicated some psychometric properties, and only 5 referred to patient (child)-reported outcomes measures.

Conclusions: Data on the psychometric properties of instruments used to assess the impact of PPC interventions were scarce. Children are not systematically involved in reporting outcomes.

Introduction

The purpose of paediatric palliative care (PPC) is to enhance the quality of life of children and their families when facing life-limiting or life-threatening illnesses. According to The World Organization of Health, palliative care for children is the active total care of the child's body, mind and spirit, and it also involves giving support to the family. It begins when illness is diagnosed and continues regardless of whether a child receives treatment directed at the disease. (1) It is estimated that while approximately 21 million children worldwide would benefit from a palliative care approach, 8 million children are in need of specialized palliative care. (2)

Measuring outcomes in PPC is considered essential to improve clinical care, to evaluate the quality of services and to secure funding for programs, and it has been identified as a priority on the research agenda, especially in PPC. (3-8) However, there is currently little evidence of the effectiveness of PPC as it is difficult to define appropriate outcome measures in this field. (9-11) Several obstacles have been identified regarding outcomes research in PPC: small sample size, the difficulty of identifying a relevant comparison group, and finally, the broad heterogeneity in children's diseases and ages. Moreover, the relevance of assessing quality of life in children within a palliative context has been criticized by some consequentialist authors, who argue that in the face of inevitable death, measuring the impact of quality of life would no longer be a priority. (12) These assumptions are often linked to false representations of paediatric palliative care largely restricted to the very end-of-life moments.

A systematic review conducted by Coombes et al. showed that there is currently no ideal outcome assessment measure available yet for use in paediatric palliative care. (13) This finding is in agreement with the conclusion of Knapp et al. and Huang, who found none of the generic Quality of life measurement instruments to be valid for use in a paediatric palliative care context. (12,14)

Measuring quality of life has become a growing interest and an endpoint in many clinical settings. However, in studies of palliative care, quality of life may become the principal or only endpoint of consideration. (15) Quality of life outcomes are now also commonly called patient (or person)-reported outcomes (PROs) to reflect more clearly the broad spectrum of outcomes that are measured, such as pain, fatigue, depression and observable physical symptoms such as nausea and vomiting, which are included in the assessment. Measuring children's quality of life in a PPC context remains challenging for at least three reasons. First additional dimensions have been suggested by some researchers, such as the ability to cope with illness, spiritual dimension and satisfaction with life. (16) Second, depending on the nature of the disease and age of the child, proxy-assessments by a relative or other close observer are often used. Third, the diversity of diseases and contexts dealt with by PPC often makes it very difficult to disentangle the impact of disease severity and treatment from the impact of PPC interventions.

Quality of life is seldom measured directly, but rather explored through a combined assessment of several aspects labelled as dimensions, and although there is disagreement about the aspects that should be included, there is a consensus that QoL should be considered a multidimensional construct. Thus, regardless of the instrument used, items of different nature targeting different dimensions will be included.

Commonly, instruments measuring health-related outcomes rely on one of two theoretical frameworks depending on the relationship between the items and the construct to be measured.[17] The distinction between reflective and formative models in the field of QoL was introduced by Fayers et al. (15)

In a reflective model, the construct manifests itself, and the items are effective direct indicators of that construct, e.g., on a scale intended to measure anxiety, all items will reflect a certain degree of anxiousness. In a formative model, the items form or build the construct and are called causal items, e.g., in an instrument intended to measure stress, the amount of stress is measured by assessing many items that all contain stress-evoking events. In the field of QoL measurements, many instruments used in PPC settings have a hybrid nature and contain reflective and causal items. They most often are conceptualized as multidimensional scales and contain direct observable, self-reported and proxy-reported items. Thus, it is challenging to assess their psychometric characteristics.

This systematic review aims to identify and describe the instruments that have been used to assess the impact of PPC interventions and to assess their psychometric properties.

Methods

Search strategy

This study complied with the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines (18). The methods were prespecified and documented in a protocol (PROSPERO registration number: CRD42017080541).

We conducted primary systematic literature searches using a combination of keywords, including child, infant, paediatric, adolescent, young adult, palliative care, comfort care, supportive care, bereavement care, respite care, organization, standards, planning, outcomes, results and effects. (see Table 6 for the full search strings). We used broad keywords and MeSH terms to maximize inclusiveness and searched 5 databases: Embase, Scopus, Cochrane Library, Psychinfo and PubMed (Medline), for studies published from January 1, 2006 to June 1, 2016.

The year 2006 for starting the inclusion of studies was chosen because the first **I**nternational **M**eeting for **P**alliative **C**are in **C**hildren took place the same year in **T**rento, Italy (IMPaCCT), leading in 2007 to the first publication of standards for PPC in Europe. (19)

Table 6. Search strategy employed for studies presenting outcomes measured in paediatric palliative care

MBASE n=489)	(palliative and care or 'comfort'/exp or comfort and care or supportive and care or end and of and ('life'/exp or life) and care or advanced and care or respite and care or 'bereavement'/exp or bereavement and care and [2006-2016]/py) and (organization or organizations or plan or program or programs or project or projects or standards or standard or planning and [2006-2016]/py) and (infant or infants or child or children or paediatric or paediatric or adolescent or adolescents or young and adult or young and adults or toddler or toddlers and [2006-2016]/py) and (outcome or outcomes or result or results or effect or effects and [2006-2016]/py)
COPUS n=279)	(TITLE-ABS-KEY (organization OR organizations OR plan OR program OR programs OR project OR projects OR standards OR standard OR planning) AND PUBYEAR > 2005) AND ((TITLE-ABS-KEY (outcome OR outcomes OR result OR results OR effect OR effects) AND PUBYEAR > 2005) AND (((TITLE-ABS-KEY (bereavement care)) OR (TITLE-ABS-KEY (respite care)) OR (TITLE-ABS-KEY (advanced care)) OR (TITLE-ABS-KEY (supportive care)) OR (TITLE-ABS-KEY (end of life care)) OR (TITLE-ABS-KEY (comfort care)) OR (TITLE-ABS-KEY (palliative care))) AND (TITLE-ABS-KEY (infant OR infants OR child OR children OR paediatric OR paediatric OR adolescent OR adolescents OR young adult OR young adults OR toddler OR toddlers) AND PUBYEAR > 2005)))
ochrane ibrary n=844)	(palliative care OR comfort care OR supportive care OR end of life care OR advanced care OR respite care OR bereavement care) AND (infant OR infants OR child OR children OR paediatric OR paediatric OR adolescent OR adolescents OR young adult OR young adults OR toddler OR toddlers) AND (outcome OR outcomes OR result OR results OR effect OR effects) in Title, Abstract, Keywords, Publication Year from 2006 to 2016 in Trials'

sycinfo n=134)	1 (palliative care or comfort care or supportive care or end of life care or advanced care or respite care or bereavement care).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] (12563) 2 (infant or infants or child or children or paediatric or paediatric or adolescent or adolescents or young adult or young adults or toddler or toddlers).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] (738711) 3 (outcome or outcomes or result or results or effect or effects).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] (1868626) 4 1 and 2 and 3 (691) 5 limit 4 to yr="2006 - 2016" (557) 6 (organization or organizations or plan or program or programs or project or projects or standards or standard or planning).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] (709649) 7 5 and 6 (199)
edline n=404)	Search (palliative care OR respite care OR advanced care OR supportive care OR bereavement care OR "end of life care")AND (child OR children OR infant OR infants OR paediatric OR paediatric OR paediatric OR adolescent OR adolescents OR young adult OR young adults OR toddler OR toddlers) AND (outcome OR outcomes OR result OR results) AND (measure) Filters: Publication date from 2006/01/01 to 2016/06/01"

Study selection

All types of paediatric palliative care interventions or programs were eligible for inclusion, such as supportive care, respite care, and bereavement care.

Furthermore, all types of outcomes, not restricted to quality of life but focused on the child, the siblings or the parents, were taken into account.

A study was included if all of the following criteria were fulfilled: (1) the full text was written in English, French, German, Italian or Dutch; (2) the study sample included a clear description of infants/children/adolescents ranging in age from 0-18 years; (3) children's diseases were included in the Directory of life-limiting diseases (20) or were labelled as life-limiting or life-limited diseases, or complex chronic conditions; (4) the study included empirical data; (5) the study presented a combined description of a PPC intervention, outcome and measure instrument; (6) a minimal definition of PPC was presented in the study.

In contrast, a study was excluded if any of the following criteria were fulfilled: (1) sickle-cell disease, diabetes, obesity, perinatal deaths, chronic pain; (2) age greater than 18 years; (3) a study restricted to a specific molecule or treatment assessment, to pain as the single outcome, or to an economic assessment.

Children with sickle cell diseases are very rarely referred to PPC teams. Management of pain alone was not a criterion of inclusion. Perinatal deaths were also excluded because they concern a specific population with particular PPC needs and would need to be searched independently.

Data extraction and analysis

After retrieving all records, the duplicates were removed. All studies were initially screened on the basis of title and abstract, and then on the basis of the full-text. Three authors (M.F., I.A. and J.D.) independently assessed the eligibility of the studies. Any discrepancy were discussed and resolved by consensus. Quality of studies was assessed using a checklist, the Standard Quality Assessment Criteria for Evaluating Primary Research papers from a Variety of Fields. (21)

Box 1 offers detailed information on the data extraction and analysis.

Three authors (M.F., I.A. and J.D.) independently assessed the eligibility of the studies: M.F assessed all articles, and I.A and J.D each assessed half of the articles. Any discrepancy were discussed with the other two authors (I.A. and J.D.) and resolved by consensus. M.F., J.D., I.A. and A.D. selected the categories to build the extraction table. M.F. and J.D independently extracted the data on the study characteristics (publication year, country, study design, objectives of the study, sample size, type of disease, types of intervention provided, types of outcomes assessed, instruments used) and psychometric characteristics of the instruments. The following information on psychometrics was collected: content validity, internal consistency, construct validity, reliability, and acceptability. Moreover, when an included study did not report any information on psychometrics but referred to other articles regarding a measure, we assessed the additional articles to evaluate the evidence they provided.

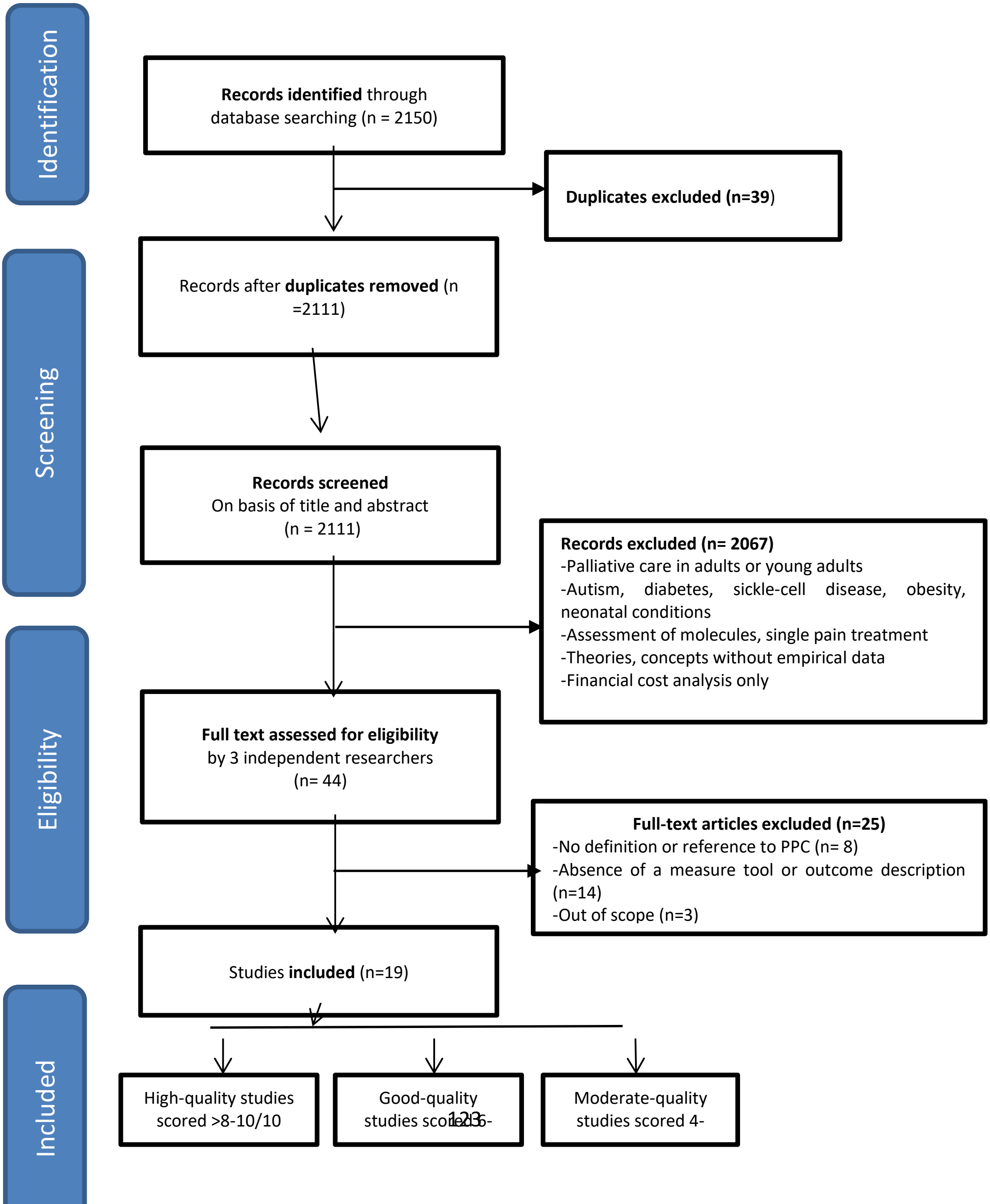
Additionally, the quality of the studies included was assessed using a checklist, the Standard Quality Assessment Criteria for Evaluating Primary Research papers from a Variety of Fields.21 This scale offers 2 versions with separate questions: one adapted to qualitative studies and the other suitable for quantitative studies. Two independent researchers, M.F. and J.D., assessed the quality of the articles using the same scale. Discrepancies in scores were discussed until a consensus was reached. Total scores obtained were converted on a 10-point scale for comparison among all 19 studies. Depending on their score, the articles were classified into 4 categories. The quality of the articles that were scored >8-10/10 designated as category 1 (high quality), >6-8/10 as category 2 (good quality), >4-6/10 as category 3 (moderate quality) and below 4/10 as category 4 (poor quality).

The reliability of the measuring instruments

There are several ways to interpret a reliability coefficient of a given value depending on the type of characteristic measured and on the method of obtaining the estimate of reliability. In classical test theory, a reliability coefficient can be interpreted as the proportion of the observed variance that is “true” rather than the “error variance”. However, one difficulty with expressing the reliability coefficient as a dimensionless ratio of variances is that it is difficult to interpret in terms of an individual score (22). The standard error of measurement (SEM) is defined in terms of the standard deviation (σ) and the reliability (R) as $SEM = \sigma \times \sqrt{1-R}$. Knowledge of the SEM makes it possible to construct a 95% confidence interval (95% CI = ±1.96 x SEM) around a person’s observed score so that the amount of measurement error around that score can be quantified in a meaningful way (17). Thus, we decided to compute the SEM wherever possible, using either the reported reliability index or any other reliability figures available in the literature.

Results

Figure 6 PRISMA flow diagram of the included studies assessing outcomes



After duplicates were removed, 2111 studies were screened on the basis of titles and abstracts, and 44 studies were screened on the basis of the full text. This procedure identified 19 studies that met all the inclusion criteria.

Study characteristics

Table 7 provides an overview of the main characteristics of the included studies.

Table 7. Overview of the included studies assessing outcomes in paediatric palliative care

References	Design	Methods	Study period	Country	Age	Pathology	Sample size	Instruments	Quality appraisal (1= good quality)
Akard et al. (2015)[23]	RCT	Mixed	**NR**	USA	7-17 y	Cancer	Intervention group n=15 Control group n=13	PedsQoL 4.0 (Paediatric Quality of life questionnaire)	2
Al-Gharib et al. (2015)[24]	Cross-sectional	Quantitative	2010-2011	Libanon	7-18 Y	Cancer	n=85	NEST questionnaire (needs at the end of life scale)	3
Arland et al. (2013)[25]	Retrospective cohort	Quantitative	1990-1995 1996-2005	USA	1 m-19 Y	Cancer (brain tumor)	n=22 n=92	Chart review	3
Bradford et al. (2012)[26]	Prospective cohort	Quantitative	2010-2011	Australia	0-18 y	LLC	Intervention group n=6 Control group n=6	QOLLTI-F questionnaire (quality of life in life-threatening Illness-Family carer version)	3
Conte et al. (2016)[27]	Match-paired cohort	Quantitative	2008-2012	Canada	**NR**	LTC	11 pairs	Chart review	3
Eaton et al. (2008)[28]	Prospective	Qualitative	**NR**	UK	5-16 Y	LTC	n=11 families	Interviews	2
Friedrichsdorf et al. (2015)[29]	Retrospective cohort	Quantitative	2002-2008	USA	0-17 Y	Cancer	Intervention group n=30	SCCC questionnaire (survey about caring for children with cancer)	2

							Control group n=30		
Gans et al. (2015)[30]	Prospective	Quantitative	2010-2012	USA	1-20 Y	LLC	n= 93/50/18 survey	Customized survey	3
Groh et al. (2013)[31]	Prospective	Quantitative	2011-2012	Germany	1 m-18 Y	Severe illness	n=40	CQ1, CQ2 customized children's questionnaire HPS (Haüsliche Pflegeskala), HADS (Hospital Anxiety &Depression Scale), QOLLTI-F (Quality of life in life-threatening Illness-Family carer version)	3
Harris et al. (2016)[32]	Prospective	Mixed	2013-2014	UK	3 m-21 Y	LLC	n=32 families	Family empowerment scale	3
Lyon et al. (2013)[33]	RCT	Quantitative	2011-2012	USA	14-21 Y	Cancer	Interv. gr =17 dyads Control gr =13 dyads	Statement of treatment preferences (STP) Decisional Conflict Scale (DCS), Quality of Participant-Interviewer Communication, Congruence in treatment preferences	1
Lyon et al. (2014)[34]	RCT	Quantitative	2010-2012	USA	14-21 Y	Cancer	Interv. gr =17 dyads Control gr =13 dyads	Satisfaction questionnaire, Five wishes, Beck anxiety inventory (BAI), Beck depression inventory (BDI), PedsQL 4.0, Spiritual Well-Being Scale	1

Remedios et al. (2015)[35]	Pre-post study	Mixed	2011-2012	Australia	0-18 Y	LLC	58 children/caregivers	Sort form -12 items Health Survey (SF-12 v1), General Health Questionnaire (GHQ), Chalder fatigue scale (CFS), quality of marriage index (QMI), multidimensional scale of perceived social support (MSPSS)	2
Schmidt et al. (2013)[36]	Retrospective cohort study	Quantitative	2000 (cohort 1) and 2005 (cohort 2)	Germany	0-18 Y	Cancer	46 families (cohort 1) 32 families (cohort 2)	Survey of caring for children with cancer (SCCC)	3
Swallow et al. (2011)[37]	Prospective	Qualitative	NR	UK	14-18 Y	LLC	15 families (5 teenagers, 15 mothers, 5 fathers)	Interviews and focus groups	1
Van der Geest et al. (2014)[38]	Retrospective cross-sectional study	Quantitative	2000-2004	Netherlands	0-18 Y	Cancer	89 parents of 57 deceased children	Inventory of traumatic grief (ITG), questionnaire on parents 'experience, parent’s rating of severity and control of symptoms during palliative phase	1
Vern-Gross et al. 2015)[39]	Retrospective (pre-post) cohort study	Quantitative	2001-2005 2007-2012	USA	0-21 Y	Cancer	57 children (cohort 1) 134 children (cohort 2)	Chart review	2

Vollenbroich et al. (2012)[40]	Retrospective study	Quantitative	2004-2007	Germany	0-34 Y	LLC	43 parents (from 38 deceased children)	Parental Questionnaire (PQ1), Hospital Anxiety & Depression Scale (HADS), Prolonged grief disorder (PG13), Questionnaire for Health Care Professionals (QHCP)	3
Wolfe et al. (2014)[41]	Multicenter pilot RCT	Quantitative	2004-2009	USA	>2 Y	Cancer	Intervention group : 51 Control group :53	Pediquest (MSAS, PedsQL)	1

Legend:

RCT: randomized controlled trial NR: not reported LLC: life-limiting condition LTC: life-threatening condition Y: yea

A total of 19 studies were included in this systematic review, of which 15 utilized quantitative methods, 1 used qualitative methods and 3 were based on mixed-methods (qualitative and quantitative methods). Regarding the design of the studies, 9 were retrospective, 6 were prospective and 4 consisted of randomized controlled trials. Among all the included studies (n=19), 14 used instruments (n=23 different instruments).

Table 8 shows an overview of the most cited instruments assessing outcomes in paediatric palliative care in the included studies.

Table 8. Overview of the most cited instruments assessing outcomes in paediatric palliative care in the included studies

Name of instrument	Dimensions assessed	Form	Number of items	Self or proxy report	References of studies using this instrument
Paediatric Quality of Life Questionnaire (PedsQoL 4.0)	Children's QoL: physical, emotional, social functioning; school.	Questionnaire Rating scale	23	Self (child) and proxy report (parent)	23,34,41
Quality of life in life-threatening Illness-Family carer version (QOLLTI-F)	Caregiver's QoL: perceived QoL, patient's environment and condition, caregiver's physical, emotional and cognitive condition, meaning in life, quality of care, relationships, financial worries	Questionnaire Rating scale	16	Self-report (parent)	26,31
Survey about caring for children with cancer (SCCC)	End-of-life care domains: symptoms and their treatment; QoL, parent sociodemographic characteristics	Questionnaire Rating scale	211	Proxy report (Parent)	29,36
Needs at end of life screening scale (NEST)	Adult's financial burden, quality of medical care, spirituality/religion, relationships	Questionnaire Rating scale	35	Self (child) and proxy report (parent)	24
Hospital anxiety and depression scale (HADS)	Parent's anxiety and depression	Questionnaire Rating scale	14	Self-report (Parent)	31,40

Three studies were based exclusively on a chart review, and 2 studies used interviews or focus groups to measure outcomes. The included studies covered the observational period from 1990 to 2014, a timeframe of 24 years, excluding 2 studies that did not specify the time period. The patient population concerned cancer patients (n=10) or children facing various life-limiting or life-threatening conditions (n=8) or having a “serious illness” (n=1). Of the studies, 8 were conducted in the USA, 7 in Europe, 2 in Australia, 1 in Canada, and 1 in Lebanon. Half of the studies collected data exclusively among parents (n=9). Children and parents were both interviewed in 5 studies. One study collected data among parents and health professionals, and another study combined the children’s, parents’ and health care professional’s perspectives. Sample sizes varied from 11 families to 134 families.

Quality appraisal of articles

Classification of the quality of the studies indicated that 5 studies had a high quality (>8/10), 5 had good quality (6-8/10), and 9 had moderate quality (4-6/10). None of the studies were labelled as having poor quality.

Types of interventions

Interventions or programs presented in the studies varied from home care (n=6), to hospital care (n=5), hospice care (n= 2), respite care (n=3), or a combination of home, community-based and hospital care (n=3).

Flexibility was found in the individualized approach of PPC interventions, where the focus is placed on the personal desire and priorities expressed by the child and his family. Table 9 shows the types of interventions offered, the outcomes expected versus achieved and the definition of PPC.

Table 9. Overview of the type of interventions and outcomes of paediatric palliative care assessed in the included studies

References	**Care setting**	**Type of intervention**	**Expected outcomes**	**Observed outcomes**	**Definition of PPC**
Akard et al. (2015)[23]	Home care	Legacy-making intervention/digital storytelling	To evaluate if a legacy-making intervention is a possible strategy to improve QoL	The intervention group that benefitted from a legacy-making intervention showed slightly better emotional and school functioning than controls. Parents reported that their child's digital story provided emotional comfort to them (n=11, 46%), facilitated communication between parents and children (n=9, 38%), and was a coping strategy for them (n=4, 17%). Parents reported that the intervention helped children express their feelings (n=19, 79%), cope (n=6, 27%), and feel better emotionally (n=5, 23%).	ND
Al-Gharib et al. (2015)[24]	Hospital care	Tertiary paediatric cancer center	To evaluate the quality of medical care received, effect of cancer on patients' relationships, degree of patients' spirituality/religiousness, and degree of parents' financial hardships	Participation in decision making significantly higher in adolescents than in children ($P= 0.004$). Compared by age, adolescents showed more readiness to learn from illness than children ($P=0.016$). All respondents reported a high quality of medical care.	PPC is an approach that improves the quality of life of patients and families facing a life-limiting disease, which may be achieved through effective symptom management as well as spiritual and psychological support from diagnosis until end of life and bereavement. PC is defined as "the active, total care of the child's body, mind and spirit, and also involves giving

					support to the family. It begins when illness is diagnosed, and continues regardless of whether or not a child receives treatment directed at the disease" (WHO)
Arland et al. (2013)[25]	Hospital and home care	Comprehensive EOL discussions, medications for symptom control, primary family liaison, and home visits.	To examine if a relationship existed between specific patient outcomes (i.e., symptoms, hospitalizations, and location of death) before and after implementation of a standardized EOL program among paediatric patients with brain tumors: patients who received the EOL program were hospitalized less often (n = 114; chi-square = 5.001 with df = 1, p < .05) than patients who did not receive the program.	Patients who received the EOL program were hospitalized less often (n = 114; chi-square = 5.001 with df = 1, p < .05) than patients who did not receive the program. The authors suggest that these patients may have had a reduced number of hospital admission days because the EOL program included comprehensive EOL care discussions, continuous infusion of midazolam, an assigned clinic nurse or PNP liaison for communication with the family and the hospice nurse, and healthcare provider home visits.	Lack of clarity in recognized definitions for both palliative care (PC) and EOL care. It is recognized that PC is focused on reducing and preventing suffering and improving quality of life (QOL) regardless of the patient's diagnosis. In contrast, EOL is recognized as part of the continuum of PC with the major focus on symptom management and prevention of suffering for patients with a life-threatening diagnosis that is expected to lead to death. The common goal of PC and EOL care is achievement of the best QOL for all affected patients.
Bradford et al. (2012)[26]	Home care	Home telehealth program: symptom management, emotional support to caregivers.	To Improve the QoL of primary caregivers of children receiving palliative care via the HTP (Home telehealth program)	The descriptive analysis of the QoL data that was collected showed no differences in QOLLTI-F scores between caregivers in the control and intervention groups. Finances and caregiver's own physical and emotional were stated as poor. The patient condition, as expected in palliative care, deteriorated over time. Domains that scored highly in both groups included quality of care, satisfaction with care and the environment for care. No relationship	ND

				was found between QOLTTI-F scores and HTP intervention.	
Conte et al. (2016)[27]	Hospice care	Not described	To compare the number of admissions and length of stay between PPCP (paediatric palliative care patients) users versus those children who received usual care.	PPCP users were more likely to have advanced directives (100% vs. 27%). After controlling for disease and age, we found no significant difference in the number of admissions; however, PPCP users had an increase in admissions postreferral compared with prereferral (median 3.08 admissions), driven by the need for critical care. We did not find a significant difference in LOS (length of stay) but observed longer admissions among PPCP users pre- (1.91 days/month) and postreferral (3.66 days/month) than usual care. More than 60% of inpatient utilization shifted to the hospice postreferral.	ND
Eaton et al. (2008)[28]	Respite care	Hospice-based and home-based respite care services	Descriptive study: satisfaction with service, parent's contribution to care, benefits of service	The areas of concern identified as significant to all the families were referral to the respite service, service organization, communication, relinquishing control to respite carers and satisfaction with service.	Definitions of hospice and home-based respite care
Friedrichsdorf et al. (2015)[29]	Home care	Not described	Reduce symptom of distress and improve the QoL experience for children	Prevalence of constipation and high distress from fatigue were more common in the PPC/oncology group; other distressing symptoms were similar between groups, showing room for improvement. Children who received PPC/oncology were significantly more likely to have fun (70% versus 45%), to	(PPC) aims to provide excellent pain and symptom management as well as psycho-social-spiritual care, regardless of the expected disease trajectory. Integration of PPC services appears to improve outcomes, such as improved pain and symptom management, improved quality of life,

				experience events that added meaning to life (89% versus 63%), and to die at home (93% versus 20%).	fewer emergency room visits, and a reduced paediatric intensive care unit (PICU) length of stay.
Gans et al. (2015)[30]	Home care, community-based service	Not described	Impact of service to decrease caregiver level of stress and worry	Worry and stress improved in the overall study population between baseline and follow-up surveys. The reductions over time were significant when controlling for the effects of family support and the child's age and disease severity, resulting in a 0.26-point reduction per survey wave for both worry and stress. Family support was also predictive of reductions in both stress and worry. Disease severity was also significantly predictive of stress. Age was not independently associated with changes in caregiver stress and worry. Caregivers' perceptions of the ability of PFC to reduce or control the child's pain and other symptoms received average ratings of 8.6 and 9.3, respectively, on a 10-point scale.	From the time of diagnosis with a LTD and as a supplement to clinical treatment, PPC provides children with holistic care that considers every aspect of the effect of the diagnosis on the child's and family's physical, psychological, and spiritual health. For the child, PC may include pain and symptom management and child life counselling and expressive therapies that provide the child with developmentally appropriate coping mechanisms and tools for self-expression. For the family, palliative care may include training on health care system navigation, treatment decisions, optimal care for their child, and bereavement counselling before and after the death of a child. Paediatric conditions are different from more prevalent terminal illnesses in adults and require different modalities and care plans. (ICPCN)
Groh et al. (2013)[31]	Home care	Palliative medical and nursing care, 24/7 on-call service, psychosocial support,	Address the needs of patients and their families, increase acceptance and effectiveness of PPC, improve QoL in children and their families	After the involvement of specialized PPHC, caregivers' satisfaction with care and quality of care significantly improved, as could be documented for 12 of 14 issues (NRS). The involvement of the PPHC team	(WHO) PPC is focused on achieving the best possible quality of life (QoL) for patients and their families and requires a multidisciplinary approach,

		coordination of professional assistance		led to a significant improvement of children's symptoms and QoL ($P < 0.001$), as perceived by the parents; and the parents' own QoL and burden relief significantly increased (QOLLTI-F, $P < 0.001$; 7-point change on a 10-point scale), while their psychological distress and burden significantly decreased (HADS, $P < 0.001$; HPS, $P < 0.001$).	encompassing physical, emotional, social and spiritual domains.
Harris et al. (2016)[32]	Hospice care	My quality	Acceptability of the tools and patterns of usage, increase in family empowerment	The mean duration of tool use was 106 days (range 2–301). The 32 families identified 114 priorities in total, with most identifying 2 or 3 (range 1–15). The most common priorities identified were seizures (24/32), constipation (9/32), pain (6/32) and sleep problems (6/32). Priorities were modified from the suggested options or entered as free text by 20 parents.	PC for children encompasses symptom control for the child, emotional and psychological support for the child and family, and addresses practical, financial and spiritual needs. The aim of PC is to maximize quality of life (QOL) rather than its duration. PC may be required for children with a wide variety of LLC, including cancer, organ failure, metabolic, genetic or degenerative illnesses, or static conditions such as severe cerebral palsy or epilepsy where LTC means that survival into adulthood is unlikely.
Lyon et al. (2013)[33]	Hospital care	Three sessions of family-centered advanced care planning (ACP): interviews	Examine the efficacy of family-centered ACP: document specific treatment preferences of patients and a surrogate understanding of what the patients would want, measure the degree of uncertainty about the course of action to be taken. improve	Significantly increased congruence was observed for intervention dyads compared with controls for 4 of the 6 disease-specific scenarios; for example, for situation 2 ("treatment would extend my life by not more than 2 to 3 months"), intervention dyads demonstrated higher congruence (= 0.660; P .001) vs. control dyads (= 0.0636;	Quality of PC is a health care priority

			quality of communication, congruence in treatment preferences, decrease decisional conflict	P=.70). Intervention adolescents (100%) wanted their families to do what was best at the time, whereas fewer control adolescents (62%) gave families this leeway. Intervention adolescents were significantly better informed about end-of-life decisions (t=2.93; effect size, 0.961; 95% CI, 0.742-1.180; P=.007). Intervention families were more likely to concur on limiting treatments than controls. An ethnic difference was found in only one situation.	
Lyon et al. (2014)[34]	Hospital Care	Paediatric Advanced care planning intervention (FACE-TC) during cure-directed treatments	Test feasibility, acceptability and safety of a paediatric ACP intervention called Face-TC	Acceptability was demonstrated with enrollment of 72% of eligible families, 100% attendance at all three sessions, 93% retention at 3 months postintervention, and 100% data completion. Intervention families rated FACE-TC worthwhile (100%), whereas adolescents' ratings increased over time (65%-82%). Adolescents' anxiety decreased significantly from baseline to 3 months postintervention in both groups. Low depressive symptom scores and high quality of life scores were maintained by adolescents in both groups. Advance directives were located easily in the medical records (100% of FACE-TC adolescents vs. none in controls). Oncologists received electronic copies. Total spirituality scores were significantly	PC currently exists in the context of a changing health care delivery system, in which death is medicalized. Four of five children with complex chronic conditions in the United States die in the hospital. Among children with cancer, 72% of those without PC died in the ICU versus 21% of those with PC . Advance care planning (ACP) is one dimension of palliative care that provides support in decision making about limiting burdensome medical interventions and advance directives.

				higher among FACE-TC adolescents versus controls.	
Remedios et al. (2015)[35]	Respite care	Out of home respite care service	Impact of respite care service on levels of fatigue, psychological adjustment, QoL, relationship satisfaction among caregivers	Caregivers had below-standard levels of quality of life compared with normative populations. Paired t-tests demonstrated that caregivers' average psychological adjustment scores significantly improved from prerespite (mean = 13.9, standard error = 0.71) to postrespite (mean = 10.7, standard error = 1); $p < 0.001$, 95% confidence interval: 1.25–5.11). Furthermore, caregivers' average fatigue scores significantly improved from prerespite (mean = 14.3, standard error = 0.85) to postrespite (mean = 10.9, standard error = 1.01; $p < 0.001$, 95% confidence interval: 1.69–7.94), and caregivers' average mental health quality of life scores significantly improved from prerespite (mean = 44.2, standard error = 1.8) to postrespite (mean = 49.1, standard error = 1.6; $p < 0.01$, 95% confidence interval: −9.56 to 0.36). Qualitative data showed caregivers sought respite for relief from intensive care provision and believed this was essential to their well-being.	Respite has been recognized as an important supportive service in paediatric palliative care. Broadly, paediatric respite involves individuals other than family members caring for the child with a medical condition for a short period of time. It is generally accepted that caregivers are the intended beneficiary of respite, with the aim to provide a break from intensive caring and thereby promote a sense of well-being.
Schmidt et al. (2013)[36]	Home care	Not described	PPC services reduce the symptoms and suffering of children, increase palliative home care, change the place of death	The children of both parental cohorts were similar in terms of disease characteristics and sociodemographic variables. Children suffered in a very similar manner from core	**ND**

				symptoms such as pain and dyspnea. However, symptom treatment increased for all symptoms. In the case of treatment of anxiety. the increase was statistically significant ($p = 0.035$). The location of care changed, with almost three-quarters of the 2005 cohort receiving palliative home care, which was significantly more than the 2000 cohort ($p = 0.007$). Additionally, fewer children in the 2005 cohort died in the intensive care unit.	
Swallow et al. (2011)[37]	Respite care	Offer a break, nursing and medical care, physiotherapy, social work, play and occupational therapy, clergy support providing residential care 24/7.	Adolescent and parent satisfaction with the service provided, skill of staff, partnership, access to leisure activities, opportunities for independence and peer support	Three themes emerged: accessibility and communication; needs and boundaries; and shaping the service. Teenagers enjoyed regular planned residential breaks, access to skilled staff and facilities to support their needs, opportunities to meet others with life-limiting conditions and fun time away from home, thereby giving parents peace of mind, a regular planned break from caregiving, opportunities to meet other parents and to spend exclusive time with their other children.	Definition of respite care
Van der Geest et al. (2014)[38]	Hospital care	Paediatric oncology center	Parents' perception and satisfaction with the interaction with HCPs (communication, continuity of care, parental involvement), symptom management, reduce long-term parental grief)	Parents' highly rated communication (4.6 +-0.6), continuity of care (4.3 +- 0.6), and parental involvement (4.6 +- 0.7) during the palliative phase. Parents' most often reported physical and psychological symptoms of their child during the palliative phase were fatigue (75%), pain	Palliative care is defined by the WHO (1998) as ''the active total care of the child's body, mind, and spirit,'' and also comprises care for the family during and after the child's death.16 According to the WHO, ''palliative care begins when illness is diagnosed and continues

				(74%), anxiety to be alone (52%), and anger (48%). Higher ratings of parents on communication ($b=- 9.08$, $P = 0.03$) and continuity of care ($b=-11.74$, $P = 0.01$) were associated with lower levels of long-term parental grief. The severity of the child's dyspnea ($b= 2.96$, $P =0.05$), anxiety of being alone ($b = 4.52$, $P < 0.01$), anxiety about the future ($b =5.02$, $P < 0.01$), anger ($b =4.90$, $P < 0.01$), and uncontrolled pain ($b =6.60$, $P < 0.01$) were associated with higher levels of long-term parental grief. Multivariate models combining the interaction with health care professionals and symptom management showed a significant influence of both aspects on long-term parental grief.	regardless of whether or not a child receives treatment directed at the disease."
Vern-Gross et al. (2015)[39]	Hospital and home care	Inpatient/outpatient follow-up, team meetings, home visits, interdisciplinary team, symptom management, psychosocial and spiritual support, coordinated care, facilitate EOL discussion, decision making and bereavement support	Reduce symptom burden, preferred location of death respected, EOL discussion, bereavement support	The median time to the first QoL/PC consultation was 17.2 months (range 9-33). At consultation, 60% of children were not receiving or discontinued cancer-directed therapy. Within the QoL/PC cohort, 54 patients had documented symptoms, 94% required intervention for >3 symptoms, and 76% received intervention for >5 symptoms. Eighty-three percent achieved their preferred place of death. Compared with the historical cohort, the QoL/PC cohort had more end-of-life discussions per patient (median 12 vs. 3; $P < 0.001$), earlier end-of-	Integration of palliative care (PC) for these children improves end-of-life care, enhances advance care planning, and substantially decreases suffering by improving communication, providing optimal symptom control, and emphasizing quality of life (QoL).

				life discussions, with longer times before do-not-resuscitate orders (median 195 vs. 2 days; $P < 0.001$), and greater hospice enrollment (71% vs. 46%, $P = 0.002$).	
Vollenbroich et al. (2012)[40]	Paediatric palliative home care team (PPHCT)	Coordination of professional assistance, provision of palliative treatment, interdisciplinary care and health care professionals (HCP), 24/7 service	To evaluate satisfaction with the service, satisfaction with the course of dying phase, reduce anxiety, depression, prolonged grief disorder	Satisfaction with the PPHCT scored a median of 10 (numeric rating scale, 0–10). The child's death was predominantly experienced as very peaceful (median, 9); 71% died at home. According to parents, involvement of the PPHCT led to highly significant ($p < 0.001$) improvements in the children's symptoms and quality of life, as well as in aspects of communication and administrative barrier reduction. Anxiety was detected in 25% of parents, depression in 19%, and prolonged grief disorder in 13%. HCPs (return rate, 83%) evaluated all investigated care domains (particularly cooperation/communication/family support) as being significantly improved ($p < 0.001$). Thirty-five percent of HCPs felt uncertain concerning paediatric palliative	The goal of PPC is to achieve the best quality of life (QOL) for patients and their families, consistent with their values, regardless of the location of the patient (WHO)

				care; 79% would welcome specific training opportunities.	
Wolfe et al. (2014)[41]	Hospital care		To determine whether feedback on patient-reported outcomes (PROs) to providers and families of children with advanced cancer improves symptom distress and health-related quality of life (HRQoL).	Feedback did not significantly affect the average MSAS, PedsQL4.0, or sickness score trends. Post hoc subgroup analyses among children age 8 years who survived 20 weeks showed that feedback improved PedsQL4.0 emotional (8.1; 95% CI, 1.8 to 14.4) and Sickness (8.2; 95% CI, 14.2 to 2.2) scores. PediQUEST reports were valued by children, parents, and providers and contributed at least sometimes to physician initiation of a psychosocial consult (56%).	Ensuring the best possible quality of life for children with cancer is a high national priority

Types of outcomes

Outcomes addressed in the studies, when focusing on children, were multidimensional and included physical (pain, fatigue, dyspnea, appetite), psychological (anxiety, depression), social (relationships) and spiritual dimensions. Quality of life, when explicitly explored, was defined as “having fun or experiencing events that added meaning to life” (23,26,27). Other outcomes were related to the satisfaction with the care and communication with health care professionals, often in parallel with the opportunity given to the child or adolescent to express wishes, treatment preferences or the place of care. Finally, the length of hospitalization or the place of death were outcomes searched through chart review and interpreted by authors as quality indicators of services.

Psychometric properties of the instruments used to measure outcomes

Table 10 presents the psychometric properties of the instruments included in the review.

Among the studies, 23 different instruments were identified (PedsQoL 4.0 in 3 studies, QOLLTI-F in 2 studies, SCCC in 2 studies, HADS in 2 studies), all of which were standardized measures. The average of the number of instruments used per study was 2, ranging from 1 to 6 instruments.

Among the psychometric properties, the alpha Cronbach was the most frequently reported value (indicated for 15 instruments among a total of 23). The alpha Cronbach is a measure of internal consistency that reveals how closely related a set of items are as a group. However, no data other than consideration of the face validity of the instruments were found among the included studies.

The standard error of measurement could be computed for 12 different instruments presented in 8 different studies. The SEM varied from 0,38 on a scale from 0-70 (with 95% CI= +-0,74) for the QOLLTI-F to 6,27 on a scale from 0-100 (with 95% CI = +-12,29) for the PedsQL 4.0.

Table 10. Overview of the instruments used in the included studies assessing outcomes in paediatric palliative care

References	**Name of instruments/methods used for assessment**	**References**	**Child/ parent/HCP report**	**N. items**	**Psychometric properties**	**SEM (standard error of measurement) and Confidence Interval (95% CI)**
Akard et al. (2015)[23]	Paediatric Quality of Life questionnaire (PedsQL 4.0)	Varni 2001 [42], 2005 [43]	Parent+Child	22	PedsQL Cronbach α 0,88	SEM PedsQL= 6,27 95% IC= +- 12,29
Al-Gharib et al. (2015)[24]	Needs at end of life tool (NEST)	Emmanuel 2001 [44]	Parent+Child		NEST Cronbach α 0,64-0,86	SEM Nest = 0,53-0,7 95% IC= +-1,18
Arland et al. (2013)[25]	Chart review	NA	NA	NA	NA	NA
Bradford et al. (2012)[26]	Quality of life in life-threatening illness-family carer version (QOLLTI-F)	Cohen 2006 [45]	Parent	16	QOLLTI-F Cronbach α 0,857	No sufficient data available
Conte et al. (2016)[27]	Chart review	NA	NA	NA	NA	NA
Eaton et al. (2008)[28]	Interview	NA	Parent	NA	NA	NA
Friedrichsdorf et al. (2015)[29]	Survey about caring for children with cancer (SCCC)	Wolfe 2000 [46], Hechler 2008 [47] Mack 2005 [48], 2008 [49] Edwards 2008 [50] Dussel 2009 [51]	Parent	211	ND	No sufficient data available
Gans et al. (2015)[30]	Chart review, own survey		Parent		NA	NA
Groh et al. (2013)[31]	Child questionnaire 1 (CQ1) Child questionnaire 2 (CQ2) Haüsliche Pflegeskala (HPS) Hospital Anxiety &Depression Scale (HADS) Quality of Life in life-threatening Illness-Family Carer (QOLLTI-F)	Gräßel 2001, 2003 [52,53] Snaith 1990 [54] Hermann-Lingen 2011 [55] Cohen 2006 [45]	Parent Parent Parent Parent Parent	71 61 10 14 19	HADS: Cronbach α 0,78-0,90 Pearson Correlation Coefficient 0,49-0,63 QOLLTI-F_Cronbach α 0,857	For HPS and HADS, insufficient data available SEM QOLLTI-F = 0,38 95% IC=0,74

Harris et al. (2016)[32]	MyQuality Family empowerment scale (FAS)	Harris 2014 [56], 2015 [57] Koren 1992[58], Nachshen 2005[59] Herbert 2009 [60]	Parent	 34	FAS: Congruence coefficients 0,88-0.98 Cronbach's α 0,78-0.89 Split-half 0.93	For FAS, no sufficient data available
Lyon et al. (2013)[33]	Statement of treatment preferences (STP) Decisional Conflict Scale (DCS) Quality of Participant-Interviewer Communication (QPIC) Congruence in treatment preferences (CTP)	Kirchhoff 2010 [61] O'Connor 1995 [62] Engelbert 2006 [63]	Parent+Child	 10	 DCS: test-retest 0,81 Cronbach α 0,78-0,92 Effect size 0.961 (95% CI, 0.742-1.180, p= 0.007)	For DCS and CTP, no sufficient data available SEM QPIC= 0,54 95% IC= +-1,06
Lyon et al. (2014)[34]	Satisfaction questionnaire Five wishes Beck anxiety inventory (BAI) Beck depression inventory (BDI) Paediatric Quality of life Questionnaire (PedsQL 4.0) Spiritual Well-Being Scale (FACIT)	 Beck 1993,1996. [64,65] Varni 2003 [66] Peterman 2002 [67]	Parent+Child	 21 22 12	BAI Cronbach's α >0,70 BDI Cronbach's α 0,91 PedsQL Cronbach's α 0,88 FACIT: Cronbach's α 0,86	SEM BAI = 3,18 95% CI= +-6,23 SEM BDI =1,71 95% CI=+-3,35 SEM PedsQL= 6,02 95% CI=+-11,8 SEM FACIT =6,14 95% CI= +-12,03
Remedios et al. (2015)[35]	SF-12 Health Survey (v.1) General health Questionnaire (GHQ) Chalder fatigue scale (CFS) The quality of marriage index (QMI) Multidimensional scale of perceived social support (MSPSS)	Ware 1994 [68] Goldberg 1997 [69] Chalder 1993 [70] Welch 2012 [71] Zimet 1988 [72]	Parent	12 12 11 6 12	SF-12 Cronbach's α 0,72-0,89 SF-12 Test-retest 0,73-0,86 GHQ Cronbach's α 0,82-0,86 CFS: Cronbach's α 0,80-0,86 QMI: Cronbach's α >0,90 test 0,65 MSPSS: Cronbach's α 0,89	SEM SF12 = 4,65 95% CI=+-9,11 SEM GHQ = 1,76 95% CI=+-3,45 SEM CFS =2,06 95% CI=+-4,04 SEM QMI = 1,74 95% CI=+-3,41
Schmidt et al. (2013)[36]	SCCC	Hechler 2008 [47] Wolfe 2000 [46]	Parent	NR	NR	No sufficient data available
Swallow et al. (2011)[37]	Interviews and focus group	NA	Parent+Child		NA	NA
Van der Geest et al. (2014)[38]	Inventory of traumatic grief, Questionnaire on parents 'experience, Parents rating of severity and control of symptoms during palliative phase	Boelen 2003 [73]	Parent	29 17 9		Only subscales no total score of the SEM could be computed
Vern-Gross et al. (2015)[39]			Chart review		NA	NA
Vollenbroich et al. (2012)[40]	Parental Questionnaire (PQ1) Hospital Anxiety & Depression Scale (HADS)	 Snaith 1990 [54]	Parent +Health	56 14	HADS: Cronbach α 0,78-0,90	No sufficient data available

	Prolonged grief disorder (PG13) Questionnaire for Health Care Professionals (QHCP)	Prigerson 2008 [74]	care professional	13 22	Pearson Correlation Coefficient 0,49-0,63	
Wolfe et al. (2014)[41]	Pediquest composed by: Memorial symptom assessment scale (MSAS) Paediatric Quality of life questionnaire (PedsQL 4.0) Sickness question	Collins 2002 [75] Varni 2001[42]	Parent+Child+Health care professional	24 22 1	MSAS Cronbach's α 0,67 PedsQL Cronbach's α 0,88	SEM MSAS = 4,94 95% CI= +-9,68 SEM PedsQL = 5,85 95% CI=+-11,47

Discussion

Paediatric palliative care is a recent subspecialty in medicine that has rapidly developed over the past two decades. Attempts to assess the impact of such interventions are progressively published, but the findings of this review show that the psychometric properties of the instruments used in this field are not well documented. The aim of PCC is to promote quality of life, but a precise description of how this construct is operationalized has not been provided in any of the included studies. As a matter of fact quality of life was addressed as an outcome directly or indirectly in only 6 out of the 19 studies. Measuring the quality of life of children in a context of a life-limiting disease requires rigorous conceptual and methodological foundations to capture core domains using a holistic and family-centered approach. Some methodological, clinical and conceptual challenges will be further discussed.

Patient-reported outcomes measures versus proxy-reported outcome measures

Overall, this review showed that outcomes measures predominantly explore parental (proxy) perspectives retrospectively concerning a past event, such as child's symptoms and circumstances of death or the satisfaction with the care received. However, retrospective studies have been found to be less reliable than prospective ones to assess outcomes. Conte et al. (76) affirms that prospective studies are needed to better evaluate the overall impact of PPC on the health care system from the perspectives of children and families. Furthermore, outcomes measures in paediatric palliative care seldom involve the perspectives of the children, as found in our review, in which only 5 studies addressed children's views. Ethical concerns are widely documented, arguing that participating in interviews may be burdensome for children facing life-limiting conditions (77). In recent decades, however, several authors have recommended the involvement of children in research showing that if the design is adapted to the child's cognitive and physical conditions and researchers adopt a compassionate and sensitive approach, participation in studies would be valued by children and their parents and would improve even the decision-making capacity (78). When children are involved in research, they are frequently associated or compared to proxy parental measures. In doing so, the discrepancy between self (by children) and proxy-reporting (by parents) is found, as reported in other studies (79-82), due to the unique subjective evaluation made by children, which clearly differs from that made by their parents.

The need for measurement instruments in PPC

It is hypothesized that the use of PROMS would improve the care quality provided. Nevertheless, this link is rarely assessed. Measurement instruments can be used for several purposes. They can be intended to be primary to guide the caregiving process rather than assess the impact of the interventions or to monitor patient-related health outcomes over time. Physicians often consider PROMS as useful for the assessment and screening of patients, whereas nurses perceive those instruments as almost relevant for clinical decision making. (83)

Difficulty of measuring outcomes lies in the fact that all the described interventions could be labelled as complex interventions, defined by Craig et al. as requiring a large number of health care professionals, organizational levels, outcomes and finally a large degree of flexibility. (84) Whether at home or in a hospital, PPC interventions are characterized by interdisciplinary teams composed of nurses, physicians, psychologists and social workers. Coordination of care is frequently ensured by several levels of care: primary home care teams collaborating with specialist hospital-based teams. Interdisciplinary teams are a core element of quality care in PPC because the aim is to promote quality of life not only for the child but also for the whole family, considering all the dimensions such as physical health, psychological well-being and social relationships.

Core dimensions to be measured

In adults, a study found that questions relating to pain, symptoms, emotional needs and family concerns are consistently considered the most useful and important components of palliative patient-reported outcome measures (PROMs) (85). Dimensions that matter the most for children in PPC are currently not known. According to its definition, palliative care of children consists of a family-centered approach to achieve the best quality of life of the child embedded among his family members. This focus should consequently be addressed through the choice of a measure that evaluates the outcomes of PPC on the child and his family. This idea was pointed out by Dussel et al. (86) However, patient involvement in the development of a PROM is paramount and has been documented for adults by Wiering et al. (87), but not in children. As already mentioned, health-related outcome measures are either based on formative (causal) or reflective models or mixed (17) For this review, we were interested in discovering which model was used for the instruments that we identified. We found that all the instruments identified in this review were based on reflective models, which indicated that the explored dimensions reflected the underlying construct, such as the quantity of pain or the degree of physical impairment.

Generic versus specific disease-oriented instruments

Another challenge is related to the choice of specific (disease specific, such as cancer) versus generic instruments (88). Generic measures may be useful to compare outcomes across different populations and interventions, especially for cost-effectiveness studies. Disease-specific measures assess the special states and concerns of diagnostic groups. Specific measures may be more sensitive for the detection and quantification of small changes that are important to clinicians or patients (89). A systematic review performed by Janssens et al. found that many generic instruments are available to assess children's health. However, they found that only a few PROMs were usable across all age ranges up to 18 years. Most PROMs excluded at least one major domain, and all of them conflated elements of functioning and well-being in the scales. (90) Additionally, the authors found at least five different psychometric properties for the Child Health and Illness Profile, the Healthy Pathways, the KIDSCREEN, and the Multi-dimensional Student Life Satisfaction Scale. (91)

Standardized versus individualized measures

Standardized measures are instruments that contain predefined domains, whereas individualized measures are instruments that encourage the patient to describe which domains are important to him. The only individualized measure of QoL is called the, scheduled evaluation of individual quality of life (SEIQoL) developed by Hickey in 1996. (92)

Standardized measures of QoL have been criticized by several authors, who argue that some psychometric properties of those measures, such as the test/retest, are not valid in a paediatric palliative care context. Rather, the same authors suggest the use of individualized measures of QoL (13,14), which seems to better reflect the subjective perception of QoL, independently of the decline in physical function. Another perspective is presented by Wolpert et al. (93), who recommended combining at least one standardized measure with one individualized measure to optimally assess QoL.

New developments in the field

Systematic reviews on PROMS have been performed for children suffering from burns (94) or who are visually impaired (95), but specific PROMS for PPC are clearly lacking.

Recently, through expert meetings and international conferences, a generic promising PROM was developed that addresses the specific methodological and conceptual challenges emerging from the paediatric palliative care context (96,97). This multidimensional instrument explores children’s perspectives on psychical, psychological and social aspects while receiving palliative care. Parental perspectives on information, confidence and worries are equally assessed using the same tool.

See Box 2 presenting the African Palliative Care Association Children’s Palliative Outcome Scale (APCA c-POS). Since no reports were published yet on the use of this new instrument in relation to an intervention we did not integrate this development in our overview.

Box 2 Presentation of the African Palliative Care Association - children's Palliative Outcome Scale (APCA c-POS)

The APCA c-POS (Downing 2012) is an instrument derived from the validated palliative outcome scale (POS), developed at Cicely Saunders Institute, Kings College London (Hearn et al. 1997)

The APCA c-POS explores in the same unique document the multidimensional outcomes of paediatric palliative care exploring physical, psychological domains for any child facing life-limiting or life-threatening conditions independently of his age or type of disease and psychological dimensions of his parents.

The instrument is composed of two parts: Part A addresses 7 questions to children (self-report) and part B 5 questions to their parents.

If the child isn't able to respond for himself in part A, due to his small age (< 6years) or his poor cognitive capacities, a proxy-report (to fill in by parents) is planned addressing the same dimensions.

Ex of question in part A exploring child's perceptions:

"Can you tell me how often you have felt happy since yesterday?"

Ex of questions in part B exploring parental perceptions:

"Have you been able to share how you are feeling about your child's illness with others when you have wanted to?"

PROMS make up an instrument that measures outcomes of care provided and provides valuable information on quality of care. Nevertheless, the impact of care should be assessed not only by outcomes measures. Documentation of the processes and types of interventions are required. As PPC intends to promote QoL, a question could concern how far PROMS contribute to the overall aim to evaluate QoL or the common ground or convergent domains assessed equivalently by PROMS compared with QoL instruments? (98)

Implementation of PROMS in clinical practice

After validation of an outcomes measure tool in paediatric palliative care, a next step would be a rigorous implementation process for which guidelines are issued. (99,100) Dissemination and implementation science is a growing research field that focuses on the best strategies to implement evidence practice. (101) Lack of education and training were found to be the most common reasons leading to the poor implementation of PROMS and therefore must be strengthened. (102) Some authors point out the importance of involving health care professionals right from the start during the development stages of a measure tool to improve its implementation. (103,104) Patient satisfaction towards a PROMS is an important element to consider in implementation research, which was explored by Recinos et al. in adults; however, similar research is lacking in children. (105)

Strengths and limitations

This systematic review offers, for the first time to our knowledge, an in-depth description of the instruments used to assess the impact of paediatric palliative care interventions.

Our assessment of the psychometric properties related to the instruments was restricted to the data that were retrievable from the publications included. We had no access to the development process used for the instruments or to the underlying concepts underpinning the construction of the scales.

Conclusion

This systematic review led to 2 observations. First, a broad spectrum of outcomes are targeted in the studies, demonstrating a lack of consensus on the dimensions that should be measured when assessing paediatric palliative care interventions. Second, the psychometric properties of outcome measurement instruments are poorly documented.

It is recommended to strengthen prospective studies involving children in a sensitive and age-appropriate way. Rigorous methods are needed to develop new instruments that are able to measure quality of life using a multidimensional and family-centered approach. The use of instruments that combine self and proxy-reports or even that address individual quality of life requires further investigation. Co-construction of the tool should be planned with health care professionals to guarantee optimal implementation into clinical care.

References

1. WHO definition of palliative care 1998a (accessed 12th February 2018) http://www.who.int/cancer/palliative/definition/en/

2. Connor SR, Downing J, Marston J. Estimating the global need for palliative care for children: a cross-sectional analysis. J Pain Symptom Manage. 2017;53(2):171-177. doi: 10.1016/j.jpainsymman.2016.08.020

3. Chong PH, De Castro Molina JA, Teo K, Tan WS. Paediatric palliative care improves patient outcomes and reduces healthcare costs: evaluation of a home-based program. BMC Palliat Care. 2018;17(1):11. doi: 10.1186/s12904-017-0267-z.

4. Goldhagen J, Fafard M, Komatz K, et al. Community-based paediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. BMC Palliat Care. 2016;3(15):73. doi: 10.1186/s12904-016-0138-z.

5. Downing J, Knapp C, Muckaden MA, Fowler-Kerry S, Marston J, ICPCN Scientific Committee. Priorities for global research into children's palliative care: results of an International Delphi Study. BMC Palliat Care. 2015;4(14):36. doi: 10.1186/s12904-015-0031-1.

6. Harding R, Wolfe J, Baker JN. Outcomes measurement for Children and Young People. Journal of Palliative Medicine. 2017;20(4):313. doi: 10.1089/jpm.2016.0525.

7. Voyles E. The development and outcomes of a paediatric palliative care program: a quality improvement process. J Pediatr Nurs. 2013;28(2):196-199. doi: 10.1016/j.pedn.2013.02.002.

8. Baker JN, Levine DR, Hinds PS, et al. Research Priorities in Paediatric Palliative Care. J Pediatr. 2015;167(2):467-70. doi: 10.1016/j.jpeds.2015.05.002.

9. Hearn J, Higginson I. Outcomes measures in palliative care for advanced cancer patients: a review. J Public Health Med. 1997;19(2):193-199.

10. Bausewein C, Daveson BA, Currow DC, et al. EAPC White Paper on outcome measurement in palliative care: Improving practice, attaining outcomes and delivering quality services - Recommendations from the European Association for Palliative Care (EAPC) Task Force on Outcome Measurement. Palliat Med. 2016;30(1):6-22. doi: 10.1177/0269216315589898

11. Araújo J, Dourado M, Ferreira PL. Measuring Instruments of the Quality of Life Paediatric Palliative Care. Acta Med Port. 2015;28(4):501-512.

12. Knapp C, Madden V. Conducting outcomes research in Paediatric Palliative Care. Am J Hosp Palliat Care. 2010;27(4):277-281. doi: 10.1177/1049909110364019.

13. Coombes LH, Wiseman T., Lucas G, et al. Health-related quality-of-life outcome measures in paediatric palliative care: a systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-949. doi: 10.1177/0269216316649155.

14. Huang I, Shenkman E, Madden V, et al. Measuring quality of life in paediatric palliative care: challenges and potential solutions. Palliat Med. 2010;24(2):175-182. doi: 10.1177/0269216309352418.

15. Fayers P, Machin D, Quality of Life. the assessment, analysis and interpretation of patient-reported outcomes. Wiley 2008, ISBN 987-0-470-02450-8.

16. Walters S. Quality of Life Outcomes in clinical trials and health-care evaluation. A practical guide to analysis and interpretation. 2009, Wiley and Sons, ISSBN 978-0-470-75382-8

17. De Vet HC, Terwee CB, Mokkink LB et al. Measurement in Medicine. Cambridge University Press 2011, 338p.

18. Moher D, Liberati A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. PLoS Med. 2009; 21;6(7):e1000097. doi: 10.1371/journal.pmed.1000097.

19. Craig F, Abu-Saad Huijer H, Benini F, Kuttner L, Wood C, Feraris PC, Zernikow B. IMPAcCT: standards for paediatric palliative care in Europe. European Journal of Palliative Care.2007;14(3):109-114. doi: 10.1007/s00482-008-0690-4. http://www.eapcnet.eu/LinkClick.aspx?fileticket=ShMQyZuTfqU%3D (accessed the 25th May 2018)

20.Hain R, Devins M, Hastings R, et al. Paediatric Palliative Care: development and pilot study of a “Directory” of life-limiting conditions. BMC Palliat Care. 2013;12:43. doi:10.1186/1472-684X-12-43.

21. Kmet LM, Lee RC, Cook LS. STANDARD QUALITY ASSESSMENT CRITERIA FOR EVALUATING PRIMARY RESEARCH PAPERS FROM A VARIETY OF FIELDS. HTA initiative #13, Feb 2004. https://www.ihe.ca/advanced-search/standard-quality-assessment-criteria-for-evaluating-primary-research-papers-from-a-variety-of-fields (accessed 25th May 2018)

22. Norman G, Streiner D (2008) Health Measurement Scales, a practical guide to their development and use. Oxford University Press. ISBN 978-0-19-923188

References of studies included:

23. Akard TF, Dietrich MS, Friedman DL, et al. Digital storytelling: an innovative legacy-making intervention for children with cancer. Pediatr Blood Cancer 2015;62(4):658-665. doi: 10.1002/pbc.25337.

24. Al-Gharib RM, Abu-Saad Huijer H, Darwish H. Quality of care and relationships as reported by children with cancer and their parents. Ann Palliat Med. 2015;4(1):22-31.

doi: 10.3978/j.issn.2224-5820.2015.01.04.

25. Arland LC, Hendricks-Ferguson VL, Pearson J, et al. Development of an in-home standardized end-of-life treatment program for paediatric patients dying of brain tumors. J Spec Pediatr Nurs. 2013;18(2):144-157. doi: 10.1111/jspn.12024.

26. Bradford N, Young J, Armfield NR,et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. J Telemed Telecare 2012;18(8):438-442. doi 10.1258/jtt.2012.

27. Conte T, Mitton C, Erdelyi S. Paediatric Palliative Care Program versus Usual Care and Healthcare Resource Utilization in British Columbia: A Matched-Pairs Cohort Study. J Palliat Med. 2016;19(11):1218-1223. doi: 10.1089/jpm.2016.0177.

28. Eaton N. 'I don't know how we coped before': a study of respite care for children in the home and hospice. J Clin Nurs. 2008;17(23):3196-3204.

doi: 10.1111/j.1365-2702.2008.02630.

29. Friedrichsdorf SJ, Postier A, Dreyfus J, et al. Improved quality of life at end of life related to home-based palliative care in children with cancer. J Palliat Med. 2015;18(2):143-150. doi: 10.1089/jpm.2014.0285.

30. Gans D, Hadler MW, Chen X, et al. Impact of a Paediatric Palliative Care Program on the Caregiver Experience. J Hospice Palliat Nurs. 2015; 17(6):559-565.

31. Groh G, Borasio GD, Nickolay C, et al. Specialized paediatric palliative home care: a prospective evaluation. J Palliat Med. 2013;16(12):1588-1594. doi: 10.1089/jpm.2013.0129.

32. Harris N, Beringer A, Fletcher M. Arch Dis Child. 2016;101(3):247-252. doi: 10.1136/archdischild-2015-308769.

33. Lyon ME, Jacobs S, Briggs L, et al. Family-centered advance care planning for teens with cancer. JAMA Pediatr. 2013;167(5):460-467. doi:10.1001/jamapaediatrics.2013.943.

34. Lyon ME, Jacobs S, Briggs L, et al. A longitudinal, randomized, controlled trial of advance care planning for teens with cancer: anxiety, depression, quality of life, advance directives, spirituality. J Adolesc Health. 2014;54(6):710-717. doi: 10.1016/j.jadohealth.2013.10.206.

35. Remedios C, Willenberg L, Zordan R.A pre-test and post-test study of the physical and psychological effects of out-of-home respite care on caregivers of children with life-threatening conditions. Palliat Med. 2015;29(3):223-230. doi: 10.1177/0269216314560008.

36. Schmidt P, Otto M, Hechler T, et al. Did increased availability of paediatric palliative care lead to improved palliative care outcomes in children with cancer? J Palliat Med. 2013;16(9):1034-1039. doi: 10.1089/jpm.2013.0014.

37. Swallow V, Forrester T, Macfadyen A. Teenagers' and parents' views on a short-break service for children with life-limiting conditions: a qualitative study. Palliat Med. 2012;26(3):257-267. doi: 10.1177/0269216311401947.

38. Van der Geest IM, Darlington AS, Streng IC, et al. Parents' experiences of paediatric palliative care and the impact on long-term parental grief. J Pain Symptom Manage. 2014;47(6):1043-1053. doi: 10.1016/j.jpainsymman.2013.07.007.

39.Vern-Gross TZ, Lam CG, Graff Z, et al. Patterns of End-of-Life Care in Children With Advanced Solid Tumor Malignancies Enrolled on a Palliative Care Service. J Pain Symptom Manage. 2015;50(3):305-312. doi: 10.1016/j.jpainsymman.2015.03.008.

40. Vollenbroich R, Duroux A, Grasser M, et al. Effectiveness of a paediatric palliative home care team as experienced by parents and health care professionals. J Palliat Med. 2012;15(3):294-300. doi: 10.1089/jpm.2011.0196.

41. Wolfe J, Orellana L, Cook EF, et al. Improving the care of children with advanced cancer by using an electronic patient-reported feedback intervention: results from the PediQUEST randomized controlled trial. J Clin Oncol. 2014.10;32(11):1119-1226. doi: 10.1200/JCO.2013.51.5981.

References of studies describing measures:

42. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Paediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. Med Care. 2001;39(8):800-812.

43.Varni JW, Burwinkle TM, Seid M. The PedsQL as a paediatric patient-reported outcome: reliability and validity of the PedsQL Measurement Model in 25,000 children. Expert Rev Pharmacoecon Outcomes Res. 2005;5(6):705-719. doi: 10.1586/14737167.5.6.705.

44. Emanuel LL, Alpert HR, Emanuel EE. Concise screening questions for clinical assessments of terminal care: the needs near the end-of-life care screening tool. J Palliat Med. 2001;4(4):465-474.

45. Cohen R, Leis AM, Kuhl D, et al. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006;20(8):755-767.

46. Wolfe J, Grier HE, Klar N et al.: Symptoms and suffering at the end of life in children with cancer. N Engl J Med. 2000;342:326-333. doi: 10.1056/NEJM200002033420506.

47. Hechler T, Blankenburg M, Friedrichsdorf S, et al.: Parents’ perspective on symptoms, quality of life, characteristics of death and end-of-life decisions for children dying from cancer. Klin Pädiatr. 2008;220:166-174. doi: 10.1055/s-2008-1065347.

48. Mack JW, Hilden JM, Watterson J, et al.: Parent and physician perspectives on quality of care at the end of life in children with cancer. J Clin Oncol. 2005;23:9155-9161. doi: 10.1200/JCO.2005.04.010.

49. Mack JW, Joffe S, Hilden JM, et al.: Parents' views of cancer-directed therapy for children with no realistic chance for cure. J Clin Oncol. 2008;26:4759-4764. doi: 10.1200/JCO.2007.15.6059.

50. Edwards KE, Neville BA, Cook EF Jr, et al.: Understanding of prognosis and goals of care among couples whose child died of cancer. J Clin Oncol 2008;26:1310-1315. doi: 10.1200/JCO.2007.13.4056.

51. Dussel V, Kreicbergs U, Hilden JM, et al.: Looking beyond where children die: Determinants and effects of planning a child's location of death. J Pain Symptom Manage 2009;37:33-43. doi: 10.1016/j.jpainsymman.2007.12.017.

52. Gräßel E: Home Care Scale for the Assessment of Subjective Burden of Mentoring or Nurturing Persons. Ebersberg: Vless Verlag, 2001.

53. Gräßel E, Chiu T, Oliver R: Development and Validation of the Burden Scale for Family Caregivers (BSFC). Toronto: COTA: Comprehensive Rehabilitation and Mental Health Services, 2003.

54. Snaith RP: The hospital anxiety and depression scale. Br J Gen Pract 1990;40(336):305.

55. Herrmann-Lingen C, Buss U, Snaith RP: Hospital Anxiety and Depression Scale – German Version. Berne: Verlag Hans Huber, 2011.

56. Harris. MyQuality. 2015. http://my-quality.net (accessed 24th May 2018).

57. Harris N, Beringer A, Fletcher M. Promoting patients' priorities through user-generated quality of life measures. 2014. http://www.health.org.uk/programmes/shine2012/projects/promoting-patients%E2%80%99-priorities-through-user-generated-quality-life (accessed 21 Oct 2017).

58. Koren PE, DeChillo N, Friesen B J. Measuring empowerment in families whose children have emotional disabilities: a brief questionnaire. Rehabil Psychol 1992;37:305-321.

59. Nachshen JS. Empowerment and families: building bridges between parents and professionals, theory and research. J Dev Disabil 2005;11:67-76.

60. Herbert RJ, Gagnon AJ, Rennick JE, et al. A systematic review of questionnaires measuring health-related empowerment. Res Theory Nurs Prac 2009;23:107-132.

61. Kirchhoff KT, Hammes BJ, Kehl KA, et al. Effect of a disease specific planning intervention on surrogate understanding of patient goals for future medical treatment. J Am Geriatr Soc. 2010;58(7):1233-1240. doi: 10.1111/j.1532-5415.2010.02760.x.

62. O'Connor AM. Validation of a decisional conflict scale. Med DecisMaking. 1995;15(1):25-30. doi: 10.1177/0272989X9501500105.

63. Engelbert R, Downey L, Curtis JR. Psychometric characteristics of a quality of communication questionnaire assessing communication about end-of-life care. J Pal Med. 2006;9(5):1086-1098. doi: 10.1089/jpm.2006.9.1086.

64. Beck AT, Steer RA. Beck Anxiety Inventory manual. San Antonio (TX): Psychological Corporation, Harcourt Brace & Company; 1993.

65. Beck AT, Steer RA, Brown GK. Beck Depression Inventory manual. 2nd ed. San Antonio (TX): Psychological Corporation, Harcourt Brace & Company; 1996.

66. Varni JW, Burwinkle TM, Seid M, et al. The PedsQL 4.0 as a paediatric population health measure: feasibility, reliability, and validity. Ambul Pediatr. 2003;3(6):329-341.

67. Peterman AH, Fitchett G, Brady MJ, et al. Measuring spiritual well-being in people with cancer the Functional assessment of chronic illness Therapy Spiritual Well-Being Scale (FACIT-Sp). Ann Behav Med. 2002;24:49e58. doi: 10.1207/S15324796ABM2401-06.

68. Ware JE, Kosinski M and Keller SD. SF-36 physical and mental health summary scales: a user's manual. Boston, MA: The Health Institute, New England Medical Center, 1994.

69. Goldberg DP, Gater R, Sartorius N, et al. The validity of two versions of the GHQ in the WHO study of mental illness in general health care. Psychol Med. 1997;27(1):191-197.

70. Chalder T, Berelowitz G, Pawlikowska T, et al. Development of a fatigue scale. J Psychosom Res. 1993;37(2):147-153.

71. Welch V, Hatton C, Emerson E, et al. Do short break and respite services for families with a disabled child in England make a difference to siblings? A qualitative analysis of sibling and parent responses. Child Youth Serv Rev. 2012;34(2):451-459.

72. Zimet GD, Powell SS, Farley GK, et al. Psychometric characteristics of the Multidimensional Scale of Perceived Social Support. J Pers Assess. 1990 Winter;55(3-4):610-617. doi: 10.1080/00223891.1990.9674095.

73. Boelen PA, Bout van den J, Keijser de J, et al. Reliability and validity of the Dutch version of the Inventory of Traumatic Grief (ITG). Death Stud. 2003;27:227e247. doi: 10.1080/07481180302889

74. Prigerson HG, Vanderwerker LC, Maciejewski PK: A case for the inclusion of prolonged grief disorder in DSM-V. In: Stroebe M, Hansson R, Schut H, (eds): Handbook of Bereavement Research and Practice: 21st Century Perspectives. Washington, D.C.: American Psychological Association Press, 2008, pp. 165-186.

75. Collins JJ, Devine TD, Dick GS, et al. The measurement of symptoms in young children with the validation of the Memorial Symptom Assessment Scale in children aged 7-12. J PainSymptom Manage. 2002;23:10-16.

76. Conte T, Mitton C, Trenaman L, et al. Effect of paediatric palliative care programs on health care resource utilization and costs among children with life-threatening conditions: a systematic review of comparative studies. CMAJ Open. 2015;3(1):e68-e75. doi: 10.9778/cmajo.20140044.

77. Wolfe J, Siden H. Chapter 39. Research in paediatric palliative care, in Goldman A, Hain R, Liben S. (Eds) Oxford Textbook of palliative Care for Children, 2012.

78. Rahimzadeh V, Bartlett G, Longo C, et al. Promoting an ethic of engagement in paediatric palliative care research. BMC Palliative Care. 2015;14(50). doi: 10.1186/s12904-015-0048-5.

79. Silva N, Crespo C, Carona C, et al. Why the (dis)agreement? Family context and child-parent perspectives on health-related quality of life and psychological problems in paediatric asthma. Child Care Health Dev. 2015;41(1):112-121. doi: 10.1111/cch.12147.

80. Lim Y, Velozo C, Bendixen R. The level of agreement between child self-reports and parent proxy-reports of health-related quality of life in boys with Duchenne muscular dystrophy. Qual Life Res. 2014;23(7):1945-1952. doi: 10.1007/s11136-014-0642-7.

81. Sattoe J, van Staa, Moll H. The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. Health Qual Life Outcomes. 2012;10:10. doi: 10.1186/1477-7525-10-10.

82. Vetter TR, Bridgewater CL, McGwin G. An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? Health Qual Life Outcomes. 2012;10:85. doi: 10.1186/1477-7525-10-85.

83. Daveson BA, Simon ST, Benalia H, et al. Are we heading in the same direction? European and African doctors' and nurses' views and experiences regarding outcome measurement in palliative care. Palliat Med. 2012 Apr;26(3):242-9. doi: 10.1177/0269216311409614.

84. Craig P, Dieppe P, Macintyre S, Michie S, Nazareth I, Petticrew M; Medical Research Council Guidance. Developing and evaluating complex interventions: the new Medical Research Council guidance. BMJ. 2008;337:a1655. doi: 10.1136/bmj.a1655.

85. Higginson IJ, Simon ST, Benalia H, et al. Which questions of two commonly used multidimensional palliative care patient reported outcome measures are most useful? Results from the European and African PRISMA survey. BMJ Support Palliat Care. 2012 Mar;2(1):36-42. doi: 10.1136/bmjspcare-2011-000061.

86. Dussel V, Medin G. Paediatric issues. In: Bruera E, De Lima L, Wenk R, Farr W (Eds.) Palliative Care in the Developing World: Principles and Practice. Houston: IAHPC Press. 2004.

87. Wiering B, de Boer D, Delnoij D. Patient involvement in the development of patient-reported outcome measures: The developers' perspective. BMC Health Serv Res. 2017;17(1):635. doi: 10.1186/s12913-017-2582-8.

88. Kane JR, Baker JN. Chapter 38. Quality improvement, in Goldman A., Hain R., Liben S. (Eds.). Oxford Textbook of Palliative Care for Children, 2012, pp. 430-440.

89. Patrick DL, Deyo RA. Generic and disease-specific measures in assessing health status and quality of life. Med Care. 1989;27(3):S217-32.

90. Janssens A, Thompson Coon J, Rogers M, et al. A systematic review of generic multidimensional patient-reported outcome measures for children, part I: descriptive characteristics. Value Health. 2015;18(2):315-333. doi: 10.1016/j.jval.2014.12.006.

91. Janssens A, Rogers M, Thompson Coon J et al. A systematic review of generic multidimensional patient-reported outcome measures for children, part II: evaluation of psychometric performance of English-language versions in a general population. Value Health. 2015;18(2):334-345. doi: 10.1016/j.jval.2015.01.004.

92. Hickey AM, Bury G, O'Boyle CA, et al. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33.

93. Wolpert M. Do patient reported outcome measures do more harm than good? BMJ. 2013;346:f2669. doi: 10.1136/bmj.f2669.

94. Griffiths C, Armstrong-James L, White P, et al. A systematic review of patient reported outcome measures (PROMs) used in child and adolescent burn research. Burns. 2015 Mar;41(2):212-24. doi: 10.1016/j.burns.2014.07.018.

95.Tadić V, Rahi JS. One size doesn't fit all: time to revisit patient-reported outcome measures (PROMs) in paediatric ophthalmology? Eye (Lond). 2017;31(4):511-518. doi: 10.1038/eye.2016.316.

96. Downing J, Ojing M, Powell RA, Ali Z, Marston J, Meiring M, Ssengooba J, Williams S, Mwangi-Powell F, Harding R. A palliative care outcome measure for children in sub-Saharan Africa: Early development findings. European Journal of Palliative Care 2012;19(6):292-294.

97. Downing J. International Observatory on end-of-life Care (IOELC) Seminar 24th October 2017. Lancaster https://dtu-panopto.lancs.ac.uk/Panopto/Pages/Viewer.aspx?id=505d0b14-129f-4b64-9d06-408abc60bf2c (accessed 20th May 2018).

98.Van Leeuwen M, Husson O, Alberti P. Understanding the quality of life issues in survivors of cancer: towards the development of an EORTC QoL cancer survivorship questionnaire. Health Qual Life Outcomes. 2018;16(1):114. doi: 10.1186/s12955-018-0920-0.

99. Evans CJ, Benalia H, Preston NJ, Grande G, Gysels M, Short V, Daveson BA, Bausewein C, Todd C, Higginson IJ; MORECare. The selection and use of outcome measures in palliative and end-of-life care research: the MORECare International Consensus Workshop. J Pain Symptom Manage. 2013;46(6):925-937. doi: 10.1016/j.jpainsymman.2013.01.010.

100. Antunes B, Harding R, Higginson IJ; EUROIMPACT. Implementing patient-reported outcome measures in palliative care clinical practice: a systematic review of facilitators and barriers. Palliat Med. 2014;28(2):158-175. doi: 10.1177/0269216313491619.

101. Holtrop JS, Rabin BA, Glasgow RE. Dissemination and Implementation Science in Primary Care Research and Practice: Contributions and Opportunities. J Am Board Fam Med. 2018;31(3):466-478. doi: 10.3122/jabfm.2018.03.170259.

102. Bausewein C, Simon ST, Benalia H, Downing J, Mwangi-Powell FN, Daveson BA, Harding R, Higginson IJ; PRISMA. Implementing patient reported outcome measures (PROMs) in palliative care--users' cry for help. Health Qual Life Outcomes. 2011;20(9):27. doi: 10.1186/1477-7525-9-27.

103.Wells S, Tamir O, Gray J, et al. Are quality improvement collaboratives effective? A systematic review.BMJ Qual Saf. 2018;27(3):226-240. doi: 10.1136/bmjqs-2017-006926.

104. Drahota A, Meza RD, Brikho B et al. Community-Academic Partnerships: A Systematic Review of the State of the Literature and Recommendations for Future Research. Milbank Q. 2016 Mar;94(1):163-214. doi: 10.1111/1468-0009.12184.

105. Recinos PF, Dunphy CJ, Thompson N, et al. Patient Satisfaction with Collection of Patient-Reported Outcome Measures in Routine Care. Adv Ther. 2017;34(2):452-465. doi: 10.1007/s12325-016-04

Key messages of chapter 4

Instruments used to measure outcomes in paediatric palliative care are seldom self-reported by children or adolescents.

Psychometric properties of those instruments are poorly documented.

No outcome measurement instrument combining self- and proxy-report, nor combining children’s and parents’ quality of life are currently available for a paediatric palliative care population.

However, the APCA childrens palliative outcome scale (CPOS) seems to be a promising tool, but its psychometric properties are not known. It was pilot-tested in 3 African countries and is only available in English. For that reason, we decided to conduct a pilot-test of the CPOS in French with the aim to assess its face and content validity, its acceptability by children and parents, and its feasibility of use by paediatric liaison teams.

CHAPTER 5. FACE AND CONTENT-VALIDITY, ACCEPTABILITY AND FEASIBILITY OF THE FURTHER DEVELOPED CHILDREN'S PALLIATIVE OUTCOME SCALE (CPOS-2): A PILOT-STUDY

This chapter was published as an article under the title:

Friedel M. Brichard B, Boonen S, Tonon C, de Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse JM, Aujoulat I. Face and content validity, acceptability and feasibility of the adapted version of the Children's Palliative Outcome Scale (CPOS-2): a qualitative pilot study. J Palliat Med. 2020;10.1089/jpm.2019.0646. [published online ahead of print, 2020 Jul 15].

ABSTRACT

Background: Instruments in French used to measure outcomes in paediatric palliative care are lacking.

Objective: To assess the face and content validity, acceptability and feasibility of a French version of the Children's Palliative Outcome Scale (CPOS).

Design: After forward-backward translation of the 12-item English CPOS to French, we conducted a qualitative pilot study.

Setting/subjects: Fourteen children and adolescents (8-18 years) with life-limiting or life-threatening conditions cared for at home, in hospital or in respite care services, 19 parents, and 9 members of 4 paediatric liaison teams (PLTs) providing palliative care in a Belgian francophone region.

Measurements: During semistructured interviews among children and parents, we used the CPOS, the Schedule for the Evaluation of Individual Quality of Life interview guide (SEIQoL) and the Quality of Life in Life-threatening Illness-Family Carer questionnaire (QOLLTI-F), in addition to three expert meetings with PLTs.

Results: No families refused to participate. All children with verbal capacities chose to be interviewed in the presence of their parents and a PLT member. The children valued being given the opportunity to share their experiences. New QOL dimensions pertaining to social, emotional, and administrative health-care related issues were added to the original version of the 12-item CPOS, leading to a 22-item CPOS-2.

Conclusions: The CPOS-2 was perceived as relevant and easy to use by the principal stakeholders. Our study paves the way for a large-scale field study assessing its psychometric characteristics and its implementation in routine clinical care.

Introduction

The main objective of paediatric palliative care (PPC) is to achieve the best quality of life (QoL). (1,2) Measuring QoL in children facing life-limiting or life-threatening conditions (LLC/LTC) is challenging due to the subjective aspect of the construct, heterogeneity in ages and diseases of the children, the diversity of PPC models and interventions and the legal and ethical requirements that must be observed when involving children in research. (3-7)

It has been recommended to develop a short, age-adapted, multidimensional patient-centered outcomes measure. This measure should include children's and parents' perspectives and should been perceived as acceptable by them. Furthermore, this measure should be feasible to use for clinicians. (3-5, 8-10) In accordance with this recommendation, Downing et al. developed the Children's Palliative Outcome Scale (CPOS) (4,11), which is a short 12-item instrument combining self (child) and proxy (parents) reports regarding health status, health-related QoL and QoL. It covers 4 dimensions: physical and psychological symptoms, information received, advance care planning and self-efficacy.

In Belgium, paediatric liaison teams (PLTs) are ensuring continuity of care for children facing LLC/LTC across various care pathways. (12) Their activities as well as the profile of children they care for have been described in previous papers. (13,14)

The aim of the pilot study was to assess the face and content validity, acceptability and feasibility of a French version of the CPOS among children facing LLC/LTC, their parents and PLTs in the French part of Belgium.

Design

This qualitative pilot study is part of the larger Belgian MOSAIK study (Move to Open Shared Advanced Interventions for Kids with life-limiting conditions) and aims to develop a measurement instrument to assess children's and parents' quality of life while receiving paediatric palliative care.

To comply with standards on ethical issues in palliative care research (15,16) and recommendations (17-19) on the development of a measurement instrument, especially regarding the assessment of face and content validity, we opted for an iterative and collaborative approach that involved the main stakeholders (children, parents, and teams) in the research process.

Data were collected through semi structured interviews conducted by the first author (MF) with children and their parents as well as three expert meetings with representatives of PLTs. Children and adolescents aged 8 to 18 who were facing an LLC/LTC and their parents were included in the study. Families were recruited through one PLT in the French part of Belgium, and an interview was planned according to their preferences of time and place.

Instruments: A forward/backward translation of the CPOS was conducted based on guidance from Antunes et al. (20) and de Vet et al. (17)

Face validity is defined as “the degree to which a measurement instrument indeed looks as though it is an adequate reflection of the construct to be measured.” (17, p.155) To assess the face validity of the CPOS for measuring children’s and parental QoL, the researchers relied on previous work by Downing et al. (4) and discussed each translated item in French with paediatric liaison teams.

Content validity is defined as “the degree to which the content of a measurement instrument is an adequate reflection of the construct to be measured.” (17, p.155) To assess the content validity of the CPOS for measuring children’s QOL, the Schedule for the Evaluation of Individual Quality of Life (SEIQoL) was used. (21,22) This semi structured interview guide aims to elicit self-generated domains of QoL as expressed by the children themselves. The SEIQoL was validated for use in children with diabetes (46), children with cerebral palsy (47), and adults with cancer (45). As this population was not the one included in our study, we considered the SEIQoL only as a guide. To verify the content validity of the CPOS for measuring parental QoL, the Quality of Life in Life Threatening Illnesses-Family Carer questionnaire (QOLLTI-F) was administered to parents. (23)

All instruments were pretested with 2 children aged 8 and 10 to ensure the comprehensibility of the questions, but these children were not included in the sample size. Children, parents and an expert panel composed of paediatric liaison team members discussed the relevance and comprehension of each item included in the further developed French CPOS.

-Table 11 shows an overview of the instruments and the interview guide used in the study.

Table 11. Overview of the instruments and the interview guide used in the pilot-study

Name	**Measure**	**Aim of the instrument**	**Context**	**Validation**	**Language**
Children's palliative outcome scale **CPOS** (4,11)	Multidimensional standardized self- and proxy-reported outcomes measure 12 questions	To measure outcomes of paediatric palliative care	Uganda, Kenya, South Africa 302 children, 299 family carers	Pilot-tested, Responsivity, acceptability, feasibility, face and content validity tested (not yet published)	English
Scheduled evaluation of individual quality of life S**EIQoL** (21,22)	Semi-structured interview guide Degree of importance/satisfaction for each of the 5	To generate self-reported domains of quality of life	Adult oncology (45) Children with Diabetes (46) with Cerebral Palsy (47)	Validation complete	French

	self-generated domains				
Quality of life family carers **QOLLTI-F** (23)	Multidimensional Standardized parental quality of life Scale 7 domains 16 questions	To measure parental quality of life	Used in PPC context (48-50)	Validation complete version 2 French	French

Acceptability of the CPOS was assessed through the ratio of the number of families willing to participate in an interview versus the number that refused, the duration of the interview and the families' comments on the acceptability of each item included in the CPOS.

The feasibility of using the CPOS was evaluated by members of the PLT who participated in the interviews through 3 expert panel reviews and think aloud techniques that captured their individual spontaneous reflections following each family interview. Expert panel reviews and think aloud techniques gave the researcher insights into the team's cognitive processes after having used the CPOS. Think aloud techniques are well-known methods used in health research. (24-26)

The interviews were led by the first author (MF) and, according to the children's preferences, included their parents and/or a member of the PLT to enhance the children's sense of security and to involve the PLT in the use of the CPOS.

Data analysis: All interviews were audio-taped and transcribed verbatim for analysis based on an integrated approach of categorization. (27,28) An initial list of codes reflecting the pre-existing domains identified in the CPOS and the QOLLTI-F were used as a framework to organize the thematic categories inductively created through the interviews. The content validity of newly generated categories was discussed during 3 expert meetings with 9 members of 4 different PLTs, including 2 paediatricians (BB and CF), 6 paediatric nurses specializing in paediatric palliative care (CT, BT, DB, MM, SB and MS), one psychologist (MJ) and 2 academic researchers (MF and IA).

Results

Fourteen children/adolescents, 19 parents, 9 PLT members and 2 researchers participated in the pilot study, which was conducted from June 2017 to July 2018. Table 12 shows the characteristics of the participating subjects (n=42)

Table 12. Characteristics of the participating subjects (n=42)

1. Characteristics of children/adolescents		**Total (n=14)**
Gender	male, female	n=7 (50%), n=7 (50%)
Age	8-10 years	n=7 (50%)
	11-13 years	n=1 (0,8%)
	14-16 years	n=2 (14,3%)
	17-18 years	n=4 (28,6%)
Life-limiting conditions:	Neurological disease	n=8 (57,1%)
	Oncological disease	n=6 (42,9%)
(For confidentiality reasons, we could not further specify the type of neurological and oncological diseases)		
Cognitive and verbal capacity *(child's capacity, as perceived by their parents and HCP, to understand questions from the CPOS, SEIQoL)*		
	Good	n=6 (42,9%)
	Moderate	n=3 (21,4%)
	Low	n=5 (35,7%)
Type of care during time of interview		
	Paediatric liaison care	n=8 (57,1%)
	Respite care	n=4 (28,6%)
	Hospital care	n=2 (14,3%)
Place where the interviews were conducted		
	Home	n=7 (50%)
	Hospital	n=7 (50%)
Number of children		
	who completed CPOS self-report	n=9 (64,3%)
	who self-generated QoL domains	n=8 (57,1%)
2. Characteristics of parents		**Total (n=19)**
Mother		n=12 (63,2%)
Father		n=7 (36,8%)
Separated		n=6 (31,6%)
Number of parents		
	who completed CPOS proxy report	n=14 (73,7%)
	who completed QOLLTI-F	n=19 (100%)

3. **Characteristics of Health Care professionals**	**Total (n=9)**
Nurses	n=6
Physicians	n=2
Psychologist	n=1

Legend

CPOS= Children’s Palliative Outcome Scale;

QOLLTI-F= Quality of Life in Life-threatening Illness - Family carer

The adapted French version of the 22-item CPOS-2 and its translated version in English can be found in the Appendices.

The results are illustrated with quotations either from the interviews (I) with the families or from the expert meetings (M).

-Table 13 provides a comparison of the dimensions covered by the original CPOS with those that emerged from the interviews with children and their parents-

Table 13. Comparison of the dimensions covered by the original CPOS with those emerged from interviews with children and their parents

	Dimensions of the original APCA-CPOS (Downing, 2018) and adapted or added questions	**New dimensions that emerged from interviews with children and their parents (using CPOS, SEIQoL, QOOLTI-F) leading to additional questions**
I. Dimensions CHILD	1. Physical elements (pain Q1 and other symptoms related to the body Q2, feeding) ** question regarding feeding changed* Q3 + one question regarding sleeping added Q8	5. Interactions (family, friends, pets) +one question regarding interactions with peers, friends added Q10 +one question regarding relationships with the family added Q11
	2. Psychosocial elements (sadness Q4, joy Q5) + one question regarding worries added Q9 + one question on “feeling to be loved” added Q12	6. Death and dying No question was added on death and dying, because theme considered to be initiated by the child/adolescent himself and not by a stanardized questionnaire.
	3. Play ** question on playing changed (broadened to “having fun”)* Q6	
	4. Information received Q7	
II. Dimensions PARENTS	1. Psychosocial elements **one question changed to reflect the impact of worries on daily life* Q13 + one question on overall perceived quality of life added Q22	5. Help and support received +one question added on help and support received Q18
	2. Information received Q15	6. Burden of administrative procedures and financial issues +one question added on burden of administrative procedures Q19 +one question added on financial issues Q20
	3. Planning for the future Q16	
	4. Self-efficacy (Feeling confident/able to share feelings Q14 and to take care of the child Q17) + one question on burden of care added Q21	

Legend

CPOS= Children’s palliative outcome scale (multidimensional standardized scale)
SEIQoL= scheduled evaluation of individual Quality of life (semi-structured interview guide)
Q= Question. The number of the question correspond to the adapted 22 items CPOS in French.

Face and content validity of the CPOS

Regarding children's QoL, all 4 dimensions included in the original CPOS were **confirmed**, namely, physical and psychosocial dimension, playing, and the information received.

Two of those dimensions (physical/psychosocial dimensions) led to three more specific questions. One question addresses the **quality of sleeping (Q8 of the adapted 22-item CPOS)**:

> *"I always had nightmares that woke me up… I'm so frightened. (…). It awakens me at once, and my heart goes so fast that… When it happens, I stand up and I'm shouting everywhere." (Adolescent girl. Interview I3)*
>
> *"He struggles with the lying position in his bed. I don't know if it's a physical problem. Sometimes he doesn't sleep at all or he's up already at 5 am." (Father of a young boy. I8)*

The second question focuses on **the possibility of sharing worries with somebody (Q9)**:

> *"At night, I am telling my teddy bear all that worries me. "(Young girl, I5***)**
>
> *"No, I keep that inside of me." (Adolescent boy, I10)*
>
> *"Indeed, that's the problem! He will shout, criticize, and yell instead of explaining calmly to the psychologist… It seems he doesn't know how to express himself." (Mother of a young boy, I12)*

The third question looked at the **feeling of being valued and loved (Q12):**

A young girl with a progressive illness suggested adding a question about happiness because, according to her, this would be the most important one:

> *"You should ask if I am happy!"* adding, *"I am happy because I always have my parents by my side!" (young girl, I5)*

This was in line with the father of a young boy (I8), who suggested that the child's perception of being loved contributed significantly to his QoL.

In relation to the dimension of physical symptoms, the initial question concerning the amount of **feeding (Q3)** was changed to include any type of worry, not just about the amount of food.

In relation to the dimension of **playing**, the question **(Q6)** was broadened to all activities and sources of fun and joy in order to be more adapted to adolescents.

> *"I love playing sports, moving all around. I can't stand to keep calm." (Adolescent girl. I3)*

> *"He loves having parties, dancing, being with others and having fun." (Mother of an adolescent boy. I6)*

In addition to the confirmed dimensions addressed by the original 12-item CPOS, **one new dimension emerged regarding social relations**, which was perceived as very important by all children and adolescents and led to 2 additional questions. One question focuses on **friends (Q10)** and one on their **relations with their family (Q11).**

> *"I don't have a lot of friends. I'm missing that. (…) It's complicated to have some. I don't succeed at making them." (Adolescent boy, I6)*

> *"I really love my family. (…) I'm so happy when I'm going to see my aunt." "It's ok between me and my brother." (Adolescent boy, I6)*

In summary, all the dimensions of the original CPOS were confirmed, and questions on the newly generated dimension of social relations were added, leading to 12 questions addressing the children's QoL.

Regarding parental quality of life, all dimensions from the original CPOS were confirmed, namely, psychosocial elements, the need to plan the future, information received and the sense of self-efficacy. Within the psychosocial dimension, one question about **worries** was changed to focus on the impact that such worries have on daily life **(Q13)**:

> *"There was always this fear inside of us. We really couldn't sleep, couldn't eat, nothing. A permanent anxiety." (Mother of an adolescent girl. I3)*

Another question was added to the **overall perceived QoL (Q22)**, raising parental awareness of self-care and potential respite needs:

> *"My quality of life is almost good, although we are struggling." (Mother, I8)*

Within the dimension on self-efficacy, one question was added in relation to the perceived **burden of taking care** of their child **(Q21)**:

> *"Yes, it's a psychological burden. An enormous mental load. (…) The whole day, we are thinking about which medication we have to give him." (Father of a young boy. I8)*

Two new dimensions of parental QoL emerged. The first dimension addressed the type of **support** perceived:

> *"Dr X is open from Monday to Sunday and from Sunday to Monday. You can even call her late in the evening. … Dr X, you are super! And at that moment you feel supported. When they told us that we could always call her, ahhhhh, I had shivers up and down. Really!" (Mother of a young boy. I2)*

The second dimension focused on **administrative** issues **(Q19)**:

> *"Those files for the administration, these are crazy proceedings. We must ask Peter, Paul and Jack every time we need something. Stop now! All these papers take so much energy. (…) Even if they knew who my son his, we had to refile the paperwork, to start from the beginning, again!" (Parents of a young boy, I2)*

The relevance of one question included in the original CPOS, "Did you receive help and advice to **prepare for the future** of your child?", was questioned by some parents because it did not seem to fit with their current concerns, especially when they were overwhelmed by the present situation:

> *"Which future? We must learn how to live day after day, so this question isn't logical!" (Mother, I4)*

Conversely, for one parent of a severely handicapped child, preparing for the future was paramount:

> *"The biggest fear since she was 7 years old is: What will she become when we won't be there anymore? D. will always be dependent on somebody…." (Mother of a young girl. I7)*

We decided to remove that question because half of the population of children followed up by PLTs face neurological conditions.

Furthermore, we observed that one emerging dimension on death and dying could not be made operational in a questionnaire. In our study, one adolescent boy raised this topic by expressing how much he would like to see a dead street neighbor, yet he was facing a categorical refusal from his mother:

> *"Oh, come on, I'm in good spirits. I want to go outside and see the dead. I won't be sad, I promise you. Please! At least, I'd seen a dead person in my life. (...) Everybody will go and see him, why not me? Why are you afraid?" (Adolescent boy, I6) "And what will I do If you will be shocked?" (Mother, I6) "I know what a dead person*

> *looks like. I'm not afraid of death, because my grandmother already died." (Adolescent boy, I6)*

Although this dimension was considered very important for PLTs, they decided not to add a related question to the CPOS, arguing that good practices would recommend waiting for children to initiate any questions on death and dying rather than broaching this topic directly in a questionnaire.

In summary, all the dimensions of the original CPOS were confirmed, and new dimensions, namely, support received and burden of administrative procedures and financial issues, were added to the adapted CPOS, leading to 10 questions addressing parental QoL.

Acceptability of the CPOS as perceived by the children and their parents

All invited families (n=14) agreed to be interviewed, and the duration of interviews was considered acceptable by them (mean of 25 minutes, ranging from 12 to 75). Positive feelings about being interviewed were expressed by all children and parents:

> *"It was good*." (I3) "*I liked it*." (I5) "*I appreciated this moment*." (I6) "*The questions are easy to understand, and it was easy to respond*." (I13) Their parents also expressed positive reactions: *"Very good, very pleasant." (Father I6) "Nothing else to say. It's not intrusive at all." (Mother, I8) "I found that very good. That's why I always agree to participate." (Mother, I7)*

Furthermore, parents said that it allowed them to identify what helped them and to express to one another their mutual sense of gratitude:

> *"For 75%, it was her fathers' merit, because he was always there for her, being a huge force. And this inner force inside him never diminished. Sometimes I was asking myself: how can he stand all this? Day and night, day and night, day and night to care for her and to care for us, to clean the house, to transport here and there with the car…" (Mother, I3)*

Feasibility of using the CPOS as perceived by the PLTs

As detailed in the methods section, the PLTs were involved in the entire process of the study. They perceived the CPOS as a useful tool that shined a "*warning light*" on overlooked domains, helping them to individualize and to improve the care provided. For instance, a teenager expressed during the interview that he was longing for more contacts with his peers. The liaison nurse who had conducted the interview consequently made suggestions for him to take part in monthly gatherings of an inclusive scout movement. One month later, the mother reported that her son greatly appreciated this new activity and felt less depressed.

Discussion

Our study confirmed the face validity and strengthened the content validity of the primary dimensions included in the original 12-item CPOS, the acceptability of the CPOS to families and its feasibility of use for caregivers. Questions related to meaningful domains as expressed by the children, parents and PLT members were added to the original instrument, ultimately leading to a 22-item tool that explores children's and parents' QoL. Several issues can be discussed.

Challenges identifying meaningful outcomes to be included in a measurement instrument

Outcome measurement instruments should be derived from meaningful outcomes, defined by the patients themselves. Many studies exploring meaningful outcomes in PPC rely only on parents' and/or professionals' perspectives, which is a first step to assess the content validity of an instrument. (34, 35) However, directly identifying among what matters most to children, that is, measuring individual QoL, is recommended but challenging. Following recommendations by previous studies (3,4,10,17,36,37) and to further strengthen the evidence of the CPOS to assess meaningful QoL domains, we used the SEIQoL interview guide (21,22), through which children spoke out, elicited self-identified QoL domains and appreciated being consulted. However, in a PPC context, where many children lack cognitive capacity or are very young and consequently have difficulties expressing themselves, PLTs perceived the SEIQoL to be too complicated to be routinely used.

Furthermore, our results are consistent with a systematic review that identified 5 domains of meaningful outcomes in PPC: physical, psychological, psychosocial, existential, and information access. (29) More specifically, the importance of relationships for children found in our study was highlighted in two further studies. (30,31) The complexity of those relationships could be analyzed through the concept of normalcy, which was found to be paramount in one study including children with brain tumors. (32) Children want to have relationships, but on the other hand, they are suffering due to feeling different than their peers. Furthermore, existential dimensions should be explored through hopes, worries, meanings and life perspectives. (33) rather than using words such as religion or spirituality.

Acknowledging the ethical and psychological requirements when including children in research

The necessity of collecting data directly from children facing LLC/LTC entails adapting methods that are appropriate for their cognitive and age-specific development and ensuring a safe space by valuing their preferences. (38,39) Accordingly, in our study, we respected children's choice to be interviewed in the presence of their parents, even if this might introduce social desirability bias. However, reporting non-invitation rates and selection bias whenever possible may aid in the interpretation of research findings. (40) The ethical imperative that research in PPC must benefit children (37) was acknowledged by PLTs, who conducted the interviews and offered individualized care, responding immediately to the needs expressed by children during the interview.

Limitation and strength

Comparison of the data among the different children has not been possible because of the evolutive nature of the questionnaire. Although this study is monocentric, the relevance and comprehension of each item has been discussed by an expert panel composed of multicentric paediatric liaison teams. Potential social desirability bias could have been a factor on account of the children having been interviewed in the presence of their parents and their usual PLT. Furthermore, our sampling did not represent the wide variety of diseases labeled LLC/LTC, potentially restricting the emergence of other self-identified domains of QoL. Finally, the perspectives of very young children or those with serious verbal and cognitive deficiencies could not be considered.

Nevertheless, to our knowledge, this is the first study to test a French version of the children's palliative outcome scale in a European context of paediatric palliative care. Moreover, triangulation of data sources contributed to the content validity of the CPOS. Finally, the collaborative approach planted the seeds for a sustainable implementation process of the instrument in clinical care, as recommended. (19,41-44)

Conclusion

Face and content validity, acceptability and feasibility of a French version of the CPOS were assessed by integrating the perspectives of 14 children, 19 parents and 9 PLT members, leading to three major results. First, using a specific and unique procedure, a revised 22-item CPOS-2 was developed. Second, this instrument was perceived as relevant and easy to use by the principal stakeholders. Third, this study paves the way for an additional large-scale multicentric field study in which the psychometric characteristics as well as the challenges linked to its implementation in routine clinical care will be studied.

Research Ethics and patient consent

The pilot study was approved by ethics committee number 403, attached to University Hospital Saint Luc, Brussels, Belgium under the name MOSAIK 2017/16MAI/271 and registered under the number B403201732735. The ethics committee declared that the study followed Good Clinical Practice (GCP). Informed written consent was obtained from each parent who agreed to be interviewed. Whenever the child/adolescent was able to understand or communicate, he or she was invited to sign an adapted age-appropriate written informed assent form.

Acknowledgements

We would like to express our deeply felt gratitude to each child/adolescent and parent who agreed to share their experiences within this study; the African Palliative Care Association for their permission to use the APCA-Children's Palliative Outcome Scale; Prof. Robin Cohen for permission to use the Quality of Life in Life-threatening Illnesses-Family Carer (QOLLTI-F) questionnaire; Mandana Mehta, Pauline De Becker and Claire Mougenot, who contributed to the linguistic and semantic forward/backward translation of the CPOS; and Sophie Agboli, Thérèse Van Durme, Nathalie Chaidron, Valérie Dory and Nataly Filion for their informal support. For their unwavering support since the beginning, we would like to thank the Fonds de Soutien Marguerite-Marie Delacroix, who funded the study in the frame of a PhD thesis conducted by MF.

References

1. World Health Organization. Definition of Palliative care for Children. https://www.who.int/cancer/palliative/definition/en/ (1998, accessed 5 Jan 2019)

2. Abu-Saad Huijer H, Benini F, Craig F, et al. Steering Committee of the EAPC task force on palliative care for children and adolescents: IMPaCCT. Standards for paediatric palliative care in Europe. *Eur J Palliat Care* 2007; 14(3): 109-114.

3. Harding R, Wolfe J, Baker JN. Outcome Measurement for Children and Young People. *J Palliat Med* 2017;20(4): 313.

4. Downing J, Namisango E, Harding R. Outcomes measurement in paediatric palliative care: lessons from the past and future developments. *Ann Palliat Med* 2018;7, Suppl 3: S151-S163.

5. Knapp C, Madden V. Conducting outcomes research in paediatric palliative care. *Am J Hosp Palliat* Care 2010;27(4): 277-281.

6. Huang IC, Wen PS, Revicki DA, et al. Quality of Life Measurement for Children with Life-Threatening Conditions: Limitations and a New Framework. *Child Indic Res* 2011;4(1): 145-160.

7. Peake JN, Beecham E, Oostendorp LJM, et al. Research barriers in children and young people with life-limiting conditions: a survey. *BMJ Support Palliat Care* 2018; Jul 31. pii: bmjspcare-2018-001521.

8. Weaver MS, Darnall C, Bace S, et al. Trending Longitudinal Agreement between Parent and Child Perceptions of Quality of Life for Paediatric Palliative Care Patients. *Children (Basel)* 2018;4,8. pii: E65.

9. Wolfe J, Siden H. Research in paediatric palliative care. In: Goldman A, Hain R, Liben S (eds): Oxford Textbook of Palliative Care for Children. Oxford: Oxford University Press, 2012, pp. 441-457.

10. Harding R, Chambers L, Bluebond-Langner M. Advancing the science of outcome measurement in paediatric palliative care. *Int J Palliat Nurs* 2019;25(2): 72-79.

11. African Palliative Care Association. The APCA African Children's Palliative Outcome Scale (APCA CPOS). Manual. 2012.

12. Royal Decree defining the mission of the paediatric liaison care teams in Belgium. https://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html (2010, accessed 5 Jan 2019)

13. Friedel M, Brichard B, Fonteyne C, et al. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. *BMC Palliat Care* 2018;17(1): 77.

14. Friedel M, Gilson A, Bouckenaere D, et al. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. *BMJ Paediatrics Open* 2019;3: e000547.

15. Higginson IJ, Evans CJ, Grande G, et al. MORECare. Evaluating complex interventions in end of life care: the MORECare statement on good practice generated by a synthesis of transparent expert consultations and systematic reviews. *BMC Med* 2013; 11: 111.

16. Gysels M, Evans CJ, Lewis P. MORECare research methods guidance development: Recommendations for ethical issues in palliative and end-of-life care research. *Palliat Med* 2013; 27;13: 908-917.

17. Measurement in Medicine, 1st ed. De Vet HCW, Terwee CB, Mokkink LB (eds). Cambridge University Press, Cambridge, 2016.

18. Bausewein C, Daveson BA, Currow DC, et al. EAPC White Paper on outcome measurement in palliative care: Improving practice, attaining outcomes and delivering quality services - Recommendations from the European Association for Palliative Care (EAPC) Task Force on Outcome Measurement. *Palliat Med* 2016;30: 6-22.

19. Abma TA, Pittens CA, Visse M et al. Patient involvement in research programming and implementation. A responsive evaluation of the Dialogue Model for research agenda setting. *Health Expect* 2014;18: 2449-2464.

20. Antunes B, Daveson B, Ramsenthaler C, and al. on behalf of the POS Development Team King's College London, Cicely Saunders Institute, Department of Palliative Care, Policy and Rehabilitation. Kings College London, London, 2012.

21. O'Boyle CA, McGee H, Hickey A, et al. Individual quality of life in patients undergoing hip replacement. *Lancet* 1992; 2;339(8801): 1088-1091.

22. Hickey AM, Bury G, O'Boyle CA, et al. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. *BMJ* 1996; 6;313(7048): 29-33.

23. Cohen R, Leis AM, Kuhl D, et al. QOLLTI-F: measuring family carer quality of life. *Palliat Med* 2006; 8: 755-767.

24. Zhang L, Gallagher R, Lowres N, et al. Using the 'Think Aloud' Technique to Explore Quality of Life Issues During Standard Quality-of-Life Questionnaires in Patients with Atrial Fibrillation. *Heart Lung Circ* 2017;26(2): 150-156.

25. Verkuyl M, Hughes M, Fyfe MC. Using Think Aloud in Health Assessment: A Mixed-Methods Study. *J Nurs Educ* 2018;57(11): 684-686.

26. Lundgrén-Laine H, Salanterä S. Think-aloud technique and protocol analysis in clinical decision-making research. *Qual Health Res* 2010;20(4): 565-75.

27. Miles MB, Huberman M. Qualitative data analysis. A sourcebook of new methods. Thousand Oaks: SAGE, London, 1994.

28. Bradley EH, Curry LA, Devers KL. Qualitative data analysis for health services research: developing taxonomy, themes, and theory. *Health Serv Res* 2007; 42: 1758-1772.

29. Namisango E, Bristowe K, Allsop MJ, et al. Symptoms and Concerns Among Children and Young People with Life-Limiting and Life-Threatening Conditions: A Systematic Review Highlighting Meaningful Health Outcomes. *Patient* 2019;12(1): 15-55.

30. Ambresin AE, Bennett K, Patton GC, et al. Assessment of Youth-friendly health Care: a systematic review of indicators drawn from young peoples' perspectives, *J Adolesc Health* 2013; 52: 670-681.

31. Ghirotto L, Busani E, Salvati M et al. Researching children's perspectives in paediatric palliative care: A systematic review and meta-summary of qualitative research. *Palliat Support Care* 2019;17(1): 107-118.

32. Beecham E, Langner R, Hargrave D et al. Children's and Parents' Conceptualization of Quality of Life in Children with Brain Tumors: A Meta-Ethnographic Exploration. *Qual Health Res* 2019;29(1): 55-68.

33. Barton KS, Tate T, Lau N et al. "I'm Not a Spiritual Person." How Hope Might Facilitate Conversations About Spirituality Among Teens and Young Adults with Cancer. *J Pain Symptom Manage* 2018;55(6): 1599-1608.

34. Friedel M, Aujoulat I, Dubois AC, et al. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. *Paediatrics* 2019;143(1).

35. Coombes LH, Wiseman T, Lucas G, et al. Health-related quality-of-life outcomes measures in paediatric palliative care: a systematic review of psychometric properties and feasibility of use. *Palliat Med* 2016;30(10): 935-949.

36. Huang IC, Shenkman EA, Madden VL, et al. Measuring quality of life in paediatric palliative care: challenges and potential solutions. *Palliat Med* 2010;24(2): 175-182.

37.Baker JN, Levine DR, Hinds PS et al. Research Priorities in Paediatric Palliative Care. *J Pediatr* 2015;167(2): 467-470.e3.

38. Rahimzadeh V, Bartlett G, Longo C et al. Promoting and ethic of engagement in paediatric palliative care research. *BMC Palliat Care* 2015;16(14): 50.

39. Bluebond-Langner MH. Involving Children with Life-Shortening Illnesses in Decisions about Participation in Clinical Research: A Proposal for Shuttle Diplomacy and Negotiation. In: Kodish ED (ed.): Ethics and Research with Children. A case-based approach. New York: Oxford University Press, 2005.

40. Crocker JC, Beecham E, Kelly P et al. Inviting parents to take part in paediatric palliative care research: a mixed-methods examination of selection bias. *Palliat Med* 2015;29(3): 231-240.

41. Etkind SN, Daveson BA, Kwok W, et al. Capture, transfer, and feedback of patient-centered outcomes data in palliative care populations: does it make a difference? A systematic review. *J Pain Symptom Manage* 2015;49(3): 611-624.

42. Antunes B, Harding R, Higginson IJ et al. Implementing patient-reported outcome measures in palliative care clinical practice: a systematic review of facilitators and barriers. *Palliat Med* 2014*;*28: 158-175.

43. Bausewein C, Simon ST, Benalia H et al. Implementing patient reported outcome measures (PROMs) in palliative care - users' cry for help. *Health Qual Life Outcomes* 2011;20(9): 27.

44. Pinto C, Bristowe K, Witt J et al. Perspectives of patients, family caregivers and health professionals on the use of outcome measures in palliative care and lessons for implementation: a multi-method qualitative study. *Ann Palliat Med* 2018;7(Suppl 3): S137-S150.

45. Rodary S, Dauchy C, Beauvallet M et al. Intérêt de la mesure individualisée de la qualité de vie en oncologie: utilisation du SEIQoL chez des patients traités pour tumeur neuro-endocrine. *Revue Francophone de Psycho-Oncologie* 2005;4(1): 36-44.

46. Wagner J. Acceptability of the Schedule for the Evaluation of Individual Quality of Life-Direct Weight (SEIQoL-DW) in youth with type 1 diabetes. *Qual Life Res* 2004;13(7): 1279-1285.

47. Vinson J, Shank L, Thomas PD et al. Self-generated Domains of Quality of Life in Children with and Without Cerebral Palsy. *J Dev Phys Disabil* 2010;22(5): 497-508.

48. Groh G, Borasio GD, Nickolay C et al. Specialized paediatric palliative home care: a prospective evaluation. *J Palliat Med 2013;*16,12: 1588-1594.

49. Vollenbroich R, Duroux A, Grasser M et al. Effectiveness of a paediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med* 2012*;*15(3): 294-300.

50. Bradford N, Young J, Armfield NR et al. A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. *J Telemed Telecare* 2012*;*18(8): 438-442.

Key messages of chapter 5

The original 12-item CPOS, after cross-cultural translation into French, was pilot-tested in one center. The aim was to assess the face and content validity of the CPOS, its acceptability among children and their parents and its feasibility for use by paediatric liaison teams during routine clinical care. During semi-structured interviews with families, children self-elicited domains which they perceived to impact their quality of life. Those emerging domains were compared with the dimensions included in the CPOS. The pilot-test led to a further developed 22-items CPOS (CPOS-2). Following guidelines, the further step was to conduct a wider field-test among all six paediatric liaison teams to describe the psychometric properties of the further developed 22-item CPOS, to provide an overview of children's and parents' quality of life and to document the experiences of teams when using this instrument.

CHAPTER 6. A FIELD-STUDY

6.1. RELIABILITY AND CONCURRENT VALIDITY OF VERSION 2 OF THE CHILDREN'S PALLIATIVE OUTCOME SCALE (CPOS-2)

ABSTRACT

Background and objectives: Promoting Quality of Life (QoL) is the main goal of paediatric palliative care, but assessing QoL remains an important challenge in this particular setting. Instruments for measuring QoL in paediatric palliative care are lacking. A promising outcome measurement instrument, available in English, called the children's palliative outcome scale (CPOS), with 12 items, has been previously pilot-tested for its face/content validity, its feasibility and acceptability among families, resulting in a 22-item instrument. The CPOS-2 includes a self- (child) and a parent's (proxy) report and assesses children's and parents' QoL. The aim of the study was to assess the reliability and the concurrent validity of the CPOS-2 in French and Dutch.

Methods: We conducted a field-test, during which the CPOS-2, in French and Dutch, was tested for its reliability and concurrent validity. Interviews with children facing life-limiting conditions and their parents were conducted by members of the six paediatric liaison teams during their usual home or hospital visits. A sociodemographic questionnaire, the CPOS-2, the Fragebogen für Kinder und Jugendliche zur Erfassung der gesundheitsbezogenen Lebensqualität (KINDL) and finally the Quality of life in life threatening illness-family caregiver (QOLLTI-F) was filled-in for each family.

Results: Seventy-three families participated to the interviews. After item analysis, two items were deleted, resulting in a 20-item CPOS-2, which showed a satisfactory internal consistency (0.76). A standard error of measurement (SEM) of 7.74 % was found, however, which means that an individual total CPOS-2 score should be interpreted with caution. Furthermore, a good convergent validity of the CPOS-2 with the KINDL, and a divergent validity with the QOLLTI-F scores was found, demonstrating that the same construct - quality of life - is assessed by the CPOS-2.

Conclusion: Adding new items to the questionnaire could increase the reliability of the CPOS-2. Using factor-analysis and/or item response models on a larger data set will also

enlighten the dimensionality of the scale as well as the contribution of items to the measurement of the construct that is targeted by the CPOS-2, namely the quality of life.

Introduction

Promoting Quality of life (QoL) is the main goal of paediatric palliative care but assessing QoL remains an important challenge in this particular setting. Two systematic reviews have shown that good instruments to measure outcomes of paediatric palliative care are lacking. (1,2)

A measurement instrument must be reliable. Reliability is defined as "the extent to which scores for patients who have not changed are the same for repeated measurement under several conditions: e.g. when using different sets of items from the same multi-item measurement instrument (internal consistency); over time (test-retest); by different persons on the same occasions (inter-rater); or by the same persons on different occasions (intra-rater) "(3). Briefly, a reliable instrument, is free from measurement error, which means that it will vary as little as possible due to external circumstances or persons. Other terms have been used synonyms of reliability and include precision, consistency, reproducibility or stability.

The degree of reliability of an instrument can be found through calculation of a reliability index, whereas the measurement error can be reported through the calculation of the standard error measurement (SEM).

Reliability and measurement error are separate concepts. Reliability indices assess how well patients can be distinguished from each other[6], while parameters of measurement error assess the magnitude of the measurement error. Reliability is mostly reported as a unitless

---

[6] In classical test theory, reliability is defined as the proportion of the observed variance (σ_o^2) in the measurements which is due to 'true' differences between patients (σ_t^2). The observed variance is defined as the sum of the true variance and the variance caused by all sources of error.

Reliability= $\frac{\sigma_t^2}{\sigma_o^2} = \frac{\sigma_t^2}{\sigma_t^2 + \sigma_e^2}$

index varying between 0 and 1 (maximum reliability) while measurement error is expressed in the same units as the scale, which makes interpretation more straightforward. Moreover, reliability is a characteristic of an instrument used in a particular population, and not just of an instrument. (4)

Classical test theory (CT) partitions observed-score variance into two parts: that which is thought to be linked to the differences between subjects (also called the true variance) and that which is thought to be linked to randomness (error variance). In CT, error variance is considered a monolithic bloc and no subtle distinction is made between different sources of error measurement. Generalizability theory (GT), in contrast (5,6) recognizes that there are multiple definitions of true and error-scores and offers a flexible framework that recognises multiple causes of measurement error and estimates the magnitude of each source separately, thereby offering insights in how to optimize reliability. GT also distinguishes between *relative decisions* that focus on the dependability of the differences among individuals, and *absolute decisions*, where scores are themselves interpretable without reference to others.

Another essential requirement of an outcome measurement instrument is the criterion of validity, which is defined as “the degree to which an instrument truly measures the construct it purports to measure”. (7) Validation focuses on the scores produced by an instrument, not on the instrument itself. It is a continuous process during which the definition of the construct and its underlying theories will become stronger in parallel to the validation of the instrument. Validity is also context-dependent. Therefore, the degree of validity refers only to a certain environment and population in which the instrument has been tested and is dependent on the reliability.

Three types of validity can be distinguished. One of these is content validity, defined as the degree to which the content of a measurement is an adequate reflection of the construct to be measured[7]. The second one refers to the criterion validity, which is the degree to which the scores of a measurement instrument are an adequate reflection of a gold standard. Concurrent validity is part of criterion validity. The third dimension of validity is construct validity, focusing on the degree to which the scores of a measurement instrument are consistent with hypotheses[8].

---

[7] Content validity differs from others forms of validity testing in one important aspect: it is not based on the scores from a scale, performance differences between people, or changes based on some intervention; it is based only on the judgment of experts regarding the content of the items (9). Content validity should also be clearly distinguished from *face validity* that is defined as the extent to which a test is *subjectively* viewed as covering the concept it aims to measure. It refers to the transparency or relevance of a test as it appears to test participants.

[8] It is important to keep in mind that reliability places an upper limit on validity so that the higher the reliability, the higher the maximum possible validity. Or more formally: Validity is

As some constructs are unobservable, such as quality of life, it is challenging to judge whether an instrument truly measures the right construct. Tests of validation require the formulation of specific hypotheses and scientific knowledge, and growing understanding about the construct should drive the hypotheses. These hypotheses concern relationships of the construct under study with other constructs. If these hypotheses are confirmed, then the instrument is apparently suitable to measure the construct. Validation is a continuous iterative process, in particular when the instrument focusses on a complex multidimensional construct. (8)

Another criterion of outcome measurement instruments, linked closely to validity, is responsiveness, defined as "the ability of an instrument to detect change over time in the construct to be measured." (3) For the CPOS-2, which focusses on the construct of quality of life, it means the ability of the CPOS-2 to detect a change in quality of life over time, based on the scores obtained by this instrument on different occasions. Compared to validity, which refers to a single score, responsiveness refers to the validity of a change score (based on two measurements).

Linked to reliability, the standard error of measurement indicates the size of a measurement error. The concept of the *smallest detectable change (SDC)* is closely related to the measurement error of a measurement instrument. SDC can be defined as change beyond measurement error. This implies that if the measurement error is small, relatively small changes can already be identified as real changes and vice versa: if the measurement error is large, changes must be substantial before one can be sure that they are not due to measurement error. To determine the SDC, the SEM should be based on a test-retest reliability index and not a coefficient of internal consistency. (3)

The minimum important change (MIC) is defined as "the smallest change in score in the construct to be measured which patients perceive as important." (3) related to the CPOS-2, the assessment of the MIC is useful to understand whether changes of QoL scores are perceived as clinically relevant from the children's/parents' perspectives, in addition to the assessment of statistically significant changes.

Researchers from the African Palliative Care Association (APCA) and King's College London have designed the 12-item African Palliative Care Association Children's Palliative outcome scale (APCA C-POS). (10) The APCA C-POS is a patient-centred outcome measure (PCOM), which encompasses both patient-reported and proxy-reported measures. PCOMs aim to improve awareness of unmet needs, better symptom recognition, more discussion of quality of life and increased referrals. (11)

---

highly dependent on reliability as expressed in the formula: $Validity_{max} = \sqrt{(Reliability_{New\ test}\ x\ Reliability_{Criterion})}$

The pilot-study presented in the previous chapter (chapter 5) documented the face/content validity, the feasibility and the acceptability of the further developed children's palliative outcomes scale (CPOS-2), which was conducted among 14 children, 19 parents and 9 members of paediatric liaison care teams in Brussels, Belgium. (12) Based on those results, we conducted national multicentre field-testing of the CPOS-2, which is part of a continued development process aimed at exploring its psychometric characteristics and optimizing its design.

In this chapter we will document the reliability and the concurrent validity of the children's palliative outcome scale version 2 (CPOS-2) based on the results of the first field-study.

Methods

The global design and detailed methods of our field-test are described in chapter 2 (Methods).

This multicentric cross-sectional study was conducted in Belgium from 1st February 2019 to 1st March 2020. Six paediatric liaison teams (PLTs) (2 in Flanders, 2 in Brussels and 2 in Wallonia) participated to this study. All study materials were available in 2 languages, French and Dutch.

The study received the approval of the principal ethical committee, the comité d'éthique hospital-facultaire des Cliniques St Luc and was registered as a national clinical trial under the number B403201837760.

Recruitment of children/adolescents and their parents followed-up by PLTs was done by the teams themselves. Families were invited to take part in an interview conducted by members of a PLT. Purposive sampling of all children and adolescents cared for by one of the 6 PLTs in Belgium was carried out, with the following inclusion criteria: > one year old, not at an imminent end-of-life stage, parents able to understand French or Dutch, parents' consent to participation. Children were excluded if they were in their last days of life, if parents were not able to understand French or Dutch or if they were < 1 year old. For each family, quantitative data were collected through several questionnaires, which were printed on carbon sheets in order to keep the original within the PLT and send the copy to the research team at IRSS. Each family was attributed a code, in order to respect confidentiality.

For each of the families who participated in the study, a member of the paediatric liaison care team completed:

- an original 35-item sociodemographic and medical questionnaire including five questions from the Paediatric Palliative Screening Scale (13,14) evaluating the degree of paediatric palliative care needs;
- the self- and proxy-report 22-item CPOS-2 (10,12), evaluating children's and parental quality of life;
- the generic 35-item KINDL questionnaire (15), customized for three age categories, evaluating children's quality of life through child-self and parental-proxy report;
- and the parental self-report 16-item QOLLTI-F (16) assessing parental quality of life.

Those instruments are summarized in Table 14 and can be found in the Appendix.

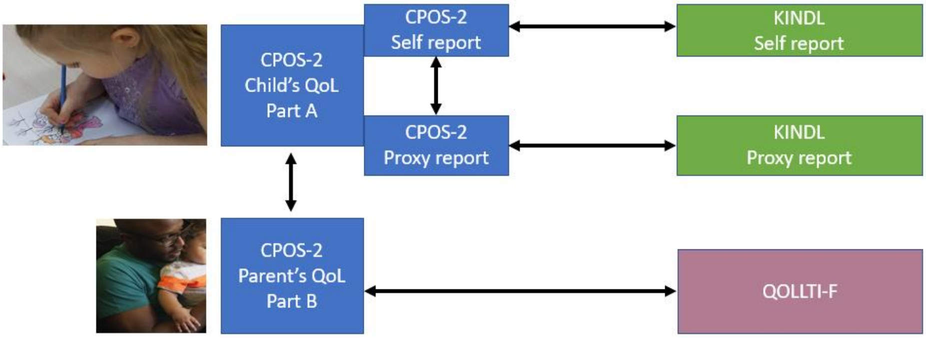
Table 14. presentations of the instruments used in the field-study

	Number of items	**Objectives**	**Designers**	**Validity**
Socio-administrative-medical data	35 items	To document background variables, level of disease, level of suffering (PaPas Scale)	Research team UCLouvain and items from PaPas scale (Bergstraesser et al. 2013,2014)	/
CPOS-2 Childrens Palliative Outcome Scale	22 items 5 dimensions 6 points Likert scale Self and proxy report	To evaluate **child's and parents' QoL** in a paediatric palliative context High score= high impact of the disease on QoL	Downing et al. 2018 (10) Friedel et al. 2020 (12)	Psychometric properties of APCA C-POS not published
KINDL Fragebogen für KINDer und Jugendliche zur Erfassung der gesundheitsbezogenen Lebensqualität	35 items 7 dimensions 3 age-adapted questionnaires 3 or 5 points Likert scale Self- and proxy-report	To evaluate **child's QoL** with and without a disease High score = High impact of the disease on QoL	Erhart et al. 2009 (15)	High concurrent validity with Kidscreen and discriminant validity with healthy children (Bullinger et al. 2008) (17)
QOLLTI-F Quality of life in life threatening illness-family caregiver	17 items 6 dimensions 10 points Likert scale	To evaluate parents' **QoL** when caring for a sick family member High score= High level of QoL	Cohen 2006 (16)	Cronbach α 0,857 Was tested among children with paediatric palliative care needs (Bradford

				et al. 2012, Groh et al. 2013) (18,19)

Figure 7 shows which instruments were compared to the CPOS-2 in the correlation study. We wanted to know whether the further developed CPOS-2 assessed the same construct (quality of life) as the already validated KINDL and QOLLTI-F questionnaire.

Figure 7. Comparison of outcome measurement instruments to assess concurrent validity



To document the reliability and the concurrent validity of the CPOS-2, we followed the steps recommended by the COSMIN guidance on developing measurement instruments in Medicine (3):

1. Explore if any items are left blank more than occasionally, and consider eliminating them
2. Explore the response pattern on each item
3. Explore the distribution of the sum scores.
4. Compute reliability indices with classical test theory and generalizability theory
5. (Conduct item analysis according to item response theory)
6. (Explore the dimensionality of the dataset with factor analysis)

Preliminary remarks:

The construct (Quality of life) to be measured was defined and elaborated previously in close partnership with stakeholders (children, parents and paediatric liaison teams).

All items were checked for understandability and semantic clarity (unambiguity).

Each item of the CPOS-2 is scored on a six-point Likert scale ranging from 0 to 5. The items are worded in such a way that a higher item score implies a poorer quality of life or, more precisely, a higher burden. The values provided by 12 out of the 22 items have to be reversed to explore the construct in the same direction.

The total score produced ranges between 0 and 60 for part A and between 0 and 50 for part B. These scores are transformed to percentage scores that are easier to interpret.

The scale contains reflective items and formative items and should therefore be considered as having a hybrid structure.

Statistical analysis

A rigorous procedure of “data-cleaning” was used. Data input was achieved by means of specifically developed data capture screens with a layout similar to the paper-based records.

Statistical calculations were performed using SPSS 25 (SPPP Inc, Chicago Il, USA) and Medcalc 11.5 (Medcalc Software Oostende) and p-values <0.05 were considered significant.

The characteristics of the distributions of the scores on different scales were explored by means of histograms and PP-plots. Descriptive statistics are presented as the mean ±standard deviation (SD) or median with inter-quartile range (IQR). To explore concurrent validity, Pearson correlation was used. Next to the observed correlation coefficients, the coefficients after correction for attenuation are also presented. [9]

Missing Value Analysis and Analyse Patterns in SPSS were used to explore patterns of mission values. Multivariate normal regression for multiple continuous variables imputation with creation of five imputations was performed, followed by an averaging procedure to prepare a dataset without missing values needed for the generalizability analysis.

Generalizability theory (G theory) is a statistical method for the analysis of psychometric test and rating scales results. It is a generalization of classical reliability theory, which examines the relative contribution of the primary variable of interest and the performance of subjects and compares these to error variance. (5,20,21) Traditional reliability

---

[9] Attenuation is a statistical concept that refers to underestimating the correlation between two different measures because of measurement error (22).

methodology conceptualizes an observed score as being determined only by these two potential sources of variance: variance attributable to the ‘‘true’’ score and variance attributable to only one other source of error encompassing all other sources of error. In G theory, various sources of error contributing to the inaccuracy of measurement are explored. G theory is a valuable tool in judging the methodological quality of an assessment method, allowing the identification of the relative contribution of different sources of measurement error to improve the precision of the scores produced by an instrument and to optimize its design.

A generalizability study was run by means of the GENOVA program. (6) First, a G-study was organised based on a person x items design (p x i) to calculate the weight of different variance components. Subsequently a decision study (D-study) was organized to explore the impact of the number of items on two kinds of reliability indices: the G-coefficient that reflects the reliability of the rank ordering of the subjects and the Ph-coefficient that evaluates the reliability needed for an absolute interpretation of the total scores.

Results

Seventy-three families accepted to participate in the field-study. Figure 8 shows the flow-chart of the number of the included families and useable records after data cleaning.

Figure 8. Flow chart of the number of included families and useable records after data cleaning

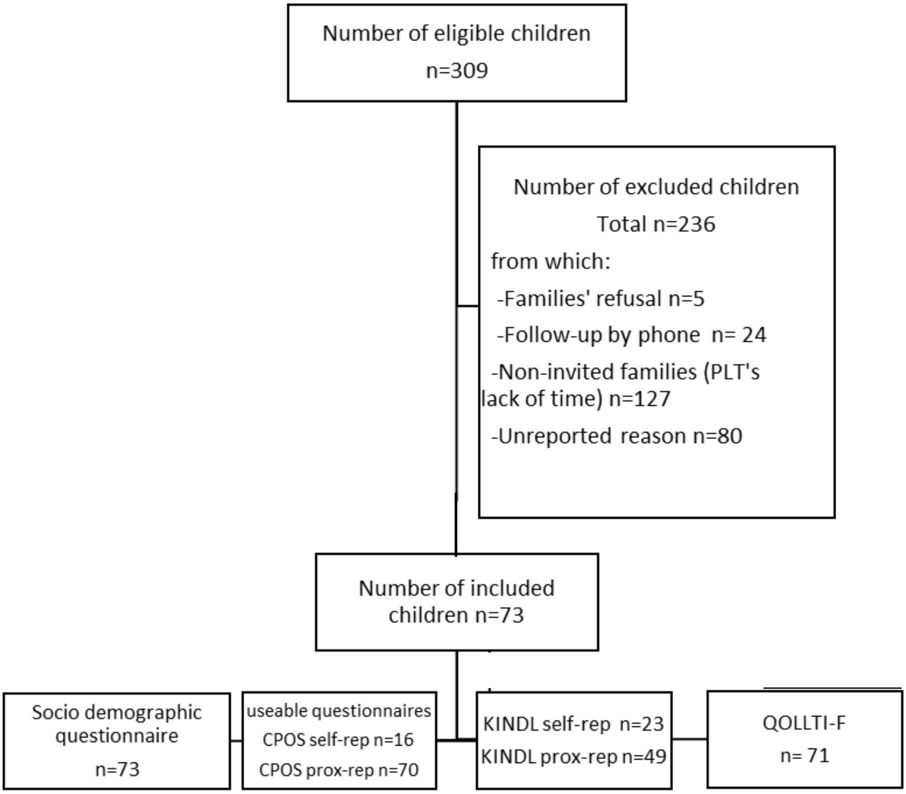


Table 15. Comparison of the sample of included children in the field-study with the group of children not included in the study

Characteristics		**Sample of children included in the field-study n (%)**	**Population of non-included children (but with same inclusion criteria) n (%)**
Gender	Total	73 (100%)	236 (100%)
Female		37 (50%)	82 (35%)
Male		36 (50%)	124 (53%)
Unknown		0	30 (13%)
Age	Total	73 (100%)	236 (100%)
1-2 years		19 (25%)	53 (22%)
3- 6 years		25 (35%)	70 (30%)
7-13 years		22 (30%)	79 (33%)
14-18 years		7 (10%)	34 (14%)
Disease	Total	73 (100%)	236 (100%)
Neurology		28 (38%)	36 (15%)
Oncology		18 (25%)	87 (37%)
Metabolic/genetic		17 (23%)	70 (30%)
Neonatology		7 (10%)	6 (3%)
Cardiology		3 (4%)	2 (1%)
Other		/	35 (14%)

As shown in Table 15, we found that our sample was representative for age ranges and gender but not for categories of diseases. In fact, onco-haematological diseases were underrepresented in our sample, compared to the population of children followed-up by paediatric liaison teams.

Item analysis

Our three-step item-analysis procedure consisted of

1. A critical review of the number of items left blank

2. A visual inspection of the response patterns and 3. The exploration of the item-total score correlations.

We looked at the items left blank in the CPOS-2 (table 16) and found that two questions presented a high amount of blank responses. These questions were: Question 7: "*Do you feel that your child still has questions about his or her illness? "(*42%) and Question 11: *"Do you feel that your child would like to change something in your family?"* (35%)

An explanation for these two items being left blank is the fact that a high number of children included in this study are non-verbal. This means that it is very difficult for parents to estimate what their children think or feel. Consequently, we decided to delete those 2 questions.

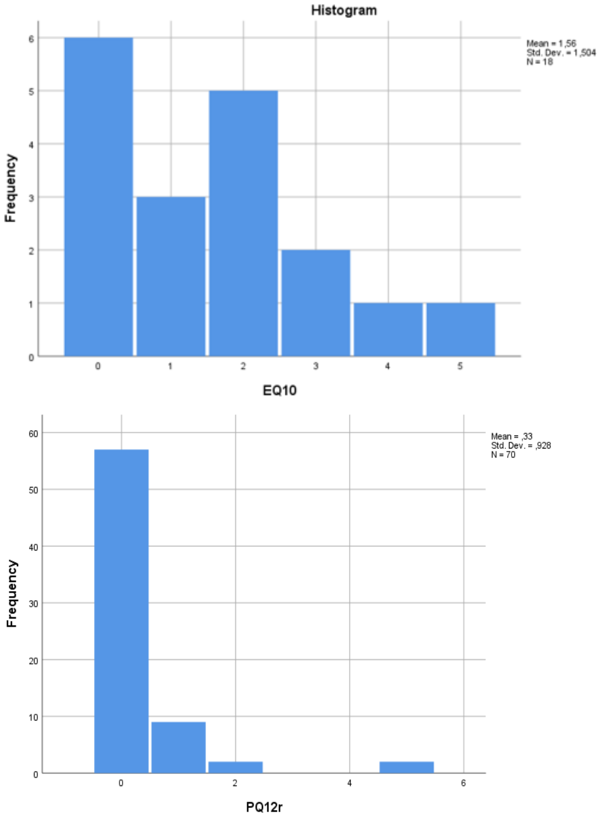
Table 16. Overview of the number of items that were left blank

Item	n/total	%
EQ1	1/23	4,35
EQ2	2/23	8,70
EQ3	1/23	4,35
EQ4	1/23	4,35
EQ5r	1/23	4,35
EQ6r	1/23	4,35
EQ7	4/23	17,39
EQ8r	0	0,00
EQ9r	4/23	17,39
EQ10r	3/23	13,04
EQ11	2/23	8,70
EQ12r	0/23	0,00
PQ1	1/70	1,43
PQ2	1/70	1,43
PQ3	2/70	2,86
PQ4	3/70	4,29
PQ5r	3/70	4,29
PQ6r	5/70	7,14
PQ7	*30/70*	*42,86*
PQ8r	0	0,00
PQ9r	3/70	4,29
PQ10r	11/70	15,71
PQ11	*25/70*	*35,71*
PQ12r	0	0,00
Q13	0	0,00
Q14r	1/73	1,37
Q15r	0	0,00
Q16r	3/73	4,11
Q17r	0	0,00
Q18r	2/73	2,74
Q19	0	0,00
Q20	0	0,00
Q21	0	0,00
Q22r	1/73	1,37

As a next step, we visually explored the response pattern for each item in order to understand whether some items were not discriminating enough, i.e. whether the response patterns were extremely skewed. Figure 9 shows an example of desired response pattern (item 10 in Part A (self-report) of the CPOS (EQ2) and of a rather strange pattern (item 12 (PQ12r) in Part A (proxy-report).

Figure 9. Example of a desired and a problematic response pattern on 2 items of part A of the CPOS-2

EQ10



We found that all items of the CPOS-2 showed an acceptable response pattern, except for Question 12: “Do you think that your child feels loved? » on which 82% of the parents and 80% of the children gave a maximum score of 5. However, from a perspective of content validity there was a consensus that a high score on this item (to interpret in a reversed way as a low score on the question) could be interpreted as an important signal. Consequently, we decided not to cut out the item.

Descriptive statistics

Next, we explored the distribution of the sum scores of the CPOS 2 and the other scales using histograms and PP plots. For all questionnaires (CPOS-2, KINDL and QOOLTI), we found a normal distribution, whether it be the self-report or the proxy-report parts. Moreover, no apparent floor or ceiling effects were discovered.[10] See Figures 10-14.

---

[10] Floor or ceiling effects can occur when a high proportion of the total population has a score at the lower or upper end of the scale respectively.

Figure 10. Distribution of the sum scores Part A self-report, Part A proxy-report and Part B of the CPOS-2 (histograms)

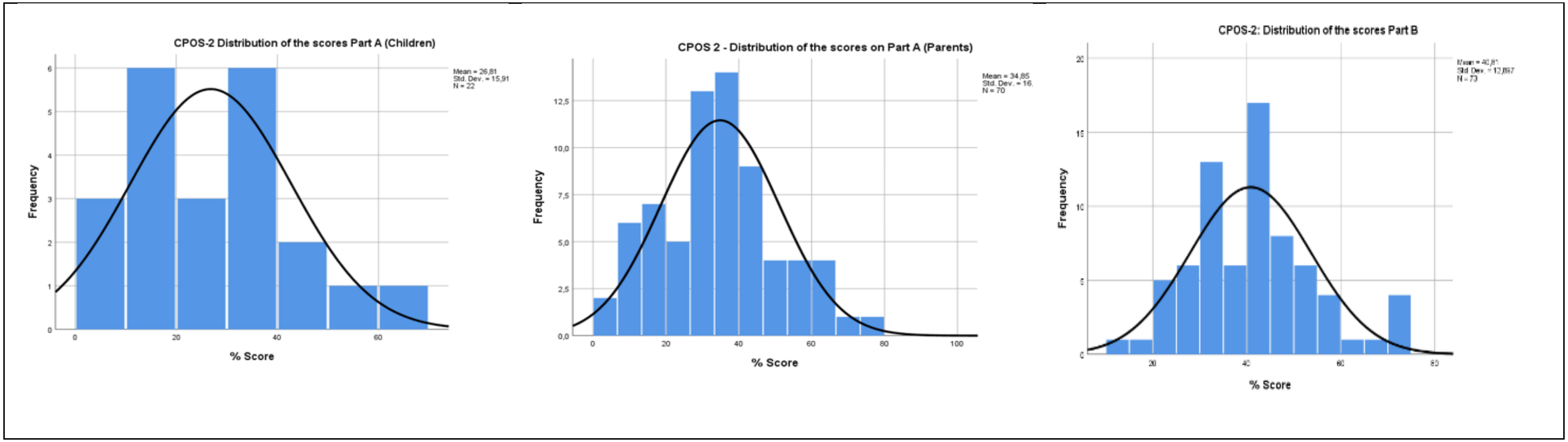


Figure 11. Distribution of the sum scores Part A self-report, Part A proxy-report and Part B of the CPOS-2 (P-Plots)

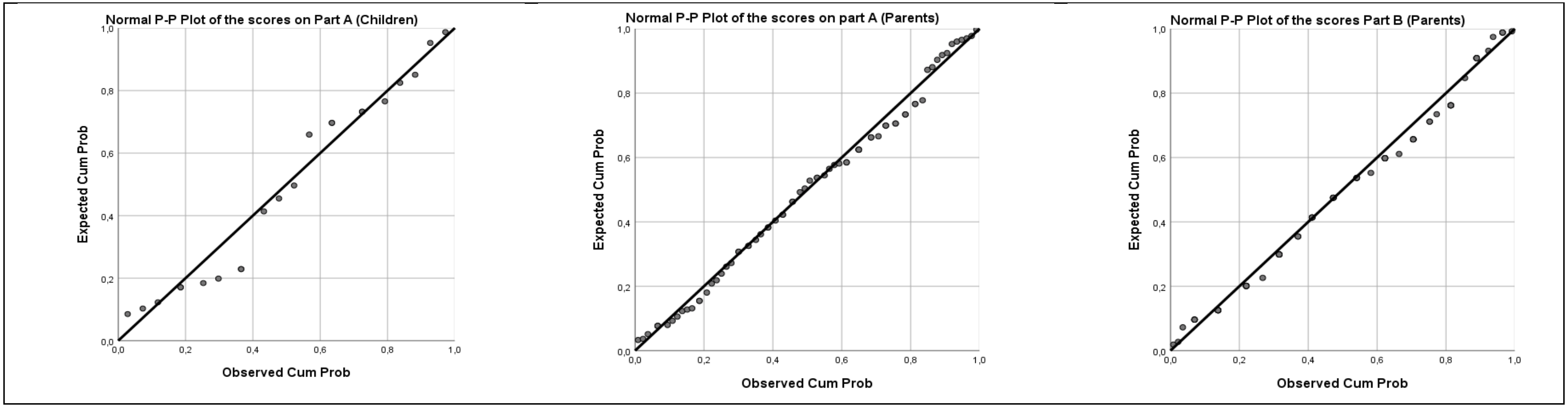


Figure 12. Distribution of the sum scores for the self-report and proxy-report of the KINDL (histograms)

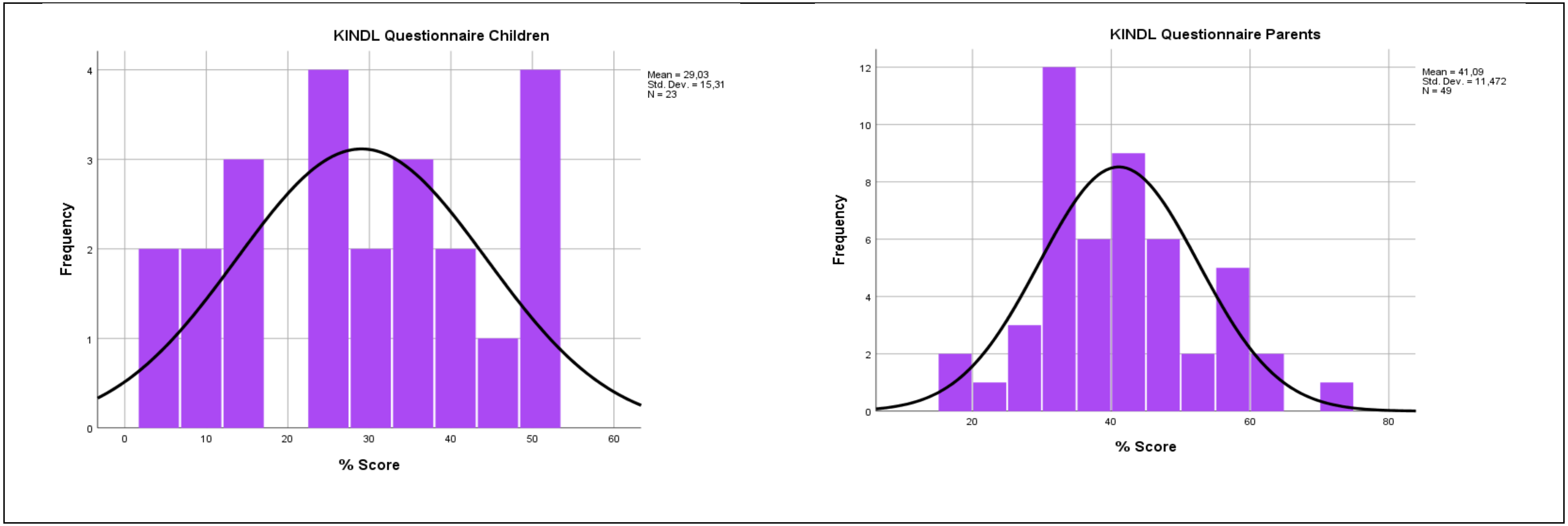


Figure 13. Distribution of the sum scores for the self-report and proxy-report of the KINDL (P-plots)

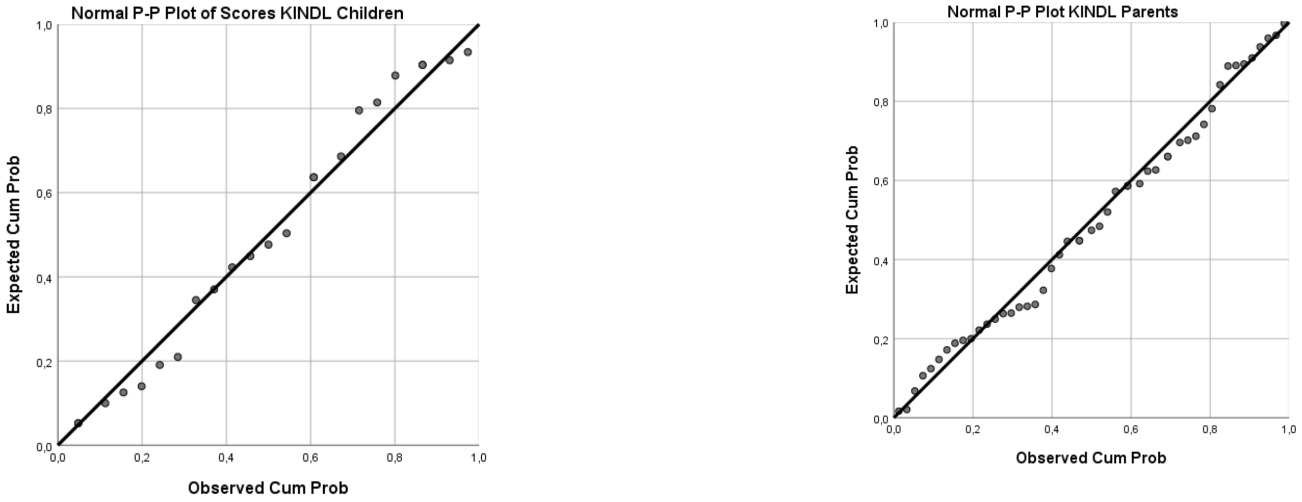
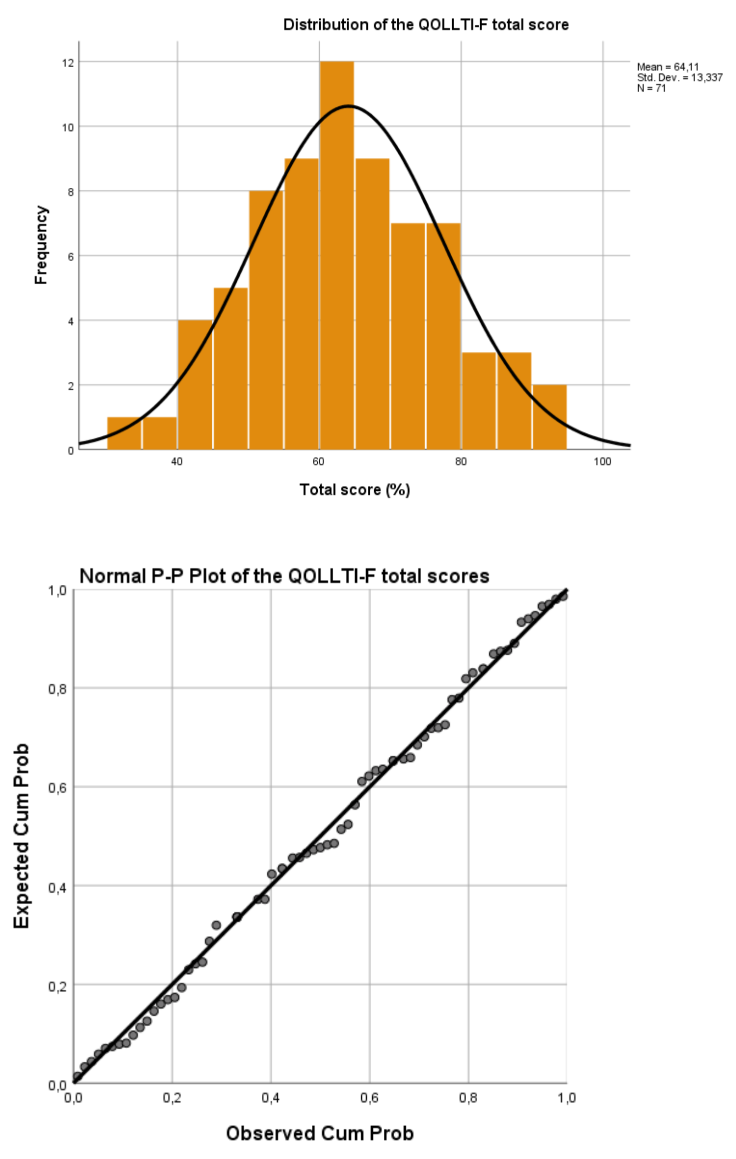


Figure 14. Distribution of the sum scores of the QOLLTI-F



Reliability of the CPOS-2

We computed reliability indices for the CPOS with classical test theory and generalizability theory. When combining both parts (A and B) of the CPOS-2, the initial alpha Cronbach for part A (self-report) was 0.52, for part A (proxy-report) 0.54, for Part B 0.59 and for the combined part A and Part B scale 0.71.

As shown in table 17, we noticed that questions 7 and 11 of the CPOS-2 correlated negatively with the total score and deleting these items appeared to beneficially impact the index of internal consistency. This provided us with an additional argument (in addition to the high number of blanks) for deleting these two items in our further analysis.

Table 17. Item-total correlation and impact on the Cronbach's Alpha when the item is deleted

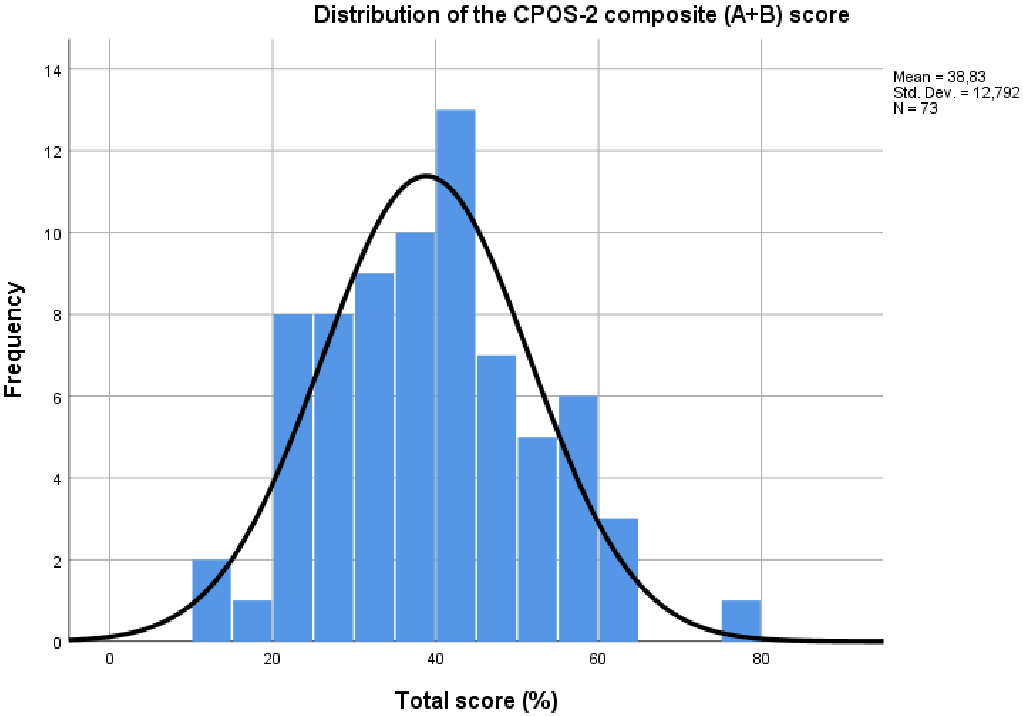
Item-Total Statistics

	Scale Mean if Item Deleted	Scale Variance if Item Deleted	Corrected Item-Total Correlation	Cronbach's Alpha if Item Deleted
PQ1	38,65	158,715	,381	,691
PQ2	37,46	146,578	,600	,665
PQ3	38,38	161,286	,295	,700
PQ4	38,65	154075	,465	,682
PQ5r	39,50	164,420	,604	,686
PQ6r	38,92	164,394	,323	,697
PQ7	*38,88*	*191,626*	*-,271*	*,755*
PQ8r	38,62	167,206	,341	,697
PQ9r	39,62	166,406	,366	,696
PQ10r	38,15	163,895	,249	,704
PQ11	*39,31*	*180,222*	*-,070*	*,736*
PQ12r	40,46	178,018	,199	,710
Q13	36,73	161,325	,380	,692
Q14	38,88	163,466	,269	,702
Q15	40,19	182,242	-,104	,722
Q16	39,23	171,785	,288	,702
Q17	40,50	177,620	,387	,708
Q18	39,65	162,795	,436	,689
Q19	36,85	163,095	,454	,689
Q20	37,00	175,680	,030	,724
Q21	38,19	161,282	,278	,702
Q22	38,27	151,645	,646	,668

After the item analysis, the adapted CPOS-2 scale consists of 20 residual items for which a Cronbach’s alfa coefficient of 0.76 was found. (part A self-report: 0.68, part A proxy-report: 0.71, part B: 0.59)

Figure 15 shows the distribution of the final combined CPOS-2 score that targets the quality of life in the family.

Figure 15. Distrubution of the composite (A and B) score of the final version of the CPOS-2



Generalizability analysis

A missing value analysis revealed no particular patterns in the missing values. However, the number of missing values was not neglectable (29% of the cases presented one or more missing values in the matrix, and the total number of missing values was 8,9%). We therefore decided to implement a multiple imputation routine as described above. After multiple imputations, full data were available for 73 families. An F x I design was used to perform the G-analysis. F stands for families, I for items and the x signifies a crossed design.

Table 18 shows the results of Step 1 (the G-analysis). The variance components are reported for each of the sources of variance that this design allows estimation of.

Table 18. Estimated variance components of the CPOS-2

Source	Estimated Variance Component	Standard Error	Percentage of total variance
F	127.74	28.05	9.7 %
I	416.96	132.23	31.5 %
fi	783.39	30.57	58.80 %

The f component shows the variance attributable to families. This component is the “wanted” variance or universe score variance that the instrument focuses on. Compared to the other sources, this effect is relatively small (approximately 10 %) although this is rather common in this type of scale. The I component represents the variance between items and the way they discriminate between subjects and represents 31% of the variance. The fi component reflects the inconsistency with which the items rank-order subjects (families) along the CPOS -2 scale in addition to all of the other sources of “error” variance (as for instance the fact that different nurses scored different patients/families).

D-Study

In step 2 (the Decision study), different kinds of reliability indices are reported in relation to the actual and virtual number of items. The G-Coefficient is defined from the so-called norm referenced perspective and reflects the reliability of the ranking of the families. With the actual number of items in the CPOS-2 (n=20) a reliability of 0.76 is found. The Phi-coefficient considers all possible sources of error-variance and reflects the reproducibility of the absolute scores. With the actual number of items, an index of 0.68 is yielded. The computed Standard Error of Measurement (SEM) is more relevant and easier to interpret. It is expressed in the same units as the variable of interest (%) and allows us to compute a 96% confidence interval around an individual score. With the actual number of items, the 95% CI = ± 1,96 x 7.74%= ±15 %. (See Table 19)

Table 19. Reliability indices of the CPOS-2 as a fonction of the number of items in the scale

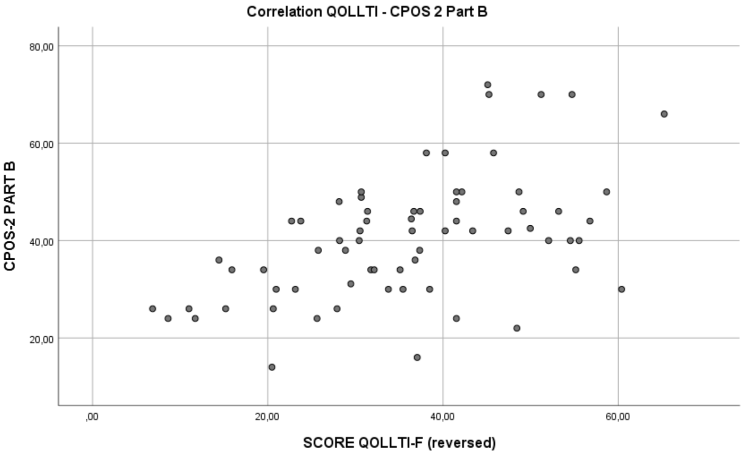
Number of Items	G-coefficient (norm referenced perspective)	Phi-Coefficient (domain referenced perspective)	Standard Error of Measurement (%)
15	0.70	0.61	8.49
20	*0.76*	*0.68*	*7.74*
25	0.80	0.72	6.92
30	0.83	0.76	5.32
35	0.85	0.80	4.89

Exploration of the concurrent validity

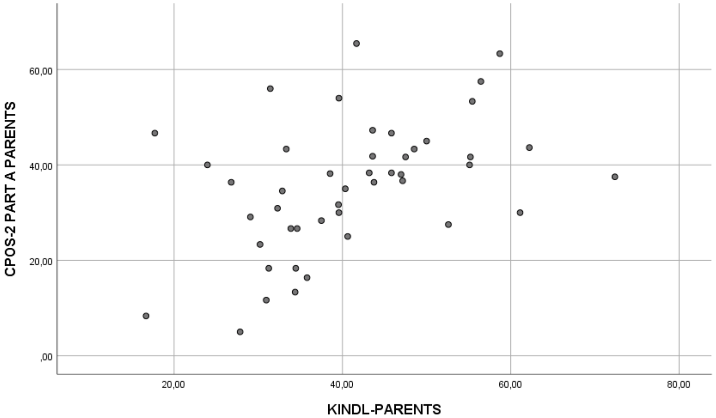
To document the **concurrent validity** of the CPOS-2, we conducted correlation studies, by computing the Pearson correlation. Figure 16 shows some of the scatterplots we used to explore the relationship between scales intended to measure similar constructs.

Figure 16. Correlation between A. The QOLLTI-F and CPOS-2 part B assessing both parent's QoL; B. The KINDL-parents and the CPOS part A parents assessing both children's QoL (proxy-report); C. The KINDL-child and the CPOS-2 part A assessing both child's QoL (self-report)

A


Correlation QOLLTI - CPOS 2 Part B
CPOS-2 PART B
80,00
60,00
40,00
20,00
,00
20,00
40,00
60,00
SCORE QOLLTI-F (reversed)

B


CPOS-2 PART A PARENTS
60,00
40,00
20,00
,00
20,00
40,00
60,00
80,00
KINDL-PARENTS

C

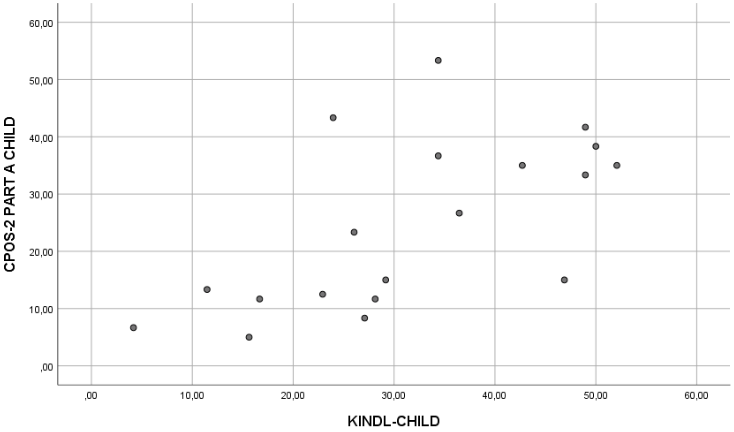

CPOS-2 PART A CHILD
60,00
50,00
40,00
30,00
20,00
10,00
,00
,00
10,00
20,00
30,00
40,00
50,00
60,00
KINDL-CHILD

Table 20 shows the correlations between the scores on the different instruments in a multi-trait-multi method matrix as was suggested by Campbell and Fiske. (23)

The observed as well as the "attenuated" correlation indices are reported. Observed correlation can underestimate the magnitude of the association because of the amount of measurement error. When correlating scores from two instruments, the observed correlation may be substantively lower if the score reliabilities from both instruments are low. To compute corrected correlation coefficients, we used the following formula:

$$r_{x'y'} = \frac{r_{xy}}{\sqrt{r_{xx} r_{yy}}}.$$

In this formula, $r_{x'y'}$ is the correlation between the two instruments corrected for attenuation; r_{xy} is the observed correlation and r_{xx} and r_{yy} are the reliability indices of the first and second instrument. In order to compute corrected correlation indexes, the following average reliability indices were used, based on existing literature: for the QOLLTI-F : 0.78; for the KINDL questionnaire for parents: 0.84 ; and for the KINDL questionnaire for children 0.82.

We found statistically significant correlations between

the CPOS-2, part A (measuring children's QoL) as reported by children and the CPOS-2 as reported by parents;

the CPOS-2 part A (measuring children's QoL) and the KINDL, both as reported by children;

the CPOS-2 part A and the KINDL (measuring children's QoL), both as reported by parents;

and finally, also between the CPOS part B and the QOLLTI-F (measuring parents' QoL), both as reported by parents.

Table 20. Observed and attenuated correlation between the scores on the CPOS-2, the KINDL and the QOLLTI-F

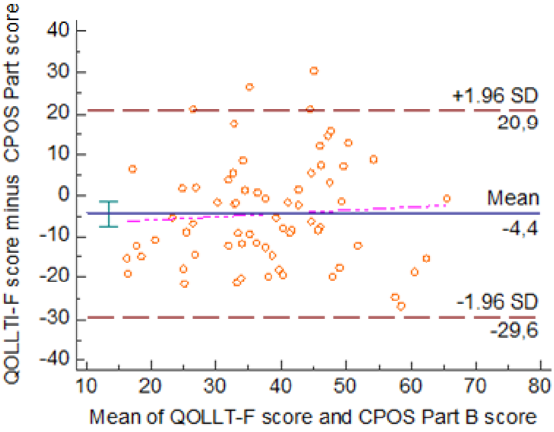
	CPOS-2 Part A Child	CPOS-2 Part A Parents	CPOS-2 Part B	KINDL Child	KINDL Parents	QOLLTI-F (reversed)
CPOS-2 A Child		0.55*	0.42*	0.62**		
CPOS-2 A Parents	*0.79°*		0.47**		0.44**	
CPOS-2 B	*0.65°*	*0.73°*				0.52**
KINDL Child	*0.83°*					
KINDL Parents		*0.57°*				
QOLLTI-F			*0.77°*			

*. Correlation is significant at the 005 level (2-tailed)
**. Correlation is significant at the 0.01 level (2-tailed)
° Coefficients corrected for attenuation

CPOS: Children palliative outcome scale;
KINDL: Kinder Lebensqualität Fragebogen;
QOLLTI-F: Quality of life in life-threatening illness-Family carer

Furthermore, to explore whether there was a linearity over the whole spectrum for the correlation between the CPOS-2 and the QOLLTI-F, a Bland-Altman plot was realised, which showed, indeed, a linearity (Figure 17).

Figure 17. Bland-Altman plot of the concordance between the scores of the QOLLTI-F score and the CPOS scores Part B



Discussion

Our findings document the reliability and the concurrent validity of the CPOS-2.

The generalizability analysis reveals that with the actual number of items (n=20) a G - coefficient of 0.76 is produced. This is a satisfactory result for a hybrid scale including a balanced proportion of reflective and formative items. Reflective items are questions which are considered to influence quality of life (e.g. Do you feel pain?) and formative questions are supposed to reflect quality of life (e.g. Are you happy? Are you sad?).

From a domain referenced perspective (=the reliability of the absolute score), however, a standard error of measurement of 7.74 % is found, which means that an individual total CPOS score (expressed as a percentage score) should be interpreted with caution since a 95% confidence interval of ±1,96 x 7.74% or ±15 % should be imagined around each value.

Regarding concurrent validity, several of our hypotheses were confirmed. We found a significant positive correlation between part B of the CPOS2 and the (reversed) scores of the QOOLTI-F, confirming that both scales measure similar constructs. We also observed a positive correlation between the part A of the CPOS and the KINDL- self report, although the latter was developed as a generic scale to asses children's wellbeing and not as a disease-specific scale. The correlations are significant but not extremely high, which supports the idea that the CPOS-2 might measure as intended a construct that is familiar but not identical to the ones targeted by the KINDL and the QOOLTI-F.

Compared to the KINDL and the QOOLTI-F, the CPOS-2 potentially offers a few new assets. It intends to measure both children's and parents' QoL in a combined scale, focusing explicitly on the well-being of the family as a unit. Additionally, it combines children's self- and parents' proxy-reports and is composed of an acceptable number of items, which reduces the burden of interviews for children, parents and the healthcare professionals who use it.

We would, however, like to suggest some strategies for improvement of the CPOS-2. Extending the questionnaire with additional items is one road to explore. A new in-depth analysis of a larger dataset using factor-analysis and/or Item response models will also enlighten the contribution of items to the measurement of the construct that is targeted by the CPOS. With the current extended MOSAIK study in 8 paediatric palliative care teams in France, we hope to reach an overall sample size of 120 to be able to explore the dimensionality of the dataset through factor analysis. Finally, to further document the reliability of the tool, a test/retest study among two paediatric liaison teams in Belgium is foreseen.

Unfortunately, due to the small number of records in our study (n=73), we couldn't proceed to an analysis from within the framework of Item response theory, neither were we able to explore the dimensionality of the dataset with factor analysis. Indeed, we would need at least a sample size of 100 to compute those analyses. We aimed to reach a sample size of 120, but this number could not be reached in this field-study. We identified the following reasons: lack of time for paediatric liaison teams to include more families, changes in staff composition such as the absence of a medical coordinator or training of a new colleague, a high number of families followed-up via telephone without consultations or home visits needed. Overall, only five families (of 78) refused to participate in the study after having been invited, demonstrating a high collaboration rate.

Conclusion

After item analysis, two items were deleted, resulting in a 20-item CPOS-2, which showed a satisfactory internal consistency (0.76). A standard error of measurement (SEM) of 7.74 % was found, however, which means that an individual total CPOS-2 score should be interpreted with caution. Furthermore, a good convergent validity of the CPOS-2 with the KINDL, and a divergent validity with the QOLLTI-F scores was found, demonstrating that the same construct - QoL - is assessed by the CPOS-2. Adding new items to the questionnaire could increase the reliability of the CPOS-2. Using factor-analysis and/or Item response models on a larger data set will also enlighten the dimensionality of the scale as well as the contribution of items to the measurement of the construct that is targeted by the CPOS-2, namely the QoL.

References

1.Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi:10.1542/peds.2018-2379

2. Coombes LH, Wiseman T, Lucas G, Sangha A, Murtagh FE. Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-949. doi: 10.1177/0269216316649155.

3. De Vet HC, Terwee CB, Mokkink LB, Knol DL. (2011) Measurement in Medicine. A Practical Guide. Cambridge: Cambridge University Press.

4. Streiner D, Norman G, Cairney J. Health measurement scales: a practical guide to their development and use. Oxford University Press. 2015. 399 p.

5. Cronbach LJ, Gleser GC, Nanda H, Rajaratnam N. (1972) The dependability of behavioural measurements: theory of generalizability for scores and profiles. New York: Wiley.

6. Brennan RL. (2002) Generalizability theory. New York: Springer.

7. Mokkink LB, Terwee CB, Patrick DL, et al. International consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes: results of the COSMIN study. J Clin Epidemiol. 2010; 63 :737-745.

8. Strauss ME & Smith GT. Construct Validity: Advances in Theory and Methododoly. Annual Reviews in Clinical Psychology. 2009; 5; 1: 1-25.

9. Messick, S. 1989. Validity In Linn RL (Ed.), Educational measurement (3rd ed., pp. 13-104). New York, NY: American Council on education and Macmillan.

10. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02.

11. Etkind SN, Daveson BA, Kwok W, Witt J, Bausewein C, Higginson IJ, Murtagh FE. Capture, transfer, and feedback of patient-centered outcomes data in palliative care populations: does it make a difference? A systematic review. J Pain Symptom Manage. 2015;49(3):611-24. doi: 10.1016/j.jpainsymman.2014.07.010.

12. Friedel M, Brichard B, Boonen S, Tonon C, de Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse J, Aujoulat I. Face and content validity, acceptability and feasibility of the adapted version of the Children's Palliative Outcome Scale (CPOS-2): a pilot study. [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646

13. Bergstraesser E, Hain RD, Pereira J. The development of an instrument that can identify children with palliative care needs: the Paediatric Palliative Screening Scale (PaPaS Scale): a qualitative study approach. BMC Palliative Care 2013; 12:20.

14. Bergstraesser E, Paul M, Rufibach K, Hain RD, Held L. The Paediatric Palliative Screening Scale: Further validity testing. Palliat Med. 2014;28(6):530-533.

15. Erhart M, Ellert U, Kurth B, Ravens-Sieberer U, Measuring adolescents' HRQoL via self and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument. Health and Quality of Life Outcomes. 2009; 7:77.

16. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006 Dec;20(8):755-67. doi: 10.1177/0269216306072764.

17. Bullinger M, Brütt AL, Erhart M, Ravens-Sieberer U; BELLA Study Group. Psychometric properties of the KINDL-R questionnaire: results of the BELLA study. Eur Child Adolesc Psychiatry. 2008;17 Suppl 1:125-132. doi:10.1007/s00787-008-1014-z.

18. Bradford N, Young J, Armfield NR et al: A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. J Telemed Telecare 2012;18(8): 438-442.

19. Groh G, Borasio GD, Nickolay C et al: Specialized paediatric palliative home care: a prospective evaluation. J Palliat Med. 2013;16,12: 1588-1594.

20. Shavelson R, Webb N, Rowley G. Generalizability Theory. American Psychologist. 1989;44:6: 922-932.

21. Bloch R, Norman G. Generalizability theory for the perplexed: A practical introduction and guide: AMEE Guide No. 68. Medical Teacher. 2012;34:11,960-992.

22. Gulliksen H. Theory of mental tests. Lawrence Erlbaum, 1987.

23. Campbell DT & Fiske DW. Convergent and discriminant validation by the multitrait-multimethod matrix. Psychological Bulletin. 1959; 56, 81-105.

6.2. CAN WE MEASURE SPICE OF LIFE? ASSESSING QUALITY OF LIFE AMONG CHILDREN FACING LIFE-LIMITING CONDITIONS AND THEIR PARENTS

ABSTRACT

Background and objectives: Outcome measurement research has been considered as a research priority in the field of paediatric palliative care, in order to improve quality of care offered to children and to foster evidence of the effectiveness of paediatric palliative care. This study aims to measure the QoL of children with life-limiting conditions, who are followed-up by Belgian paediatric liaison teams and the QoL of their parents, through the further developed children's palliative outcome scale (CPOS-2).

Methods: During a field-study in 2019, interviews were conducted with children facing life-limiting conditions, who were followed-up by paediatric liaison teams, and their parents. A socio-demographic questionnaire, the CPOS-2, the KINDL and the QOLLTI-F were filled in by paediatric liaison team members during their home or hospital visits. Descriptive statistics were conducted on the sociodemographic data, the CPOS -2 and QOLLTI-F scores.The association between clinicians' judgement of quality of life and the scores on the QOLLTI-F and CPOS-2 was explored using Spearman's correlation coefficients and boxplots. Pearson chi square test and the Fisher exact test were used to investigate significant differences between scores. Analysis was performed with SPPSS 26.O (SPSS inc., Chicago, IL, USA). Results were discussed and interpreted with six paediatric liaison teams.

Results: A total of 73 children/adolescents (1-18 years) were included in the study. A slight discrepancy between children's and parents' scores on children's QoL, as measured by the CPOS-2 and the KINDL, was found. Especially for items focusing on emotional items, children included in our study self-reported their QoL as higher than their parents did. The QoL scores are not significantly associated with the condition's severity or impaired activities of daily life. Discrepancies were not found between health care professionals' perceptions and parents' perceptions on their QoL.

Conclusion: This study provides, for the first time, an overview of the quality of life of families followed-up by a paediatric liaison team in Belgium in 2019. Quality of life scores do not seem to be associated with the severity of the disease.

Introduction

The main goal of paediatric palliative care is to improve quality of life for both the child and the family. (1,2)

Quality of life, however, is a complex construct, defined by the WHO as “an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns.“ (3)

Knowing the fundamentally subjective essence of this concept, is it possible to evaluate quality of life? Instruments to measure quality of life in a paediatric palliative care population are lacking due to many methodological and clinical challenges. (4-6)

Outcome measurement research has been considered as a research priority in the field of paediatric palliative care, in order to improve quality of care offered to children and to foster evidence of the effectiveness of paediatric palliative care. (7-9) In recent years, patient-centred outcome measures (PCOM), which encompass both patient-reported and proxy-reported measures, have largely been developed. An outcome measurement instrument evaluates ‘change in health status’ as a consequence of health care or interventions. (10) The term health status should be understood in a broad perspective of well-being or quality of life. If we refer to Donabedian’s model for evaluating the quality of care, we must look at the structure, the process and the outcomes of a care program. High quality paediatric palliative care is presumed to improve quality of life. Quality of life, however, is a multidimensional construct and is, most probably, not only influenced by the burden of disease or the quality of care. “It is important to recognise that experience of care is not the same as outcomes of care. Experiences are likely to be better if outcomes are better, but they relate more closely to how individuals are respected, listened to and heard.” (11)

It is not clear, yet, whether (paediatric palliative care) PPC outcomes overlap with quality of life or not. We assume that assessing quality of life in children facing life-limiting conditions is a way to assess meaningful outcomes of PPC, which are not limited to the burden of disease. We are, nonetheless, aware that some factors affecting children’s or parents’ quality of life may not be managed or controlled by PPC healthcare teams. Furthermore, we acknowledge that an outcome measurement instrument will probably never be able to assess quality of life in a perfectly reliable way.

To better understand the children’s perspectives on the palliative care they receive, however, and therefore to identify the meaningful outcomes of paediatric palliative care, a meta-summary suggested eight themes: the relationship with professional caregivers, pain and its management, “living beyond pain”, the relationship between paediatric patients and their families, children’s views on their treatment and service provision, meanings children give to their end-of-life situation, consequences of clinical decisions, and the relationships among children in paediatric palliative care and their peers. (12)

Those findings were confirmed by a systematic review conducted by Namisango et al., which looked at the meaningful domains of paediatric palliative care (13). She found that five domains reflected priority concerns: physical (e.g. symptoms), psychological (e.g. worries), psychosocial (e.g. relationships), existential (e.g. existential loss) and others (e.g. information access). She noticed that children’s perspectives were not systematically researched.

Despite the methodological challenges to developing an outcome measurement instrument, a promising tool developed by the African Palliative Care Association (APCA), called the APCA children’s palliative outcome scale (APCA C-POS), was developed in an African context of care (14,7). Based on the APCA palliative outcome scale for adults (15), it combines a self- and proxy-report. The C-POS is a multidimensional patient-centred outcome measure, with 5-point Likert-scale response options. It contains 12 items exploring physical and psychosocial elements. Seven items are related to children’s quality of life, which can be rated by the children themselves (self-report) and rated by their parents (proxy-report) and five items focus on parents’ quality of life.

In Belgium, paediatric palliative care is provided through paediatric liaison teams (PLTs) attached to university hospitals. They offer curative, palliative and liaison care across all care settings as a mobile team, available 24/7. Services provided in a family-centred approach to ensure continuity of care are free of charge for users. Previously published studied have shown that more than 700 children aged 0-18 years are followed-up on an annual basis by those teams in Belgium. (16) Their probable insufficient access for the Brussels region has, however, been documented. (17)

A pilot-study on the face/content validity, acceptability and feasibility of the children’s palliative outcome scale (7) has been conducted among 14 children, 19 parents and 9 representatives of paediatric liaison teams and has been published previously (18). During this pilot-study, the original APCA CPOS was further developed by adding items to the scale, which were found meaningful for children. In fact, children were invited to self-elicit domains of Quality of Life (QoL) via an instrument called the Scheduled Evaluation of Quality-of Life (SEIQoL-DW) (19), resulting in a CPOS version 2 (CPOS-2), which includes 22 items.

This chapter presents an overview of the sociodemographic characteristics and the quality of life of children facing life-limiting conditions, and their parents that are followed-up by 6 paediatric liaison teams in Belgium in 2019.

We will first present the characteristics of the population, then focus on three research questions:

1. How do the scores on the QOLLTI and the CPOS-2 correlate with the scores observed in other populations?

2. How do the appreciation of the well-being of parents and children by the responsible paediatricians' correlate with quality of life as measured by the QOLLTI- (parents) and the CPOS-2 (children's self- and parents' proxy-report)?

3. Which characteristics and/or background variables are associated with high scores on the QOLLTI and/or the CPOS-2?

Methods

The detailed data collection and data analysis are described in chapter 2 (Methods).

This multicentric cross-sectional mixed-methods study was conducted in Belgium from 1st February 2019 to 1st March 2020. Six paediatric liaison teams (2 in Flanders, 2 in Brussels and 2 in Wallonia) participated in this study. All study material was available in 2 languages, French and Dutch.

This study received the approval of the principal ethical committee, the comité d'éthique hospital-facultaire des Cliniques st Luc, of each ethical committee of the participating hospitals (Hôpital universitaire des enfants Reine Fabiola, Centre hospitalier regional Citadelle, Centre hospitalier chrétien Espérance, Universiteits ziekenhuis Leuven and Universiteits ziekenhuis Gent). It was registered as a national clinical trial under the number B403201837760.

Children/adolescents and their parents followed-up by PLTs were invited to participate in the study by the teams themselves. Families were invited to take part in an interview conducted by members of a PLT. Purposive sampling of all children and adolescents cared for by one of the 6 PLTs in Belgium was carried out with following inclusion criteria : > one year old, not at an imminent end-of-life stage, parents able to understand French or Dutch, parents' and children's consent obtained. Children were excluded if they were in their last days of life, if parents were not able to understand French or Dutch or if they were < 1 year old. For each family, quantitative data were collected through several questionnaires, which were printed on carbon sheets in order to keep the original within the PLT and to send the copy to the research team at IRSS. A code was allocated to each family, in order to respect their confidentiality.

For each of the families who participated in the study, a member of the PLT completed the following questionnaires, one for each interview conducted with the families:

- *An original 35- item sociodemographic and medical questionnaire* including five questions from the Paediatric Palliative Screening Scale (20,21) evaluating a child's life expectancy, the impact of the disease and the impact of treatment on children's daily lives and the level of the child's, parents' and siblings' suffering as perceived by the physician and rated on a 3-point Likert scale. This 35-item questionnaire was created by our research team and previously discussed with representatives of three different paediatric liaison teams.
- The *self- and proxy-report 22-item CPOS-2* (7,18), evaluating children's and parental quality of life. Each item is scored on a six-point Likert scale ranging from 0 to 5. The items are worded in such a way that a higher item score implies a poorer quality of life or, more precisely, a higher burden. The values provided by 12 out of the 22 items have to be reversed to explore the construct in the same direction. The total score produces ranges between 0 and 60 for part A and between 0 and 50 for part B. These scores are transformed to percentage scores that are easier to interpret. The scale contains *reflective* items and *formative* items and should therefore be considered as having a hybrid structure. A cross-cultural translation of the CPOS in French was made according to the guidance of Antunes et al. (22) and De Vet et al. (23) and its face/content validity, acceptability and feasibility was reported in a previous paper. (18) A translation of the CPOS-2 into Dutch was carried out by our research team in close collaboration with representatives of the paediatric liaison teams based in UZ Leuven and UZ Gent. Agreement to use the CPOS-2 in our study was obtained from the tool designers.
- The generic 35-item *Fragebogen für Kinder und Jugendliche zur Erfassung der gesundheitzbezogenen Lebensqualität* (KINDL) (24), customized for three age categories, evaluates children's health-related quality of life through child (self) and parental (proxy) report. The original KINDL was meant as a scale to measure quality of life, i.e. the higher the score, the higher the quality of life. We reversed the scoring of the items and used it as a scale to measure the impact of the disease on the quality of life (the burden). So high scores stand for a high burden and vice versa. The scale was developed to measure quality of life in healthy as well as in ill children. The available translated version in Dutch and in French was used for our study. Agreement to use the KINDL in our study was obtained from the tool designers.
- The parental self-report 17-item *Quality of life in life-threatening illness - family carer* version 2 (QOLLTI-F) (25) assessing parental quality of life. The QOLLTI-F originates from qualitative interviews exploring family burdens but also positive experiences in the caregivers' situation, which are included in the questionnaire. The QOLLTI-F includes seven subscales assessing different domains: environment, patient condition, the carer's own state, carer's outlook, quality of care,

relationships and financial worries. All items have a possible range from 0 to 10. A high total score indicates a good situation (a high quality of life or a low burden). In order for 0 to always indicate the worst situation and 10 the best situation, five items are transposed prior to calculating subscales and total scores. All subscales scores are calculated by taking the mean of the items comprising that subscale. The QOLLTI-F total score is the mean of the subscale scores. A French version of the QOLLTI-F was already available. A Dutch version was produced by our research team in close cooperation with paediatric liaison team members from UZ Leuven and UZ Gent for linguistic and semantic soundness. Agreement to use the QOLLTI-F in our study was obtained from the tool designers.

Statistical analysis

Descriptive statistics were conducted on the sociodemographic data, the CPOS QoL scores and the QOLLTI-F scores.

The association between clinicians' judgement of quality of life and the scores on the QOLLTI-F and CPOS-2 was explored using Spearman's correlation coefficients and boxplots.

We used the highest quartile value of the QOLLTI-F score (quality of life parents) and the highest quartile value of the CPOS-2 score (impact on the quality of life of the family) to define contrasting groups and compare the characteristics of families with higher quartile values with the others. Pearson chi square test and the Fisher exact test were used to investigate significant differences. Analysis was performed with SPPSS 26.O (SPSS inc., Chicago, IL, USA).

Results on the descriptive statistics were presented to each of the six paediatric liaison teams in June 2020 during four different virtual meetings, and their comments and questions enriched the interpretation of the data.

Results

A total of 73 children/adolescents (1-18 years) were included in the study. The main reason for not inviting families to participate in the study, despite them corresponding to inclusion criteria, were lack of time as reported by PLTs. Only five families refused to participate (see Figure 8 in Subchapter 6.1). Table 21 shows the characteristics of included children.

A homogeneous proportion of girls and boys were found in our sample. Few teenagers were included (10%), however, while 65% of children were < 6 years old. Most (61%) of the children included in the study had either a neurological or a metabolic/genetic disease, and few (25%) faced an oncologic disease. Only 19 (26%) of them had verbal capacities. More than half (55%) of the children received artificial nutrition and a fifth needed respiratory support. At time of interview, more than a half (53%) of the children included had been followed-up by a PLT for more than one year. See Table 21.

As shown in table 15 of Subchapter 6.1, we found that our sample was representative for age ranges and gender but not for categories of diseases. In fact, onco-haematological diseases were underrepresented in our sample, compared to the population of children followed-up by paediatric liaison teams.

Regarding socio economic elements, we found that 76% of included children had parents living as a couple, whereas 18% of the children lived in a single-parent family. Forty-five percent of parents reported having been obliged to completely stop their work in order to care for their child at home. This situation especially affected mothers (34%). Ten percent of families included in the study reported benefitting from social or financial assistance.

Table 21. Characteristics of children of families included in the field-study (Total n=73)

Characteristics of children		**N (%)**
Gender	Total	73 (100%)
Female		37 (50%)
Male		36 (50%)
Age	Total	73 (100%)
1-2 years		18 (25%)
3- 6 years		25 (35%)
7-13 years		22 (30%)
14-17 years		7 (10%)
Disease	Total	73 (100%)
Neurology		28 (38%)
Onco-hematology		18 (25%)
Metabolic/genetic		17 (23%)
Neonatology		7 (10%)
Cardiology		3 (4%)
Verbal capacity (able to understand and respond to questions as judged by parents)		19 (26%)
Medical equipment	Gastrostomy	32 (44%)
Artificial nutrition	Naso-gastric feeding tube	8 (11%)
Respiratory assistance	Oxygenotherapy	7 (10%)
	Non-invasive ventilation	6 (8%)
	Tracheotomy	3 (4%)
Intravenous access	Port-a-cath/Broviac	5 (7%)
Length of follow-up by a PLT at time of interview		73 (100%)
0-1 month		0 (0%)
1-3 months		4 (5,6%)
3-6 months		7 (9,9%)
6-12 months		11 (15,5%)
1-2 years		11 (15,5%)
2-3 years		9 (12,7%)
➢ 3 years		18 (25,4%)
Missing data		11 (15,5%)

Child's life expectancy as assessed by paediatrician	73 (100%)
1-6 months	2 (2,7%)
7-11 months	1 (1,4%)
1-2 years	6 (8,2%)
3-5 years	7 (9,6%)
>5 years	7 (9,6%)
Totally unpredictable	45 (62%)
Missing data	5 (7%)

Focusing on quality of life scores, Table 22 shows the mean scores self-reported by children on their quality of life as measured by the CPOS-2 and the KINDL. Parents' proxy-report through CPOS-2 and KINDL are also indicated. Finally, parents' perspectives on their own quality of life as measured by the CPOS-2 part B and the QOOLLTI-F are included, too.

As mentioned earlier, in subchapter 6.1, we found a significant correlation between scores on part A of the CPOS-2, part A (measuring children's QoL) as reported by children and as reported by parents (observed correlation coefficient 0.55, and corrected correlation coefficient 0.79). We also looked at each item in detail by computing correlations between children and parents scores. We found that for questions 1,2,6,8, 10 and 12 of the CPOS-2, the correlations were statistically significant, whereas for the questions 3,4,5,9, and 11, all linked to emotional status, the scores between children and parents differed considerably.

Q3 Is there anything about food that has been bothering you? Q4 Can you tell me if you have been sad? Q5 Can you tell me if you have been happy? Q9 When something bothers you, can you talk to someone about it? Q11 If you had a magic wand, is there something you would you like to change in your family?

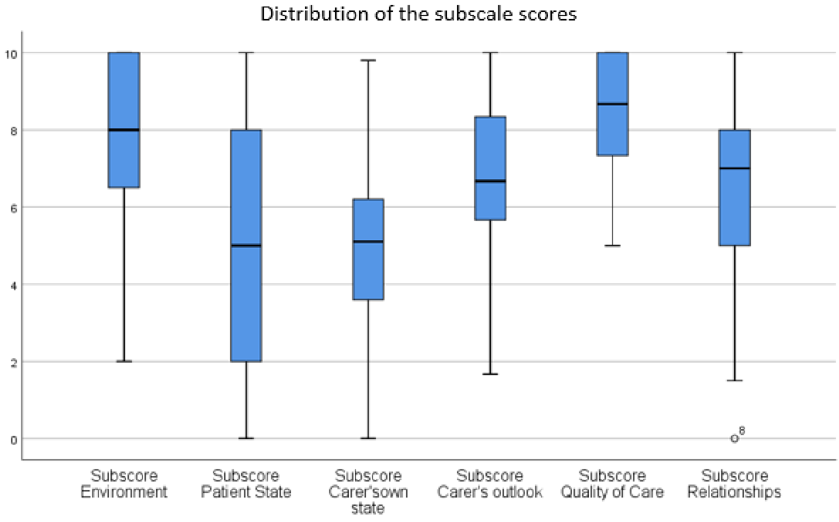
Regarding their quality of life, we found that parents had a relatively low score of quality of life as rated on the QOLLTI-F (mean of 64,11) and on the CPOS -2 part B (mean of 59,15). The scores were the lowest in the 2 subscales of the QOLLTI-F called Patient State and Carer's own state (Figure 18). These 2 subscales correspond to the 2 following questions: "During the last 2 days, the condition of my child, whom I care for, has afflicted me." and "During the last 2 days, he level of control I have on my life has been a problem."

Table 22. Children's and parent's QOL scores as measured by the CPOS-2, the KINDL and the QOLLTI-F questionnaires

	Number n	Converted to mean % and Standard deviation
Children's QOL		
CPOS-2 *reversed score*		
Self-report part A	22	73.19% (15.91)
Proxy-report part A	70	65.15% (16.25)
KINDL reversed score		
Self-report	23	70.97 % (15.31)
Proxy-report	49	58.91% (11.47)
Parental QOL		
CPOS-2 reversed score		
Proxy-report part B	73	59.15 % (12.89)
QOLLTI-F	71	64.11% (13.34)

CPOS-2 Children palliative outcome scale version 2. QOLLTI-F Quality of life in life-limiting illness-family carer QoL Quality of life The CPOS-2 and KINDL scores were reversed to be aligned with the QOLLTI-F scores and reflect the quality of life (highest QoL =100%).

Figure 18. Distribution of the subscales scores of the QOLLTI-F



To put the QoL scores in our study into perspective, Table 23 offers an overview of scores produced by KINDL and QOLLTI-F in other populations.

We found that mean child's KINDL scores were close to the ones found in our study (around 70%). The norm in healthy children being 76.8% (26,27). Two other studies, one in children with diabetes (28), and another one among children with congenital heart disease (29) found that the mean QoL scores were higher than those of healthy children of the same age reference group.

Parental QoL scores found in our study, as measured by the CPOS-2 and the QOLLTI-F, were comparable to the scores found in two other studies conducted among parents who had a child receiving palliative care (30,31). Those studies pointed to the impact of financial, emotional and physical dimensions on parents QoL. Table 23 presents the QoL scores found in different studies who used the KINDL and the QOLLTI-F.

Table 23. Comparison of QoL scores among different studies using the KINDL and the QOLLTI-F

KINDL	**N (age)**	**Disease**	**Results**	**Mean score (SD)**
Khair et al (2012), UK	84 (6-17 y)	Haemophilia	The highest impairments (KINDL) in the 8-to 12-year-old group were in the dimension "school" (55.01 ± 17.2) and self-esteem (59.5 ± 17.1), whereas scores for 6- to 7-year-olds were much higher for these dimensions (75.0 ± 31.0 and 75.0 ± 23.1, respectively)	KINDL TOTAL scores 6-7 y : 77.61 (14.2) 8-10 y : 70.40 (8.9) 13-17 y : 70.38 (12.3)
Hövels-Gürich et al (2007), Germany	40 (5-12 y)	Congenital heart disease	Children 5 to 7 years old reported better QoL (total score) than the same age reference group. Self-reported QoL scores for 8 to 12-year-olds did not differ from those in control subjects in any domain.	Full text not available
Müller-Godeffroy et al (2008), Germany	50 (6-16 y)	Spina Bifida	Children with spina bifida (8–12 y) reported lower HRQoL in all dimensions ("emotional," "self-esteem" and "friends") and total score (medium to large effect sizes). Adolescents reported lower scores on peer relations. Most medical parameters as well as limitations in ADL were not significantly associated with HRQOL. Our findings confirm the results of studies which dispute a linear inverse association between condition severity and HRQOL	8-12 y: 69,6 (95% IC 57,8-67,1) 13-16 y: 69,7 (95% IC 59,6-74,8)

Wee et al. (2005)	30 (mean age: 10.7 +/- 1.35 years	Diabetes mellitus	Overall, children with DM reported better HRQoL than healthy children. Although this appeared counter-intuitive, several explanations are possible: (1) the development of resilience to the disease over time, (2) our subjects are well-managed, (3) response shift, (4) the provision of high quality medical care, (5) compared to normal children, diabetic subjects and their families pay greater attention to health issues.	The reliability coefficients were (overall, scales): KINDL-Kid DM (0.79, 0.44-0.65), KINDL-Kid Healthy (0.71,0.60-0.80), KINDL-Kiddo DM (0.77, 0.37-0.74) and KINDL-Kiddo Healthy (0.84, 0.21-0.79).
QOLLTI-F				
Groh (2013), Germany	40 parents	Various Life-limiting conditions		QOLLTI-F total score before intervention median 5.8 (IQR: 1) After intervention 7.1 (IQR: 1.3) <0.001
Bradford (2012), Australia	10 parents	Various Life-limiting conditions	Two domains of caregiver quality-of-life require further study: their finances and their emotional and physical state	QOLLTI-F total scores Mean IG : 5,4-6,2 Mean CG: 6,6-7

The assessment of the level of distress by the responsible paediatrician

Our second research question concerns the correlation between the appreciation of the well-being of parents and children by the responsible paediatrician, with the quality of life as measured by the QOLLTI- (parents) and the CPOS (children + parents).

We found that for 45 children (62%), their specialist paediatrician estimated that a child’s life expectancy was completely unpredictable (Table 21). Moreover, they perceived that for 67% of the children illness may have a very high impact on their daily activity, and for 34% of them, medical treatment would have a very high impact on the children’s quality of life.

Paediatricians rated the level of parental suffering as very high for 39 parents (57%), whereas the child's suffering was estimated as being very high for 28% of them. For 13% of included children, paediatricians found the level of the child's suffering difficult to estimate. Siblings' suffering was found difficult to estimate in 32% of cases (Table 24).

Table 24. Levels of child's, parent's and sibling's suffering as perceived by the paediatrician (Total of 73 children, 100%)

	Difficult to assess	Low	Medium	Very high	Missing values
Child's level of suffering	12,3%	20,5%	34,2%	26,0%	6,8%
Parents' level of suffering	6,8%	4,1%	30,1%	53,4%	5,5%
Siblings' level of suffering	24,7%	6,8%	31,5%	15,1%	21,9%

We conducted correlation studies to compare children's and parents' perceptions on their QoL (as measured by the CPOS-2 and the QOLLTI-F) with paediatricians' perceptions on their level of suffering. As shown in Table 25 and Figures 19-21, we found that paediatricians frequently find it difficult to assess the level of distress/suffering in siblings (and to a lesser extend in children). But when they make an assessment on children's and parents suffering, their judgment correlates well with scores of the QOLLTI-F as well as of the CPOS (part B for the parents and part A for the children). It should be noted that a high score on the CPOS-2 indicates a high burden or impact on QoL, whereas a high score of the QOLLTI-F shows a high quality of life (low burden or impact of the disease).

Table 25. Correlation of paediatrician's assessment of parental suffering with the CPOS-2 and QOLLTI-F scores

	Level of distress Parents	CPOS-2 score (%) (Part B)	QOLLTI-F score (%)
Level of distress Parents		0.47**	-0.50**
CPOS-2 score (%) (Part B)	*0.61°*		-0.54**
QOLLTI-F score (%)	*-0.56°*	*-0.78**	

Spearman's correlation coefficients

**. Correlation is significant at the 0.01 level (2-tailed)

° Coefficients corrected for attenuation

.

Figure 19. Level of distress/suffering in parents as assessed by the paediatrician in relation to the scores of the QOLLTI-F

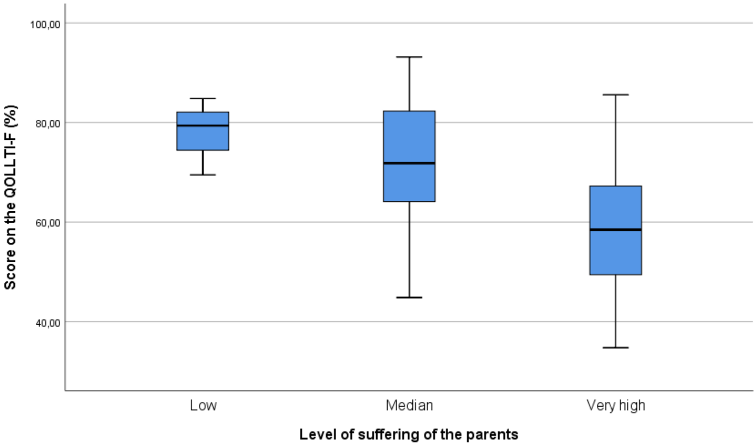


Figure 20. Level of distress/suffering in parents as assessed by the paediatrician in relation to the scores on part B of the CPOS-2

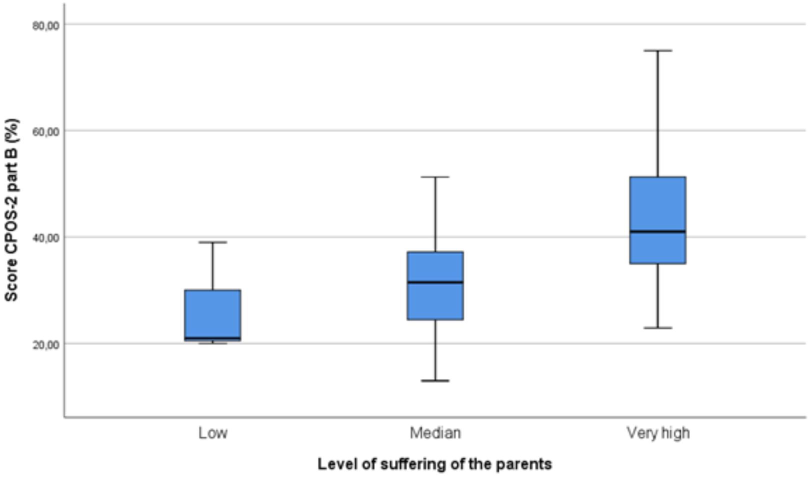
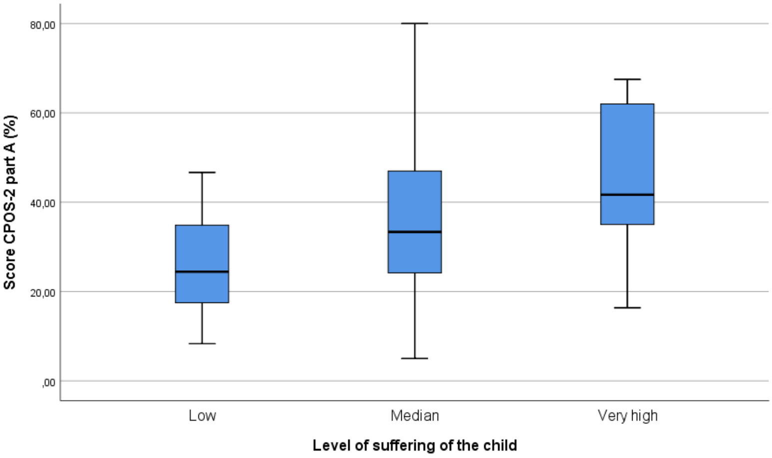


Figure 21. Level of distress/suffering in children as assessed by the paediatrician in relation to the scores on part A of the CPOS-2



Factors associated with QOL as measured by the QOLLTI and the CPOS-2

Finally, as a third focus, we explored which characteristics and background variables were associated with highest quartile scores on the QOLLT (high score = high quality of life) and/or the CPOS-2 (high score = higher impact of the disease or lower quality of life)

As seen on Table 26, we found no association with the age or sex of the child; with the type of disease the child was suffering from nor with the length of the follow-up by the PPC teams. Four parental characteristics were also investigated: socio-economic status, level of education, ability to cope with the situation (as evaluated by the PPC nurse) and the perceived impact on daily life, and no significant association was found (not reported in the table).

The most striking finding was that no association at all was found between all of the indices describing the degree of disability of the child and the quality of life of parents, as measured in our sample by the QOLLTI, and in the family as measured by the CPOS.

The only two factors associated with a high QOLLTI score were the estimation by the paediatrician of the child's and parents' level of suffering.

A high CPOS-2 score (measuring the burden of the disease), was associated with the paediatrician's perception on parents' and child's level of suffering and the need for specific equipment, more specifically the presence of a nasogastric feeding tube and/or the need for oxygen therapy.

Those results, however, must be evaluated with caution because of the small size of our group (n=73).

Table 26. Association between baseline characteristics and background variables and highest quartile QOLLTI-F and CPOS-2 scores

	QOLLTI-F score, highest quartile (n= 17)	QOLLTI-F score, rest of quartiles (n= 56)	P-value	CPOS-2 score, highest quartile (n= 21)	CPOS 2 score, rest of quartiles (n=52)	P value
Age						
1-2 y	2	16	0.55 [a]	6	12	0.46 [a]
3-6 y	6	20		6	20	
7-13 y	7	15		7	15	
14-17 y	2	4		1	5	
> 17 y	0	1		1	0	
Sex Male (n, %)	7 (43%)	29 (51%)	0.31 [b]	10 (47.5%)	26 (50%)	0.53 [b]
Type of pathology						
A= Neurological	8	15	0.10 [a]	6	17	.84 [a]
B= Onco-hematological	1	17		4	14	
C= Metabolic/genetic	6	18		8	16	
D= Neonatal	0	3		1	2	
E= Cardiac	1	0		0	1	
Disability indices						
Verbal capacity						
A=Normal	5	18	0.15 [a]	4	19	0.58 [a]
B= A few words, limited vocabulary	0	5		1	4	
	1	12		4	9	

C= vocalization, sounds, screams	6	16		9	13	
D=no verbal communication at all						
Cognitive capacity						
A= Normal	5	22	0.32 [a]	5	22	.17 [a]
B= Limited (limited verbal interaction)	1	5		0	6	
C= smiles, facial expressions, produces sounds	1	9		4	6	
D= severely limited, severe mental retardation	7	16		9	14	
Mobility						
A= Normal mobility in accordance with age (sitting, walking, etc ...)	3	15	0.65 [a]	2	16	0.26 [a]
B= sits, walks with assistance, axial hypotonia	1	9		3	7	
C= wheelchair or another device needed	6	12		5	13	
D= bedridden, no spontaneous mobilization	5	15		9	11	
Ability to eat						

A= Normal	4	17	0.26 [a]	3	18	0.46 [a]
B= Eats with help	3	6		2	7	
C= Naso-gastric tube	6	8		4	10	
D= Gastrostomy	3	19		9	13	
Duration of follow-up						
0-1 months	1	3	0.73 [a]	1	3	0.69 [a]
1-3 months	0	7		3	4	
3-6 months	2	9		3	8	
6-12 months	2	9		5	6	
1-2 years	3	6		1	8	
2-3 years	5	13		5	13	

[a] P value based on Pearson chi-square test [b] P value based on Fisher exact test.

Discussion

This study provides, for the first time, an overview of the QoL of families followed-up by a PLT in Belgium in 2019. A sample size of 73 children aged 1-18 years and their parents were included. It shows a slight discrepancy between children's and parents' scores on children's QoL, as measured by the CPOS-2 and the KINDL. Especially for items focusing on emotional items, children included in our study self-reported their QoL as higher than their parents did. Children's QoL, as rated by the CPOS-2 and the KINDL, demonstrated a relatively high QoL (mean of 72/100) compared to parents' QoL as, measured by parents themselves, on the QOLLTI-F (mean of 64/100) and the CPOS-2 part B (59/100).

Within comparison to other studies (26-29) we found that the mean child's QoL scores, as rated by the CPOS and the KINDL, were similar. Like the results found in this field-test, those studies suggest that health-related QoL measured by KINDL is not significantly associated with the condition's severity or impaired activities of daily life. Several hypotheses can be provided. Children can adapt over time to their disease and condition, showing an increased resilience. A second reason can be linked to response shift. A third one to the provision of high medical care offered to families. Those studies, however, also indicated, that QoL might be highly influenced by relations with peers, especially for adolescents.

Estimating children's life expectancy is unpredictable and might therefore not be a reliable criterion for referring to a paediatric liaison team, in contrast to the surprise question ("Would you be surprised if your patient would die in the next 6-12 months?") often used as a valid criterion in adult palliative care. In Belgium, the palliative care indicators tool for adults, called PICT, was officially introduced as a referral criterion to palliative care services and includes the surprise question (32). For a paediatric palliative care population, one single prospective cohort study found that the surprise question on child's life expectancy was a highly sensitive prognostic tool for identifying children who are in the last 3 and 12 months of life (33). In another qualitative study conducted among 10 Belgian specialist paediatricians in a single university hospital, not being able to attend school, the intuitive perception of the family's suffering, its ability to cope and the need to assist the child technically with medical equipment at home were the most reported criteria by hospital paediatricians for referral to a paediatric liaison team (34). This is consistent with the findings of this field-study, in which the presence of a medical equipment and the paediatrician's perceptions on a family's level of suffering were statistically significantly associated with a high CPOS-2 score.

In our study, we found that parents often wanted to talk about ancient events they had gone through, rather than limiting themselves to the last two days (as requested by the CPOS, KINDL and QOLLTI-F questionnaires). This suggests that some experiences linked to the announcement of the diagnosis or the care pathway might be remaining or even traumatic. This is in line with the results of two studies looking at parental suffering, measured by the degree of post-traumatic stress disorders (PTSD) when their child faced a serious illness. Indeed, 10% of mothers and 18 % of fathers showed full PTSD, even five years after child's end of cancer treatment (35,36). Might quality of life be associated with level of resilience, and how is parental resilience entangled with adolescent resilience levels? A recent study among cancer patients showed that "higher adolescents and young adults (AYA) distress predicted better maternal resilience, whereas higher maternal distress predicted worse AYA resilience. Thus, processes of resilience between AYAs and their mothers may differ." (37)

The discrepancy between self- (child) report and proxy- (parent) report of children's health-related QoL found in our study was confirmed in other studies (38-42). Those different perceptions were found particularly in psychosocial domains of health-related QoL such as emotional functioning, and these differences were age-related (43-44). Although some authors (45) suggest that a single proxy-report (by parents) would be enough for rating children's pain intensity, for example, other authors recommend always requesting child and parental reports in order to acknowledge the different perceptions of each and to foster the acceptance of the psycho-social dynamics of the child-parent dyad made of loyalty, trust and interdependence (8, 42-43,46-47).

Discrepancies were not found between health care professionals' perceptions and parents' perceptions on their QoL, although results of the qualitative implementation study (see chapter 3.4.4) showed the contrary. If clinical teams perceive a family's quality of life as low, cognitive bias might jeopardize the shared decision-making process. To prevent this, Carnevale et al. suggest recognizing first "the children as active agents, that is, persons who have interests and capacities to participate in discussions and decisions that affect them and other people"(48). He also promotes an "empathic attunement", that he describes as an attempt to "sense the emotional perspective of the other, but also implies a stunning to grasp the person's understanding of the situation to the greatest possible extent" (49). In brief, using patient-centred outcome measurement instruments such as the CPOS-2 might help health care professionals to objectify their intuitions and feelings. On a long-term basis, when used several times with the same family, the CPOS-2 could document the impact of paediatric palliative care on children's and parents' quality of life. We do, however, acknowledge that an outcome measurement instrument might not be able to fully capture one's subjective quality of life, reflected in a score, but might be used as a tool to address sensitive issues, discover unmet needs and facilitate the shared decision-making process.

Limitations

The relatively small sample size included in our study might be a limitation for generalisation of our findings. It represents 24% (n=73/309) of the whole population of children (with the same inclusion criteria) cared for by paediatric liaison teams in Belgium, as reported by them in a timeframe of 12 months. Outcome measure instruments used in our study were not able to allow for the self-report QoL in children with cognitive capacities. This limit has been already reported in other studies. To overcome this barrier, suggestions include relying purely on parental or professional proxies focusing on core outcomes identified for children with severe neurological impairment (50), using pictograms or drawings (51) or communication boards, electronic touch pads or adapted devices (47).

Further studies with larger sample sizes, especially for children with verbal capacities living with an oncological disease, are warranted to better document their perspectives on quality of life and identify potential subgroups.

Conclusion

This study provides, for the first time, an overview of the quality of life of families followed-up by a paediatric liaison team in Belgium in 2019, as assessed by the children's palliative outcome scale-2. Quality of life scores do not seem to be associated with the severity of child's disease.

References

1.Craig F, Abu-Saad Huijer H, Benini F, Kuttner L, Wood C, Feraris PC, Zernikow B. [IMPaCCT: standards of paediatric palliative care]. Schmerz. 2008;22(4):401-8. doi: 10.1007/s00482-008-0690-4.

2. World Health Organization (WHO). Definition of palliative care. 1998. https://www.who.int/cancer/palliative/definition/en/ (consulted 15th June 2020).

3. World Health Organization. Definition of quality of life. 1998 https://www.who.int/healthinfo/survey/whoqol-qualityoflife/en/ (consulted 15th June 2020)

4. Coombes LH, Wiseman T, Lucas G, Sangha A, Murtagh FE. Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-949. doi:10.1177/0269216316649155

5. Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi:10.1542/peds.2018-2379.

6. Huang IC, Shenkman EA, Madden VL, Vadaparampil S, Quinn G, Knapp CA. Measuring quality of life in paediatric palliative care: challenges and potential solutions. Palliat Med. 2010;24(2):175-82. doi: 10.1177/0269216309352418.

7. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02.

8. Harding R, Chambers L, Bluebond-Langner M. Advancing the science of outcome measurement in paediatric palliative care.Int J Palliat Nurs. 2019;25(2):72-79. doi: 10.12968/ijpn.2019.25.2.72.

9. Baker JN, Levine DR, Hinds PS, Weaver MS, Cunningham MJ, Johnson L, Anghelescu D, Mandrell B, Gibson DV, Jones B, Wolfe J, Feudtner C, Friebert S, Carter B, Kane JR. Research Priorities in Paediatric Palliative Care. J Pediatr. 2015;167(2):467-70.e3. doi: 10.1016/j.jpeds.2015.05.002.

10. Donabedian A. Explorations in Quality Assessment and Monitoring: The Definition of Quality and Approaches to Its Assessment. Volume I. Ann Arbor, MI: Health Administration Press; 1980.

11. Witt J, Murtagh FEM, de Wolf-Linder S, Higginson IJ, Daveson BA. Introducing the Outcome Assessment and Complexity Collaborative (OACC) Suite of Measures. 2015 https://www.kcl.ac.uk/cicelysaunders/attachments/Studies-OACC-Brief-Introduction-Booklet.pdf (consulted 13th July 2020).

12. Ghirotto L, Busani E, Salvati M, Di Marco V, Caldarelli V, Artioli G. Researching children's perspectives in paediatric palliative care: A systematic review and meta-summary of qualitative research. Palliat Support Care. 2019;17(1):107-118. doi: 10.1017/S1478951518000172.

13. Namisango E, Bristowe K, Allsop MJ, Murtagh FEM, Abas M, Higginson IJ, Downing J, Harding R. Symptoms and Concerns Among Children and Young People with Life-Limiting and Life-Threatening Conditions: A Systematic Review Highlighting Meaningful Health Outcomes. Patient. 2019;12(1):15-55. doi: 10.1007/s40271-018-0333-5.

14. African Palliative Care Association. Children’s Palliative Outcome scale (APCA C-POS). Manual. 2012.

15. Powell RA, Downing J, Harding R, Mwangi-Powell F & Connor S on behalf of the APCA M&E Group. Development of the APCA African Palliative Outcome Scale. J Pain Sympt Manage. 2007, 33: 229-232.

16. Friedel M, Brichard B, Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium:

A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care. 2018;17(1):77. doi: 10.1186/s12904-018-0324-2.

17. Friedel M, Gilson A, Bouckenaere D, Brichard B, Fonteyne C, Wojcik T, De Clercq E, Guillet A, Mahboub A, Lahaye M, Aujoulat I. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. BMJ Paediatr Open. 2019;3(1):e000547. doi:10.1136/bmjpo-2019-000547.

18. Friedel M, Brichard B, Boonen S, Tonon C, De Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse JM, Aujoulat I. Face and Content Validity, Acceptability, and Feasibility of the Adapted Version of the Children's Palliative Outcome Scale: A Qualitative Pilot Study [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646.

19. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33. doi: 10.1136/bmj.313.7048.29.

20. Bergstraesser E, Hain RD, Pereira J. The development of an instrument that can identify children with palliative care needs: the Paediatric Palliative Screening Scale (PaPaS Scale): a qualitative study approach. BMC Palliative Care 2013; 12:20.

21. Bergstraesser E, Paul M, Rufibach K, Hain RD, Held L. The Paediatric Palliative Screening Scale: Further validity testing. Palliat Med. 2014 Jun;28(6):530-533.

22.Antunes B, Daveson B, Ramsenthaler C, et al. The Palliative care Outcome Scale (POS). Manual for cross-cultural adaptation and psychometric validation. On behalf of the POS Development Team King’s College London, Cicely Saunders Institute, Department of Palliative Care, Policy and Rehabilitation. London: Kings College London, 2012.

23. De Vet HC, Terwee CB, Mokkink LB, Knol DL. Measurement in Medicine. A Practical Guide. Cambridge University Press. 2011.

24. Erhart M, Ellert U, Kurth B, Ravens-Sieberer U, Measuring adolescents’ HRQoL via self and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instru Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006;20(8):755-67. doi: 10.1177/0269216306072764.ment. Health and Quality of Life Outcomes. 2009; 7:77.

25. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006;20(8):755-67. doi: 10.1177/0269216306072764.

26. Khair K, Littley A, Will A, von Mackensen S. The impact of sport on children with haemophilia. Haemophilia. 2012;18(6):898–905.

27. Müller-Godeffroy E, Michael T, Poster M, Seidel U, Schwarke D, Thyen U. Self-reported health-related quality of life in children and adolescents with myelomeningocele. Dev Med Child Neurol. 2008;50(6):456–461pmid:18371093.

28. Wee HL, Lee WW, Ravens-Sieberer U, Erhart M, Li SC. Validation of the English version of the KINDL generic children's health-related quality of life instrument for an Asian population--results from a pilot test. Qual Life Res. 2005;14(4):1193-1200. doi:10.1007/s11136-004-2957-2.

29. Hövels-Gürich HH, Konrad K, Skorzenski D, et al. Long-term behavior and quality of life after corrective cardiac surgery in infancy for tetralogy of Fallot or ventricular septal defect. Pediatr Cardiol. 2007;28(5):346–354.

30. Groh G, Borasio GD, Nickolay C, et al. Specialized paediatric palliative home care: a prospective evaluation. J Palliat Med. 2013;16(12):1588-1594. doi: 10.1089/jpm.2013.0129.

31. Bradford N, Young J, Armfield NR et al: A pilot study of the effectiveness of home teleconsultations in paediatric palliative care. J Telemed Telecare 2012;18(8): 438-442.

32. Desmedt M., Identification du patient palliatif & attribution d'un statut lié à la sévérité des besoins : de l'usage d'un nouvel outil, le PICT, Cliniques universitaires Saint-Luc, 2016.

33. Burke K, Coombes LH, Menezes A, Anderson AK. The 'surprise' question in paediatric palliative care: A prospective cohort study. Palliat Med. 2018;32(2):535-542. doi:10.1177/0269216317716061.

34. Matraka, Nora. Critères d'identification des enfants nécessitant l'intervention de l'équipe de liaison en soins palliatifs pédiatriques selon les médecins des Cliniques Universitaires Saint-Luc : analyse qualitative et test d'un outil d'aide à la décision : la Paediatric Palliative Screening Scale. Faculté de santé publique, Université catholique de Louvain, 2018. Prom. : Aujoulat, Isabelle ; Friedel, Marie.

35. Ljungman L, Boger M, Ander M, Ljótsson B, Cernvall M, vonEssen L. et al. Impressions That Last: Particularly Negative and Positive Experiences Reported by Parents Five Years after the End of a Child's Successful Cancer Treatment or Death. PLoSONE. 2016;11(6):e0157076. doi:10.1371/ journal.pone.0157076.

36. Ljungman L, Hovén E, Ljungman G, Cernvall M, von Essen L. Does time heal all wounds? A longitudinal study of the development of posttraumatic stress symptoms in parents of survivors of childhood cancer and bereaved parents. Psychooncology. 2015;24(12):1792-8. doi: 10.1002/pon.3856.

37. Lau N, Yi-Frazier JP, Bona K, Baker KS, McCauley E, Rosenberg AR. Distress and resilience among adolescents and young adults with cancer and their mothers: An exploratory analysis. J Psychosoc Oncol. 2020;38(1):118-124. doi: 10.1080/07347332.2019.1656317. Epub 2019 Aug 28.

38. Khadka J, Kwon J, Petrou S, Lancsar E, Ratcliffe J. Mind the (inter-rater) gap. An investigation of self-reported versus proxy-reported assessments in the derivation of childhood utility values for economic evaluation: A systematic review. Soc Sci Med. 2019;240:112543. doi: 10.1016/j.socscimed.2019.112543. Epub 2019 Sep 17.

39. Silva N, Crespo C, Carona C, Bullinger M, Canavarro MC. Why the (dis)agreement? Family context and child-parent perspectives on health-related quality of life and psychological problems in paediatric asthma. Child Care Health Dev. 2015;41(1):112-21.

40. Lim Y, Velozo C, Bendixen R. The level of agreement between child self-reports and parent proxy-reports of health-related quality of life in boys with Duchenne muscular dystrophy. Qual Life Res. 2014; 23(7): 1945–1952.

41. Sattoe J, van Staa A, Moll H. The proxy problem anatomized: child-parent disagreement in health related quality of life reports of chronically ill adolescents. Health Qual Life Outcomes. 2012; 10: 10.

42. Vetter TR, Bridgewater CL, McGwin G. An observational study of patient versus parental perceptions of health-related quality of life in children and adolescents with a chronic pain condition: who should the clinician believe? Health Qual Life Outcomes 2012; 10: 85.

43. Jardine J, Glinianaia SV, McConachie H, Embleton ND, Rankin J. Self-reported quality of life of young children with conditions from early infancy: a systematic review. Paediatrics. 2014;134(4):e1129-48. doi: 10.1542/peds.2014-0352.

44. Lee YC, Yang HJ, Lee WT, Teng MJ. Do parents and children agree on rating a child's HRQOL? A systematic review and Meta-analysis of comparisons between children with attention deficit hyperactivity disorder and children with typical development using the PedsQLTM. Disabil Rehabil. 2019;41(3):265-275. doi: 10.1080/09638288.2017.1391338. Epub 2017 Oct 23.

45. Fayed N, Avery L, Davis AM, et al. Parent Proxy Discrepancy Groups of Quality of Life in Childhood Epilepsy. Value Health. 2019;22(7):822-828. doi:10.1016/j.jval.2019.01.019.

46. Weaver MS, Darnall C, Bace S, et al: Trending Longitudinal Agreement between Parent and Child Perceptions of Quality of Life for Paediatric Palliative Care Patients. Children (Basel) 2018;4,8. pii: E65.

47. Nelson KE, Feinstein JA, Gerhardt CA, Rosenberg AR, Widger K, Faerber JA, Feudtner C. Emerging Methodologies in Paediatric Palliative Care Research: Six Case Studies. Children (Basel). 2018;5(3):32. doi: 10.3390/children5030032.

48. Carnavale FA. A thick conception of children's voices: a hermeneutical Framework for childhood research. Int J of Qual Res. 2020;19. Epub June 19, 2020.

49. Carnavale FA. A conceptual and moral analysis of suffering. Nursing ethics. 2009; 16(2):174-183.

50. Ribbers S, Wager J, Hartenstein-Pinter A, Zernikow B, Reuther M. Core outcome domains of paediatric palliative care for children with severe neurological impairment and their families: A qualitative interview study. Palliat Med. 2020;34(3):309-318. doi:10.1177/0269216319885818

51. Sourkes BM. Children's Experience of Symptoms: Narratives through Words and Images. Children (Basel). 2018;5(4):53. doi:10.3390/children5040053

6.3. FROM INTUITION TO MEASUREMENT: A QUALITATIVE STUDY AMONG PAEDIATRIC LIAISON TEAMS EXPERIENCES OF USING THE CHILDREN'S PALLIATIVE OUTCOME SCALE - VERSION 2 (CPOS-2)

Background and objectives: Paediatric palliative care aims to improve quality of life of children facing life-limiting conditions, but outcome measurement instruments are seldom used by clinical teams. Effective implementation of outcome measurement instruments is needed. This field-study, conducted in 2019, aims to understand how paediatric liaison teams experienced using a standardized instrument for the evaluation of children's and parents' quality of life. More specifically, we aimed to collect information on the meaning they experienced when using the CPOS-2, on perceived barriers and facilitators, and on possible consequences on their clinical practice.

Methods: During family interviews conducted by members of paediatric liaison teams, outcome measurement instruments were used (CPOS-2, KINDL, QOLLTI-F). After each interview, a standardised 7-item questionnaire was filled-in by the interviewer exploring their impressions. Furthermore, a focus group was conducted by the researcher (MF) in each of the six paediatric liaison teams. Qualitative analysis of transcripts was carried out.

Results: Four main themes emerged from the questionnaires and the focus groups: meaning of using outcomes measurement instruments, impact on care a sperceived by teams, resources identified and difficulties faced. Using outcome measures seems to deepen the relationship between families and health care professionals. Beyond the metrics, the CPOS-2 is perceived as a tool to identify overlooked domains, to introduce difficult conversations, to objectify intuitions and to deepen the relationships with families.

Conclusion: Our results showed an overall enthusiastic participation of most of PLTs to using the CPOS-2, and major benefits for children and parents who were included in the study, as reported by PLTs. Nevertheless, as sensitive topics may arise when using outcome measurement instruments, some precautions must be taken to ensure the emotional security of both families and teams.

Introduction

Paediatric palliative care aims to enhance the quality of life of children facing life-limiting conditions, but research has shown that no outcome measures have yet been validated for this group of children (1,2).

Yet, outcome measure instruments are needed in the field of palliative care and their implementation in clinical care would improve the quality of palliative care and the satisfaction of patients and their families (3-6).

Several challenges to the effective use of outcome measures in clinical palliative care have been reported. These include the lack of a coordinator, the consideration of cognitive and emotional processes of each individual and the importance of training/education before implementation (7-8).

There is a growing interest in the principles of implementation research to address the challenges of the “know–do gap in real-world settings” and linking research and practice. This type of research “uses multiple disciplines and methods and emphasises partnerships between community members, implementers, researchers, and policy makers.” (p.2214, 9)

In Belgium, paediatric liaison care teams (PLTs) provide curative and palliative care in a seamless way to ensure continuity of care across all care settings (10). Since 2010, structural funding ensures the main activities of five PLTs in Belgium. These provide free of charge care 24/7 to all children facing life-limiting or life-threatening conditions, and to their families. A needs assessment conducted in 2016 among five PLTs (see subchapter 3.1.) indicated that PLTs strive to ensure children’s quality of life through a family-centred approach by managing pain and other distressing symptoms and promoting the overall well-being of all family members (10). Apart from using pain scales, they declared that they were not using instruments to measure children’s quality of life. Instead they reported pursuing their goal “by gut feeling”, on an intuitive basis. However, PLTs showed great interest in participating in the development of an outcome measurement tool able to assess children’s and parents’ quality of life. In addition to the five structurally-funded PLTs, we included also one team that is not recognized and not funded as a paediatric liaison team, to increase the potential recruitment of families. This added team has a slightly different organization of care. It provides mainly ambulatory care for oncological patients and does not offer home visits on a 24/7 basis.

Based on this needs assessment, the development of an outcome measurement instrument was decided upon in collaboration with the paediatric liaison teams. After a cross-cultural validation from English to French, we conducted a pilot-test of the African palliative care Association children’s palliative care measurement instrument, APCA-CPOS (3). During this pilot-test, the instrument was further developed in relation with quality of life domains elicited by the children themselves and their parents. Its face/content validity, its acceptability among 14 children and 19 parents and its feasibility of use for paediatric liaison teams was assessed (11). See chapter 5.

In line with the recommended steps for the development of an outcome measure instrument, we conducted a wider field-test among all six paediatric liaison teams. This field-test pursued three objectives:

Documenting the psychometric properties of the further developed CPOS-2 (as reported in subchapter 6.1);

Describing the quality of life of children and their parents followed-up by paediatric liaison teams (as reported in subchapter 6.2);

Understanding the experiences of paediatric liaison teams when using the CPOS-2 (this subchapter 6.3).

In this field-study, in which 73 families participated, we wanted to triangulate the perspectives of children, parents and healthcare teams (PLTs) by using quantitative and qualitative methods to better understand how quality of life could be measured among children and their parents.

Families were invited by PLTS to participate in interviews. Interviews took place during a routine planned visit at home or in hospital, provided parents had given their consent and children their assent. Children could choose to be interviewed alone or in presence of their parents. PLTs completed the socio-demographic questionnaire and passed the CPOS-2, then the Fragebogen für KINDer und Jugendliche zur Erfassung der gesundheitsbezogenen Lebensqualität, called KINDL (12), a generic health-related quality of life questionnaire, and finally the Quality of life in life threatening illness-family caregiver, called QOLLTI-F (13), which assesses the Quality of life of family carers.

As explained previously, PLTs are not familiar with outcome measurement instruments. As PLTs showed interest in using outcome measure instruments in their routine clinical care, we found it important to document their perceptions of using the CPOS-2.

This chapter describes the qualitative study we conducted to address the third objective of the field-test.

Our specific objective for this study was to understand how paediatric liaison teams experienced using a standardized instrument for the evaluation of quality of life in their clinical context. More specifically, we aimed to collect information on what it meant to them, on perceived barriers and facilitators, and on possible consequences for their clinical practice.

Methods

Data collection

Qualitative data on the implementation process were collected between February 2019 and February 2020 through a standardized 7-item questionnaire and focus groups.

A standardized 7-item questionnaire completed by the PLT member after each interview conducted with families. Questions looked at PLT members' feelings and experiences upon using outcome measures and their perceived impact on care provided. The 7 open-ended questions were:

1. How was the overall interview with the child and his parents?
2. How do you feel immediately after the interview?
3. Did anything during the interview surprise you, challenge you?
4. Do you feel that you have learned something new about the family?
5. Do you think that the interview changed the way you will care for the child and the family?
6. What was easy or difficult about using those questionnaires?
7. Are there any elements that you would suggest changing to improve the questionnaires?

Adiidtionnaly, a focus group was conducted in each of the six PLTs, led by the researcher (MF) and based on a four-question interview guide. These focus groups were conducted between April 2019 and December 2019 and comprised a total of 27 PLT members (5 paediatricians, 20 paediatric palliative nurses and 2 coordinators/social workers). The objective was to confirm and to better understand the results issued from the PLTs' written responses to the questionnaires that they had filled in after each interview with families.

The 4 questions of the interview guide were:

1. How is the study going so far for the team and the families?
2. What are the difficulties and resources encountered?
3. How does this change (or not) the way you care for families?
4. How do you plan to continue the study?

The focus groups were scheduled halfway through the data collection among families. This timing was chosen in order to explore what PLTs experienced while conducting the interviews among families but also to provide additional information on recruitment, instruments or general interview guidance. A focus group interview is described as "a carefully planned discussion designed to obtain perceptions on a defined area of interest in a permissive, non-threatening environment." (14)

Data analysis

A thematic analysis was carried out on the results of the questionnaires, highlighting the feelings, resources and difficulties faced by the PLTs during the interviews with families. A first scheme of categories was derived and discussed with a second researcher (Isabelle Aujoulat).

Regarding the focus groups, a synthesis of the verbatim was sent to each PLT to validate the faithful transcript of the focus group. Only one PLT corrected one item (gender of a child wrongly captured during the focus group). Thematic analysis of the data was based on four main categories, from which three referred to predefined categories (impact on care, resources discovered, difficulties faced), whereas one new category emerged (meaning-making process).

The results of the analysis of the questionnaires and focus groups were compared, discussed with a second researcher (Isabelle Aujoulat), then validated through four virtual meetings in June 2020 that included a total of 12 representatives of the six PLTs (5 paediatricians, 5 paediatric palliative nurses, one psychologist, one coordinator/social worker).

Results

Experience of conducting the interviews

Looking at the process, we found that the time for conducting the interviews varied from 15 to more than 60 minutes, including the use of the CPOS-2, the KINDL and the QOOLTI-F questionnaires. The use of the CPOS-2 alone took around 10 to 15 minutes, according to PLTs.

Forty percent (n=29) of the interviews were conducted in the children’s homes and 60% took place in hospital, either during day-patient stays or during consultations (n=34).

We found that, in total, 15 different health care professionals, mostly female, conducted the interviews with families (n=73). The majority of these professionals were paediatric nurses trained in paediatric palliative care. Across four teams, all the interviews were led by paediatric nurses within the PLTs. In one PLT, one paediatrician carried out two interviews and in another, the child’s hospital psychologist conducted all interviews (n=8).

Table 27 shows the characteristics of the interview process.

Table 27. Characteristics of the interview process

Duration of interviews (on a total of n=73)	N (%)
15-30 min	18 (25%)
31-45 min	20 (27%)
46-60 min	16 (22%)
More than 60 min	7 (10%)
Not reported	12 (16%)
Place of interviews (on a total of n=73)	
Home	29 (40%)
Hospital	28 (38%)
Day Hospital/consultation	4 (5%)
Office of PLTs	2 (3%)
Not reported	10 (14%)
Characteristics of interviewers (on a total of n=15)	
Gender: female	13 (86%)
Paediatric Nurses	13 (86%)
Paediatrician	1 (7%)
Psychologist	1 (7%)

We present hereafter the results from the questionnaires and the focus groups, classified in the four areas (see Figure 22) and illustrate them with quotes from PLT members.

Meaning for paediatric liaison teams

An important finding is related to an emergent theme, that of the meaning given by the PLTs to the use of outcome measurement instruments. PLTs affirmed that these had promoted the quality and the depth of their relationship with the families, shaping the way for discussing some sensitive issues, such as death and dying.

> *"There's a kind of increased confidence. Since the interview, the mother calls me more often, which she didn't do before" (PLT1).*
>
> *"It puts you in touch... with his entire extended network. "(PLT1)*
>
> *"Doing these questionnaires opened the door to the family's feelings and problems. "(PLT3)*

This shows that the interview was considered by both sides as a gift. Furthermore, they recognized the added value of using outcome measure instruments in order to promote the visibility of their team and to reinforce their legitimacy in the eyes of decision-makers.

> *"Using these questionnaires helps us to be more aware of deeper things. We enrich our experience and it helps us to individualize family care. We also move from an intuitive experience to identifying certain elements, naming them (PLT3).*

Impact on care as perceived by paediatric liaison teams

PLTs affirmed that using outcome measure instruments had changed the way they cared for children and their families. Through the interview process, PLTs discovered hidden aspects of parental experiences, which led them to change their perception of the families' resources and coping strategies (characterised by PLTS as peace radiating from the family, courage, perseverance, confidence for the future, emotional intelligence, faith) or, on the other hand, the families' suffering (defined by PLTs as sadness, worries, anxiety, fears, isolation, distress, need for respite care, unrecognized suffering of siblings, intrusion of other family members leading to less intimacy, financial worries). Mothers who had been perceived by PLTs prior to the interview as strong, were perceived for the first time as vulnerable. New resources were discovered by PLTs for families who had been labelled by them as experiencing high level of suffering.

Asking questions included in the CPOS made them more aware of the complex situations faced by families, sometimes modifying the perception they had of the presumed level of a family's quality of life.

> *"What struck me most was the positive energy in this family. When you see the child with all these problems, you think... where is the quality of life? And actually, no... in the end, quality of life criteria are so individual. Often we project ourselves... " (PLT1)*
>
> *"I was very challenged by one father's attitude, zenitude... when his child was seriously ill, he told me that he assessed his own quality of life as good. Whereas I would have said that his quality of life was horrible! "(PLT2)*

This led to an improved desire among PLTs to better care for families by relying on the families' own resources and on the interdisciplinary team members, such as psychologists, social workers or education therapists. Two specific areas that PLTs reported being more aware of after using the questionnaires related to sibling needs and parental respite care needs.

> *Measuring outcomes helps us to improve the quality of care, to better understand families' experiences, to be more professional or very caring (that's synonymous for us). Measuring outcomes helps us to improve our sensitivity, to pay more attention to the small details. "(PLT3)*

They stated that they learned from each interview, building a "toolkit" (comprising all their professional experiences) step by step, helping them to better address family needs during conversations.

Interestingly, in one team, interviews were conducted exclusively by the psychologist, who reported not seeing any change in care provided.

Resources and discoveries made by paediatric liaison team

PLTs experienced predominantly positive feelings in administering the questionnaires among the participating families. They discovered that parents valued expressing themselves, and perceived interviews as a relief rather than a burden. PLTs identified resources such as the openness with which the families shared their experiences and feelings and the fact that interviews were perceived by PLTs as a tool for continuing to take care of families.

> *"The mother said, are you coming for me?" "She felt like I was taking care of her. The interview was like a time to be listened to, to be supported. "(PLT 1)*

Results also showed that interviews gave families the opportunity to express their gratitude to PLTs for the follow-up received and helped PLTs to discover how positively parents valued their support, which overall strengthened trust and confidence in their relationships, from both sides.

The results suggest that flexible teams who could find solutions to unexpected situations, such as organizational changes (medical coordinator left, staff shortage,…) were found more receptive to use the CPOS-2.

The ambivalent role of hope was abundantly discussed among one PLT. They explained how important it was to leave room for parental hope, and not judging it as a form of denial.

> *“Some families tell us: "Leave us hope! Sometimes it is hope that a miracle will happen (to heal their child), but not always. Sometimes that hope means: leave us something we can fight for, something we can endure. It is the hope that the bond with the child will stay. The hope that this day could be a good day. Sometimes it's the hope that the child will be able to go home or stay at home. The hope of being together with the child. One mother kept her little one skin-to-skin, every day, every minute. Families can experience hope AND be very clear about what is happening. They know, they are aware AND they continue to hope." (PLT3)*

Difficulties faced

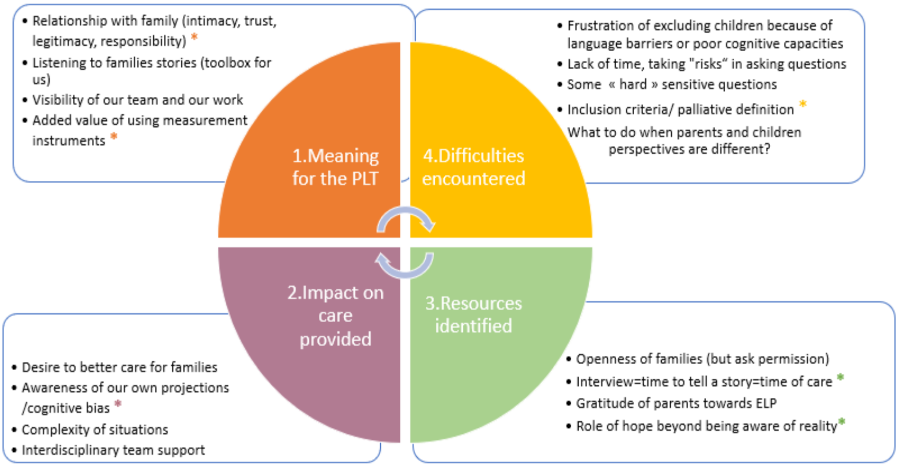
Difficulties encountered with the questionnaires, according PLTs, included the fact that non-verbal children with neurological conditions were unable to understand or answer questions, as well as language barriers faced by some parents, making them ineligible to be included in the study. This was perceived as frustrating by PLTs. Some PLT members expressed some concerns about asking questions on the psychosocial wellbeing of the child when his health state was deteriorating or when children's and parents' report (CPOS and KINDL) didn't correlate. One PLT member raised doubts about the capacity of rating quality of life with a numerical value, judging it as probably too restrictive. The last difficulty mentioned focused on the blurred definition of a palliative stage, leading one team to hesitate on how to apply inclusion criteria. PLTs reported a variety of emotions when conducting the interviews with the families. Asking the questions included in the CPOS were perceived by some of them as a risk and should be therefore preceded by families for permission to encroach upon their privacy. It is also considered important to carefully consider the title of the CPOS. A disclosure of the palliative stage must be planned before using an instrument including the term "palliative". In some clinical contexts, the palliative culture optimizes the smooth implementation of outcome measures. PLTs also pointed out the importance of reflecting on the best timing to assess outcomes among children and their parents. It seems whether the CPOS should be only used among well-known families or whether it is an appropriate means for learning from each other right at the beginning.

> *"What is difficult is the 'palliative' terminology, which is not understood by everyone in the same way. The terms are not trivial. It scares doctors more than it scares families. We sometimes get into fights; we don't have the same opinions... Yet we know that we should introduce the palliative paediatric liaison team early, as soon as the diagnosis is announced, but we don't do it. We should no longer use the term palliative, but only continuing care. That says it all, we are there, we are there no matter what. "(PLT4)*

> *"In summary, we included only children who were very, very sick, in palliative phase, but not children who receive liaison care. "(PLT5)*

> *"I think we're choosing patients... who to include in the study... that would be a selection bias... but no, we're meeting the criteria, look. We exclude when: less than a year old, when you're at the end of your life, when you don't speak the language... So it's actually good! We don't choose! "(PLT2)*

Figure 22. Synthesis of results obtained from questionnaires completed by paediatric liaison teams after each interview with families (n=70) and results from the focus groups among paediatric liaison team members (n=27 PLT members)



Discussion

Our results showed an overall enthusiastic participation of most of PLTs to use the CPOS-2 and major benefits for children and parents who were included in the study, as reported by PLTs. Using outcome measures seems to deepen the relationship between families and health care professionals.

We would like to discuss more specifically two themes to consider when implementing outcome measurement instruments among clinical teams and compare them to the existing literature. They focus on methods and on the sense-making process.

Precautions regarding methods

We found a discrepancy between parental and professional perceptions on children's or parents' quality of life. This is confirmed in other studies, which indicated that health professionals' implicit cognitive bias on under- or over-estimating children's and parental quality of life might negatively influence the quality of care provided (15,16). Because this discrepancy is commonly observed, attention should be focused on the way of dealing with those different perspectives, and of addressing this issue in a sensitive and professional way, maybe in using other interdisciplinary team resources such as a psychologist, or relying on supervisions (17,18).

Interestingly, PLTs perceived the CPOS as a discussion prompt list to raise sensitive topics (e.g. on advance care planning or end of life care issues) or to discover meaningful domains which had been left hidden. Important work was done by Ekberg et al. (19) who established for the first time a discussion prompt list to use by parents themselves and to help them engage in difficult conversations with clinical teams. Although hope was not an area directly addressed in the CPOS-2, it was expressed by families while they were responding to the questions of the CPOS-2. We discovered that parents needed to and took the opportunity to talk about different issues which might go beyond the items in the CPOS-2. This suggests that the CPOS-2 was perceived as an instrument to initiate conversations which might expand to several meaningful themes, specific to each family.

Our results suggest that when nurses were designated to use the CPOS, the implementation process was facilitated. As they are the main care co-ordinators within a PLT, they have frequent contact with children. On the other hand, according to the psychologist who conducted the interviews with the CPOS-2, it didn't have an impact on care, as she declared being accustomed to addressing similar areas already in her consultations with families.

Another important issue raised by PLTs concerns the inclusion of non-verbal children and generally families who encounter language barriers. These are perceived as potentially even more vulnerable families who should have equal access to opportunities to share their experiences. Attention should be placed on translating the CPOS into Arabic and Turkish to maximise the inclusion of those families, because PLTs reported that a high proportion of families with language barriers were not invited to participate in the study.

Sense-making process on using outcome measurement instruments

Sense-making is the process by which people give meaning to their collective experiences. It has been defined as "the ongoing retrospective development of plausible images that rationalize what people are doing" (p.409, 20).

An overall result focused on the surprises experienced by the teams when discovering resources and difficulties faced by families, but also unexplored areas of families' needs. The qualitative data suggests that using outcome measures might help professionals to reframe their initial perceptions of parental experiences and coping strategies by identifying their implicit cognitive bias (projections) and mitigating them. By using outcome measures, paediatric liaison teams became aware of those cognitive biases and this led them to perceive the instrument as a means of stepping back from their initial perceptions, from an intuitive perspective to a more objective one.

Another benefit perceived by the PLTs when using the CPOS consisted of building a stronger connection with families. In fact, during the interviews families also expressed their feeling of gratitude for the care they were receiving. This finding is confirmed by another study, which stated that using outcome measures might foster mutual trust and nurturing the relationship, elements considered as paramount in paediatric palliative care (Mitchell et al. 2020). A mutual exchange between families and health care teams while using the CPOS was observed. We observed a gift/counter gift dynamic, characterized by parental expression of gratitude towards PLTs and their experience of the interview as a moment of care, counterbalanced by PLTs' impression of learning from the experiences of families. Although this phenomenon, in which each part may perceive a benefit (parents and teams) from each other is under-documented, its understanding might facilitate the implementation process of outcome measures and should be further researched.

Involving the PLTs throughout the study and conducting an iterative evaluation of the implementation process at several points by integrating feedback-driven adaptations of the interventions is described as effective for practice change. Supporting team communication, particularly, helps collective and individual sense-making and helps to understand the interaction between process, context and outcomes (21).

In synthesis, several elements might have facilitated the use of CPOS-2 among paediatric liaison teams: whether paediatric nurses conducted the interviews during their planned visits, whether PLTs perceived an enrichment to their practice when listening to families' stories learnt from their experience, and whether PLTs were flexible and could overcome unexpected events impacting their organization. Conversely, a high workload due to staff shortages and/or changes, the absence of a medical co-ordinator and a lack of a palliative culture were identified as important barriers to the use of outcome measurement instruments. Those findings are consistent with other studies (22,23).

Conclusion

This qualitative study describes the experiences of paediatric liaison teams while using an outcome measurement instrument, and the meanings associated with such a process. Overall, the use of the CPOS-2 was perceived by PLTs as an important means to deepen the relationships with families, facilitating difficult conversations. Using the CPOS-2 is, however, also perceived by some PLTs as a risk, leading to the expression of deep emotions and feelings, and should therefore be carefully monitored within an interdisciplinary team.

References

1.Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019 Jan;143(1). pii: e20182379. doi: 10.1542/peds.2018-2379. Epub 2018 Dec 7.

2. Coombes LH, Wiseman T, Lucas G, Sangha A, Murtagh FE. Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use. Palliat Med. 2016;30(10):935-949. Epub 2016 May 31.

3. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02. Epub 2018 Jun 19.

4. Guo P, Gao W, Higginson IJ,Harding R. J Palliat Med. 2018;21,4:414-414.http://doi.org/10.1089/jpm.2017.0674.

5. Harding R, Chambers L, Bluebond-Langner M. Advancing the science of outcome measurement in paediatric palliative care. Int J Palliat Nurs. 2019;25(2):72-79. doi: 10.12968/ijpn.2019.25.2.72.

6. Bausewein C, Daveson BA, Currow DC, Downing J, Deliens L, Radbruch L, Defilippi K, Lopes Ferreira P, Costantini M, Harding R, Higginson IJ. EAPC White Paper on outcome measurement in palliative care: Improving practice, attaining outcomes and delivering quality services - Recommendations from the European Association for Palliative Care (EAPC) Task Force on Outcome Measurement. Palliat Med. 2016;30(1):6-22. doi: 10.1177/0269216315589898.

7. Bausewein C, Simon ST, Benalia H, Downing J, Mwangi-Powell FN, Daveson BA, Harding R, Higginson IJ; PRISMA. Implementing patient reported outcome measures (PROMs) in palliative care--users' cry for help. Health Qual Life Outcomes. 2011;9:27. doi: 10.1186/1477-7525-9-27.

8. Antunes B, Harding R, Higginson IJ; EUROIMPACT. Implementing patient-reported outcome measures in palliative care clinical practice: a systematic review of facilitators and barriers.Palliat Med. 2014;28(2):158-75. doi: 10.1177/0269216313491619.

9. Theobald S, Brandes N, Gyapong M, El-Saharty S, Proctor E, Diaz T, Wanji S, Elloker S, Raven J, Elsey H, Bharal S, Pelletier D, Peters DH. Implementation research: new imperatives and opportunities in global health. Lancet. 2018;17;392(10160):2214-2228. doi: 10.1016/S0140-6736(18)32205-0.

10. Friedel M, Brichard B, Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care. 2018;17(1):77. doi: 10.1186/s12904-018-0324-2.

11. Friedel M, Brichard B, Boonen S, Tonon C, De Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Namisango E, Degryse JM, Aujoulat I. Face and Content Validity, Acceptability, and Feasibility of the Adapted Version of the Children's Palliative Outcome Scale: A Qualitative Pilot Study [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646

12. Erhart M, Ellert U, Kurth BM, Ravens-Sieberer U. Measuring adolescents' HRQoL via self-reports and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument. Health Qual Life Outcomes. 2009;7:77. doi: 10.1186/1477-7525-7-77.

13. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006;20(8):755-67.

14. Krueger RA & Casey MA (2009), Focus groups: A practical guide for applied research (4th ed.). Thousand Oaks, CA: Sage.

15. Callaghan KA, Fanning JB. Managing Bias in Palliative Care: Professional Hazards in Goals of Care Discussions at the End of Life. Am J Hosp Palliat Care. 2018;35(2):355-363. doi: 10.1177/1049909117707486.

16. FitzGerald C, Hurst S. Implicit bias in healthcare professionals: a systematic review. BMC Med Ethics. 2017 Mar 1;18(1):19. doi: 10.1186/s12910-017-0179-8.

17. Jalmsell L, Lövgren M, Kreicbergs U, Henter JI, Frost BM. Children with cancer share their views: tell the truth but leave room for hope. Acta Paediatr. 2016;105(9):1094-9. doi: 10.1111/apa.13496.

18. Feudtner C, Carroll KW, Hexem KR, Silberman J, Kang TI, Kazak AE. Parental hopeful patterns of thinking, emotions, and paediatric palliative care decision making: a prospective cohort study. Arch Pediatr Adolesc Med. 2010;164(9):831-9. doi: 10.1001/archpaediatrics.2010.146.

19. Ekberg S, Herbert A, Johns K, Tarrant G, Sansone H, Yates P, Danby S, Bradford NK. Finding a way with words: Delphi study to develop a discussion prompt list for paediatric palliative care. Palliat Med. 2020;34(3):291-299. doi: 10.1177/0269216319888988. Epub 2019 Nov 22.

20. Weick K, Sutcliffe KM, Obstfeld D. Organizing and the process of sensemaking. Organization Science 2005;16(4): 409–421.

21. Luig T, Asselin J, Sharma AM, Campbell-Scherer DL. Understanding Implementation of Complex Interventions in Primary Care Teams. J Am Board Fam Med. 2018;31(3):431-444. doi: 10.3122/jabfm.2018.03.170273.

22. Damschroder LJ, Aron DC, Keith RE, et al. Fostering implementation of health services research findings into practice: a consolidated framework for advancing implementation science. Implement Sci. 2009;4:50.

23. Hower KI, Vennedey V, Hillen HA, Kuntz L, Stock S, Pfaff H, Ansmann L. Implementation of patient-centred care: which organisational determinants matter from decision maker’s perspective? Results from a qualitative interview study across various health and social care organisations. BMJ Open. 2019;9:e027591. doi:10.1136/bmjopen-2018-027591.

Key messages of chapter 6

The further developed 22-item CPOS-2 demonstrates satisfactory reliability and validity. It seems able to evaluate children’s and parents’ QoL in a paediatric palliative care context. Compared to other, longer, generic quality of life questionnaires, it seems to evaluate the same construct, namely quality of life. The QoL scores do not seem to correlate with the severity of a disease. Paediatricians’ perceptions on familyies’ suffering correlate with QoL scores. Beyond the metrics, paediatric liaison teams perceive the CPOS as a useful roadmap to open the dialogue with families on sensitive issues, to identify hidden needs and to individualize care plans.

PART III DISCUSSION AND CONCLUSION

CHAPTER 7. GENERAL DISCUSSION

Main results

Exploring and measuring the quality of life (QoL) of children facing life-limiting conditions was a challenge in Belgium, where this topic had never been researched before.

Our goal was to make the experience of those children and their families more visible, by documenting their perspectives on quality of life. We aimed to give children a voice, to value parents' experiences and to listen to their usual health care professionals on how they perceived outcome measurement instruments. Using quantitative and qualitative methods, our research turned out to be a co-constructed shared pathway with paediatric liaison teams to further develop an outcome measurement instrument, a second version of the children's palliative outcomes scale (CPOS-2), allowing uncertainty and fine-tuning via an iterative process.

Our research provides for the first time, to our knowledge, facts and figures of paediatric palliative care (PPC) in Belgium and a profile of children followed up by paediatric liaison teams. Around 720 children aged 0-18 years are followed up on an annual basis in Belgium (1). Between 2010 and 2014, however, we identified more than 22,500 children in hospital with a chronic complex condition, from which only 1.7% were referred to a paediatric liaison team, suggesting insufficient access to PPC services (2).

Our systematic review of outcome measurements in PPC showed that, although QoL is the main goal of PPC, robust instruments to assess QoL in children with life-limiting conditions are lacking (3). This is an appalling fact, meaning that quality of life in this particular setting cannot yet be assessed.

Based on the original APCA c-POS (4), and after its cross-cultural validation for a French version, a pilot-test we conducted in the Brussels region assessed the face/content validity of the further developed CPOS-2, its acceptability among 14 children and 19 parents, and its feasibility among 9 paediatric liaison team members. We combined an individual quality of life interview guide, the scheduled evaluation of individual quality of life (SEIQoL-DW), (5) and a questionnaire assessing parents' quality of life (QOLLTI-F) (6) to verify and complement the dimensions of the items included in the original 12-item CPOS. At the end of the pilot-test, an adapted version of the CPOS (CPOS-2) with 22 items to evaluate children's and parents' QoL was produced (7).

A subsequent national field test, involving all six paediatric liaison teams in Belgium, included 73 families, focusing on children's and parents' QoL in a PPC context. After item analysis, two items were deleted from the original CPOS (4) resulting in a 20-item CPOS-2, which showed a satisfactory internal consistency (0.76). A standard error of measurement (SEM) of 7.74 % was found, however, which means that an individual total CPOS-2 score should be interpreted with caution. Furthermore, a good convergent validity of the CPOS-2 with the KINDL (7), and a divergent validity with the QOLLTI-F scores (6) was found, demonstrating that the same construct - QoL - is assessed by the CPOS-2.

An overview of children and parents followed-up by paediatric liaison teams showed similar perspectives overall between children and parents, except for psycho-social dimensions. The implementation process of the CPOS-2 among paediatric liaison teams showed that the use of an outcome measurement instrument may deepen the relationship and trust between teams and families, helping them to address sensitive issues.

The good quality of collaboration with paediatric liaison teams was probably influenced by the professional background of the researcher (MF), who had previously worked as a paediatric nurse in one of the teams involved in this research. Paediatric liaison teams, who collaborated actively in this research, perceived the researcher as a legitimate individual to conduct the study. This might have been an essential component to fuel the collaboration. In qualitative studies, it is common to report the attitude adopted by the researcher in a transparent way in order to expose all potential influences on data interpretation. Professional backgrounds, but also the type of relations between researchers and participants, should be described, acknowledged and not labelled as bias (8-11).

The quadruple aim that should be achieved by health systems (12) suggests focusing on the QoL of health care professionals in addition to the QoL of patients, the quality of care and the cost-effectiveness of health interventions. This approach is consistent with the new definition of palliative care which, since 2019, aims to improve not only the QoL of patients but also that of their carers:

"Palliative care is the active holistic care of individuals across all ages with serious health-related suffering due to severe illness, and especially of those near the end of life. It aims to improve the quality of life of patients, their families and their caregivers." (13)

In line with the quadruple aim, at each step of our research we strived to choose methods that would respect the QoL not only of the child and his parents, but also the QoL of teams. For instance, by letting them decide where and by whom the interviews would be conducted, and by selecting instruments which would be the least burdensome (e.g. length of questionnaires, number of questionnaires). All focus groups and meetings with paediatric liaison teams were conducted in situ, at their workplace, in order to facilitate their attendance.

We would like to discuss now some specific themes which appeared relevant in our study.

Social stigma on the term "palliative care" as a barrier for its early integration

During the implementation process, we found that the term "palliative" in the name of an outcome measure instrument may be problematic for some families for whom disclosure of the children's palliative stage hasn't occurred yet. Using those instruments may also be difficult among teams who do not define themselves as a palliative care team, but prefer other names such as liaison care team or supportive teams. Instruments which assess palliative care outcomes might be easier to implement if a palliative culture already pre-exists among health care workers.

A misunderstanding of the purpose of palliative care (PC) persists. "The original message that palliative care is focused on total care, helping to live until the person dies, is being replaced and linked to feelings of fear, anxiety and death, instead of compassion, support or appropriate care. Society is still afraid to speak its name, and specialized units are identified as "places of death" as opposed to "places of life" meant to treat suffering. This issue is prohibitive to the implementation and development of PC policies worldwide. It is imperative to identify what message PC professionals are relaying to patients and other health care specialists and how that message may condition understandings of the right to access PC." (14)

Barriers to the integration and acceptance of paediatric palliative care have been studied among patients and families, with findings showing that more reluctance is found within health care professions than in families (15,16). The strong stigma attached to palliative care has been reported, and education of the public, patients and health care providers is paramount if early integration of palliative care is to be successful (17). This is also the position of Friedrichsdorf et al. (18), who advocate for explaining clearly and honestly what palliative care is. On the other hand, some authors have argued that palliative care should remain invisible and completely integrated into existing care, in order to be better accepted by families (19). The term liaison care, which is the term chosen in Belgium for designated paediatric palliative care teams and which focuses on the co-ordination and continuity of care, might improve acceptance of those teams.

In addition to the social stigma associated with the term palliative care, the identification of children with PPC needs remains a hindrance to its early integration. In our study, we found that only 1.7% of children diagnosed with a chronic complex condition had access to a paediatric liaison team (2). The paediatric palliative care screening tool (PaPas) is a practical screening tool that signposts PPC needs within the clinical setting (20,21). This facilitates early referrals to PPC without having to specify individual prognoses that are often uncertain. Other benefits include optimised continuity of care and implications for resource allocation. (22-26). The psychometric properties of this instrument have, however, not yet been assessed or reported.

Families with linguistic barriers and non-verbal children - a call to be more inclusive

In our field-study, many families could not be included because of language barriers. Those families are increasingly followed-up by paediatric liaison teams. For some families who do not clearly understand the local language, therefore, it would be useful to find a translator or a cultural mediator or make the CPOS available in several languages commonly used in that population, to improve the recruitment of families with language barriers and to address equity in the research process.

Emerging methods in PPC research include mixed-methods and patient-centred outcomes research, with specific attention given to children living with severe neurological conditions, unable to speak and /or whose parents encounter linguistic barriers (27,28). Ribbers has suggested meaningful outcomes for non-verbal children, based mostly on parental experiences/perspectives (29). These meaningful outcomes confirm the items included in the CPOS-2 and concern symptom control, respite and support, and the concepts of normalcy, security, empowerment and coping with the disease. A distinction should be made between parental experiences of having a child with a neurological disease versus other diseases, as often the care pathways are longer than those for children with cancer, with no diagnosis in the case of a rare disease or poor prognosis from the outset. Sourkes has suggested using drawings, pictograms and electronic devices to help non-verbal children express their feelings and report their outcomes (30).

Addressing hope while discussing advance care planning with children, adolescents and their parents

Discussions on death and dying with children and dealing with parental hopes emerged as important themes during the pilot and field-study.

Discussing end-of life issues with children, adolescents and their parents is complex. A subtle balance is needed between listening to their hopes while also exploring their wishes regarding treatment, care plans, preferred place to be cared for and also, in some circumstances, discussing the place of death (31).

Paediatric advance care planning seeks to ensure end-of-life care conforming to the patients’/their families' preferences. Being followed-up by a paediatric palliative care team leads to an integrated advanced care plan (ACP) and predicts the place of death according to medical orders on treatment limitations (32). This is in line with another study, in which children benefitting from PPC were more likely to have had an ACP addressed before death. Patients with PPC were more likely to have their goal of care (GOC) documented, completion of physician order for life-sustaining treatment (POLST), do-not-resuscitate code status, and hospice involvement at the time of death, compared to those who did not receive PPC. (33)

Studies that explored the role of advance care planning/discussing end of life (EOL) issues in PPC (32,34) showed that differences are found between adolescents' and parents' perspectives. The possibility of discussing those issues should be provided by PPC teams, acknowledging at the same time the different perceptions, feelings and perspectives of children and parents, according to the Trieste Charter (35,36). Decision-making tools which involve children and adolescents are progressively documented. In Belgium, the 5 wishes tool (37,38) was recently translated and adapted for a French version at the Hôpital universitaire des Enfants Reine Fabiola.

The role of hope, spirituality and resilience has been recently studied in the field of paediatric palliative care (31,39-46). It appears that hope influences the shared decision-making process and advance care planning. Acknowledgement of the role of hope by PLTs may help parents to shift the goal from a cure to the hope for better symptom control and improved quality of life.

In our study, we found that parental hope was not always focused on a treatment aimed at a cure, rather that it was also related to being with their child and to relieving his pain. For a long time, parental hope was wrongly perceived by professionals as a sign of denying children's fatal prognosis. In contrast, we found that hope can co-exist with parental awareness about their child's life-limiting condition. This is consistent with a study which showed that parents hoped for better QoL and symptom control (43,45,47).

Evaluating the quality of care or the quality of life?

The main goal of paediatric palliative care is to improve quality of life for both the child and the family (48,49). Quality of life is, however, a very subjective and complex construct, defined by the WHO as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns." (50)

Knowing the fundamental subjective essence of this concept, is it possible to evaluate QoL?

Outcome measurement research has been considered as a research priority in the field of paediatric palliative care in order to improve quality of care offered to children and to foster evidence of the effectiveness of paediatric palliative care (4,51,52). In recent years, patient-centred outcome measures (PCOM) encompassing both patient-reported and proxy-reported measures have been widely developed. An outcome measurement instrument evaluates 'change in health status' as a consequence of health care or interventions (53).

The term health status should be understood in a broad context of well-being or quality of life. If we refer to Donabedian's model for evaluating the quality of care, we must look at the structure, the process and the outcomes of a care program.

High quality paediatric palliative care is presumed to improve quality of life. Yet, an unknown gap may be found between the quality of care and the quality of life. Those two concepts can not be overlapped.

Initially, the designers of the CPOS intended to evaluate the outcomes of paediatric palliative care, in other words, the quality of care provided. In our pilot-study (chapter 6), by comparing the items of the CPOS and a tool assessing individual QoL, we found that some dimensions included in the original 12-item CPOS reflected or impacted the QoL. Furthermore, in our field-study (chapter 7-9), in comparing the adapted 22- item CPOS-2 with other standardized QoL instruments we found that the CPOS-2 effectively measured the same construct, namely the QoL.

Quality of life is a multidimensional construct and is not solely influenced by the burden of disease or the quality of care. "It is important to recognise that experience of care is not the same as outcomes of care. Experiences are likely to be better if outcomes are better, but they relate more closely to how individuals are respected, listened to and heard." (54).

Results from the studies presented below show that a certain type of care certainly influences the perceived QoL.

The Paediatric Quality of Life Evaluation of Symptoms Technology (PediQUEST)-study in the USA collected, via a computer-based system, patient-reported outcomes among 102 children with advanced cancer over a 9-month period. Patient-reported outcomes were mainly based on distressing symptoms, as assessed by the Pediquest-Memorial Symptom Assessment Scale, including the PedsQL 4.0. The main results of this study indicate that high-intensity therapies administered in the last month of life led to lower quality of life scores in children, compared to those who received mild therapies at end of life (55). This is consistent with a recent systematic review looking at the factors that may reduce QoL at end of life. Authors found that QoL of children at the end of life may be reduced by therapy with a curative intent (56).

Furthermore, an evidence base of mainly low- and moderate-quality studies (n= 42) showed that accessing specialist paediatric palliative care was associated with less intensive care at the end of life, more advance care planning and fewer in-hospital deaths (57).

We can conclude that quality of paediatric palliative care is the responsibility of paediatric liaison teams, and excellent symptom control, sensitive communication skills and effective coordination of care 24/7 an integral part of it. Improvements must be made to focus on respite care needs and siblings needs. We found that the quality of presence and the coordination of care ensured by paediatric liaison teams, whether it be through telephone calls, home visits or during ambulatory consultations, was deeply valued by families (1,7). Knowing that there is someone who cares was perceived as a relief. This is in line with a recent study conducted among family caregivers of children with medical complexity, which found that the experience of specific care coordination activities was associated with higher caregiver mental health-related quality of life (58).

Our findings suggest that some dimensions, not directly related to the disease, and rarely evaluated by usual QoL questionnaires, may influence children's and parental quality of life: the quality of relations among family members, the financial situation of parents, the informal social support around the family and the family's spiritual and/or religious resources. All these dimensions seem to contribute to the "spice of life".

Interestingly, in our own study, the CPOS and QOLLTI- scores of parents and children do not seem to be linked to the degree of disability nor the type of pathology the children lived with (chapter 9).

What is the CPOS-2 really measuring: QoL or the burden of disease on QoL?

What does the CPOS-2 measure? Is it the burden of a disease or is it rather the reflection of quality of life? Looking precisely at the 20 items of the CPOS-2, we found a balanced amount of reflective (reflecting quality of life) and formative (impacting quality of life) items.

Furthermore, during the interviews, and in addition to items included in the CPOS-2, we observed that children and parents addressed issues that were meaningful for them in that particular moment, suggesting that the CPOS-2 may be used as a roadmap, or a prompt list, to help clinical teams to address sensitive issues and individualize care plans.

The correlation studies showed that the self-report children QoL scores were close to the parents' proxy-reports, except for the emotional dimension. Therefore, self-report from a child should be encouraged among children who can express themselves. Furthermore, our results suggest that children's and parental QoL are interconnected, influencing each other closely. This was confirmed by a meta-analysis demonstrating that a statistically significant association was found between overall parent and child distress ($r = .32$, $p < .001$), such that increased parent-reported distress was associated with increased distress in their children (59). Measuring quality of life in a family should be viewed as measuring a complex system in which many subtle interactions may not be fully captured by an instrument.

We would like to recall one moving moment we experienced during the pilot-test to illustrate the complex construct of quality of life, which cannot be restricted to physical impairment (7). A 9-year old girl, seriously impaired by an advanced neurodegenerative disease, which had progressively paralyzed her four limbs leading to a full dependency on external support for her basic needs:

“You should ask if I’m happy!” (referring to the items of the CPOS-2).

Researcher replied: “ok, I”ll ask you: are you happy?”

She said very weakly: “Yes.”

“Ok, and would you tell me what makes you happy?”

Her, with a large smile on her face: “Because I always have my parents at my side.”

During another interview, a father emphasized how much being loved would influence, according to him, the quality of life of his young boy dying from a rare genetic disease progressively deteriorating all his abilities (walk, vision, mental capacity). This father added that being loved is a basic need, as important as food and, in his child’s case, even more important than food. He also stressed the importance of parents supporting each other as a couple in order to withstand the frightening situation he experienced with his dying child.

These two situations highlight the fundamental role of being supported, cared for and loved, and how it may significantly influence the QoL as perceived by children and parents. The burden of a disease is obviously also impacting daily life, but it seems that is not the most influential factor on the perceived feeling of well-being.

Other studies have demonstrated how much the quality of relations within the family and between the family and the clinical teams, as well as a family’s social network, may play an important role in a family’s QoL (26,60-63).

It may be surprising, but some family caregivers even reported having found a positive meaning in caring for their child facing life-limited conditions (64).

Those findings are aligned with the theory on the sense of coherence developed by Antonovsky (65), which consists of three dimensions: comprehensibility, meaningfulness and manageability. Those dimensions orientate the definition of quality of life privileging what adds value to life instead of focusing on the deterioration of physical functions or the burden of a disease. In fact, according to this theory, quality of life seems not to be impacted purely by the severity of a disease, but also by the comprehensibility, the meaningfulness and the manageability children and their parents are experiencing. Paediatric palliative care teams can support children and parents to better understand information on symptoms and treatment options. Giving meaning to a life-limiting condition is certainly not the role of teams, but they can nevertheless help families to deal with difficult emotions and experiences by listening and acknowledging their feelings. This is in line with some items included in the CPOS assessing worries, social support, information received and parents’ perceived self-efficacy.

Benefits of using outcome measurement instruments, as perceived by clinical teams

Results obtained from the pilot and field-study showed that PLTs, beyond the metrics, valued using the CPOS-2 to identify unmet needs (e.g. sleeping problems in children, respite care needs of parents or worries among siblings). Overall, the CPOS was perceived by PLTs as a clinical guidance tool to deepen their relationship with families, resulting in reinforced intimacy and trust, which was considered an opportunity to address sensitive issues such as advance care planning or the child's end-of-life.

PLTs were surprised to discover that using an outcome measurement instrument during an interview with a family could represent mutual benefits. Interviews were seen for teams as an opportunity to learn from the families and build a "toolkit of experiences" transferable to the accompaniment of other families. On the other hand, parents expressed their feeling of being cared for during the interview, by having the opportunity to share their resources and their difficulties.

Conducting a study on palliative outcome measurement instruments was an opportunity to improve the awareness of clinicians for palliative care if they had been involved in the study since the outset. At the early stage of the MOSAIK study, we identified, through discussions with PLTs, which outcomes were the most important to them. We found that symptom control and family well-being were the most important ones cited. When we discussed with each of the teams how they assessed this, most caregivers reported an intuitive approach based on their "gut feeling" and without using any formal scale. Having made this discovery, teams were highly motivated to discover outcome measures in order to objectively inform their judgment. Organizing meetings with whole interdisciplinary teams (especially clinicians, physicians, paediatricians) and explaining the objectives of the study may help them to be involved as an integral part of the research process. A bottom-up and collaborative approach throughout the study between researchers and clinical teams is paramount to enhance the implementation process of outcome measurement instruments.

Strengths and Limitations

To our knowledge, our research was the first to provide data on paediatric palliative care activities in Belgium and the profile of children followed-up by those teams.

It documented an original model of care suitable in paediatric palliative care, namely the Belgian liaison care model ensuring continuity of care for children across all care health settings, in a seamless way from curative to palliative stages and coordinated mainly by paediatric nurses.

The research methods, characterized by a triangulation design, took into account multiple perspectives, which enriched the interpretation of data. Methods chosen helped to describe in the most global way the complex interconnected facets of quality of life as perceived by children with life-limiting conditions, their parents and paediatric liaison teams.

The collaborative approach with paediatric liaison teams throughout the different studies was presumably an important element which may lead to a smoother implementation of the CPOS-2 in the future. The collaborative process with all PLTs in each region of Belgium to one common research enhanced professional identity and awareness, opening the way to other future projects to develop paediatric palliative care in Belgium.

Nevertheless, the small sample of families found in our studies, do not permit the generalizibility of our results. A high number of children with no verbal capacity are currently followed-up by paediatric liaison teams and could not self-respond to the questions of the CPOS-2. Furthermore, an increasing number of families which didn't speak Dutch or French could likewise not be included in our research.

We were aware that choosing the list of complex chronic conditions (as elaborated by Feudtner) would not completely overlap the population of children facing life-limiting conditions, but we were limited to using this list, as it was the only one compatible with the ICD-9 used by Belgian hospital registration systems in use between 2010-2014, our study period.

When starting the research, we wanted to also include the perspectives of siblings and their experiences of their quality of life, as they are often overlooked. For methodological reasons and the already complex study design of our research, we ultimately couldn't investigate this important domain.

Future perspectives, a roadmap for further research.

On-going collaborations

A growing interest in the MOSAIK study since 2018 from paediatric palliative care teams in France led, in August 2019, to a signed co-operation agreement between UCLouvain and the university hospital of Toulouse, notwithstanding the complete lack of funding. After passing all ethical and administrative approvals in France and following exactly the same protocol as the MOSAIK study in Belgium, data collection among eight French PPC teams started in February 2020 and will continue until December 2020. An estimated sample size of n=80 is expected, which will further document the psychometric properties of the CPOS-2. With this extended sample of 150 families, factor analysis will be carried out to better understand the validity of the CPOS-2 and to facilitate its fine-tuning.

Fruitful informal collaborations on methods of using CPOS have been created with the Cicely Saunders Institute (UK, ERC research grant) and researchers who conceived the initial APCA C-POS. A common paper is in progress, focusing on recruitment strategies and methods in countries that used the CPOS (UK, Turkey, Jordan, Kenya, Ouganda, South Africa, Namibia and… Belgium).

The Belgian paediatric palliative care group was created in 2015, under the initiative of Dr Marleen Renard and Ilse Ruysseveldt from the Katholieke Universiteit Leuven (KUL). It consists of around 30 members, from various interdisciplinary fields, who worked on the redaction of PPC guidelines. A website was created in 2018 and the guidelines focusing on symptom control, organization of care, ethics and advance care planning will be published in 2020 (66). MF coordinated one working group focusing on the organization of PPC in Belgium from 2017-2019. A strengthening of the BPPC collaboration with the Cancer Plan is foreseen.

European projects on patient centred outcomes research

A perspective for fostering better implementation of the CPOS-2 would be to make it available on a digital device app in order to enable children and adolescents to self-report on their quality of life from home.

A similar European project is on-going for paediatric cancer patients throughout five European countries: "MyPal aims to foster early palliative care for cancer patients by leveraging patient reported outcome (PRO) systems through their adaptation to the personal needs of the cancer patient and his/her caregiver(s). MyPal will exploit technological advances in digital health to support patients, family members and healthcare providers in gaining value through this systematic and comprehensive PRO-based intervention. MyPal will demonstrate and validate the proposed intervention in two clinical studies, a randomized controlled trial for adults with hematologic cancers and an observational study for children suffering from solid tumors and hematologic malignancies, hence targeting different age groups and cancer types." (67)

What further needs in the field of paediatric palliative care practice and research will emerge over the coming decades?

A recent publication from an expert review panel identified five common challenges in paediatric palliative care research: "patient diversity and small population size; interdependencies and dynamic interactions between child, family members, and disease processes over time; outcomes and measurement; workforce and infrastructure limitations; and presumed burden of PPC research on participants. The panel suggested the following themes be labelled as priorities: bolstering training and development of PPC investigators; developing core resources; advance symptom measurement (and measurements of other exposures and outcomes); improving symptom management and quality of life interventions; improving communication, elicitating care goals, and decision making; understanding family impact and facilitating or improving family adaptation and coping; and analyzing and improving systems of care, policy, and education." (68)

We would like to suggest some recommendations, based on the findings of the MOSAIK study and on the above-mentioned paper and discussed with paediatric liaison teams during our research meetings.

Implications for clinical practice, education, research and policy makers

Implications for clinical practice

As we found that the term palliative care is still misunderstood, paediatric palliative care teams should improve their communications strategies to inform what services they offer. This can take place, for example, through the creation of booklets, videos, symposia or stories. There is a need for a strengthening of the awareness of siblings' needs, who still remain overlooked.[11] More systematic preventive psychological consultations with siblings should be offered. Other creative spaces for siblings to express feelings and experiences in a recreational way would be beneficial. Parental respite care needs should be explored during home visits and available resources in these areas provided. For families with language barriers, paediatric liaison teams need the help of a translator or cultural mediator. The translation of the CPOS-2 into Arabic and Turkish is encouraged, considering that families speaking those languages represent a large part of the families followed-up by paediatric liaison teams.

In clinical care, we recommend promoting the use of materials (drawings, pictograms, electronic devices…) which allow caregivers to reach children's perceptions and feelings, even if they are non-verbal or facing cognitive impairment. The implementation of tools (booklets, videos,…) to address death and dying with children, to involve adolescents in shared-decision making processes and to implement advance care planning in an age-appropriate way should be promoted.

Regarding the organization of care, further implementation of the CPOS-2 on a regular basis in clinical care is needed in order to systematically document children's quality of life and monitor pain symptom management, potential psychosocial issues and other unmet needs, whether in hospital, respite care facilities, at home or in residences for disabled children. Reporting those measures in the usual data reporting systems is paramount to facilitate the long-term evaluation of the impact of paediatric palliative care. The creation of a electronic device (app) for the CPOS-2 might attract adolescents and facilitate its further implementation among PLTs. A harmonized registration and reporting system on patients' characteristics and clinical activities of all paediatric liaison teams should be fostered, which would help to document and evaluate the provision of services and staff needs. The working group with Cancer Plan focusing on this issue should be strengthened in the future.

---

[11] A co-authored published book chapter details how to accompany bereaved siblings. See Appendix (Articles and book chapters).

Classification of clinical activities carried out in French PPC teams, the Equipes ressources francophones de soins palliatifs pédiatriques, could be drawn upon for inspiration (69).

The creation in Belgium of a patients' association assembling adolescents and young adults facing life-limiting conditions should be promoted in order to defend their voices and give them the opportunity to be heard and to discuss policies concerning palliative care and end-of-life care. A model can be found through the group Palliative Care Voices in the United Kingdom, founded by two young adults, Lucy Watts and Huyaam Samuels, both affected by life-limiting conditions. "Palliative Care Voices exists to elevate the voice of direct stakeholders (that of people with lived experience) to allow them to be heard and to be involved in palliative care advocacy on the global stage, as well as at a national, regional and local level" (70).

Implications for education

Basic training on palliative care should be included in undergraduate health care curricula of medical faculties and nursing schools, as recommended by the Royal Belgian Academy of Medicine (71) and the resolution of the European Council on Palliative Care in Europe in 2018 (72). This education should be based on the 12 core competencies that have been recommended by the Children's Palliative Care Education taskforce of the European Association of Palliative Care (73):

1.Demonstrate and apply the core aspects of palliative care in the setting where children and families are based.

2.Demonstrate the range of knowledge on infants', children's and adolescents' development and family functioning, and how these are affected by a life-threatening illness.

3. Enhance physical comfort throughout the child's disease trajectories including end-of-life care.

4.Identify and respond to the child's psychosocial, educational and spiritual needs in palliative care.

5.Assess and respond to the needs of family carers.

6.Respond to the challenges of clinical and ethical decision-making in children's palliative care.

7.Facilitate communication and decision-making during crisis and end-of-life care.

8.Demonstrate capacity for interdisciplinary teamwork and inter-professional collaboration.

9.Develop interpersonal and communication skills appropriate to children and adolescents, including demonstrating capacity to break bad news and teaching parents how to provide care for the seriously ill child.

10.Assess the grief process, respond to the distinct needs of bereaved parents, siblings and significant others and provide appropriate support.

11.Practise reflective practice, self-awareness, and self-care.

12.Raise community awareness about palliative care for children and adolescents.

Based on those core competencies and since 2012, with the support of a dozen academics and experts in paediatric palliative care from different university hospitals, a 6-day interdisciplinary course in paediatric palliative care is offered at Haute Ecole Léonard de Vinci (74), as a continuous education program.

Implications for research

Studies should be conducted to better understand which type of profile needs specific paediatric palliative care interventions. Clinical tools for identification of patients needing paediatric palliative care and elaboration of clear referral criteria to paediatric liaison teams are encouraged.

The exploration, through qualitative studies, of anticipatory grief in children and their perspectives on death and dying and how they are influenced by parental expectations, feelings of loyalty and mutual trust, especially at end-of-life, should be fostered. Furthermore, the complex mechanisms around parental hope and their coping and resilience strategies when they care for their child with a life-limiting condition should be identified in order to develop practical guidance for interdisciplinary teams to enhance family resilience processes.

Best strategies should be identified for communications regarding paediatric palliative care, which is understood restrictively as end-of-life care, in order to overcome the associated social stigma that represents a barrier to access to palliative care services.

The impact of paediatric palliative care home services and respite care services on children's/parents' quality of life should be documented based on reliable instruments in prospective field-studies.

The impact of perinatal palliative care on long-term parental grief should be explored through mixed-methods prospective studies.

Robust Health care cost-analysis among paediatric palliative care populations should be promoted to foster a better understanding of how paediatric palliative care services may reduce emergency visits, hospital stays and deaths in intensive care units.

A mixed clinical-research team should be created to better advocate for patient involvement in the field of paediatric palliative care research (such as the Martin House research centre in the UK) and to promote implementation strategies based, for example, on the Promoting Action on Research Implementation in the Health Services-framework (PARiHS). This framework fosters the relationship between evidence, context and facilitation, to achieve knowledge translation of paediatric palliative care (75). A favourable breeding ground would be the Belgian paediatric palliative care group (66), which already brings together interdisciplinary professionals from all Belgian regions and has elaborated guidelines in paediatric palliative care for Belgium. The active representation of PPC experts within the three Belgian Palliative care Federations and the Federal Palliative Care Evaluation unit should be promoted and structurally ensured.

Implications for policy makers

Based on the first study analysing the annual reports of paediatric liaison teams and their time spent searching for funding, we advocate for better structural funding of paediatric liaison teams based on real data (number of children followed-up and on harmonized clinical activities indicators) instead of a flat-rate.

In order to achieve alignment with the Royal Decree defining the missions of paediatric liaison teams (76), we recommend providing sufficient long-term funding of all professionals within interdisciplinary paediatric liaison teams, including a medical coordinator, a psychologist and a social worker, as this is not yet the case for all paediatric liaison teams.

Funding opportunities to foster paediatric palliative care research need to be generated, as we observed challenges in finding these.

Consistent with a bottom-up approach, integration of PPC experts in the federal commission on the evaluation of palliative care and of euthanasia should be fostered to better take into account the reality of the paediatric palliative care field.

References

1.Friedel M, Brichard B, Fonteyne C, Renard M., Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014. BMC Palliat Care.2018;17(1):77. doi:10.1186/s12904-018-0324-2

2. Friedel M, Gilson A, Bouckenaere D, Brichard B, Fonteyne C, Wojcik T, De Clercq E, Guillet A, Mahboub A, Lahaye M, Aujoulat I. Access to paediatric palliative care in children and adolescents with complex chronic conditions: a retrospective hospital-based study in Brussels, Belgium. BMJ Paediatr Open. 2019;3(1):e000547. Published 2019 Sep 29. doi:10.1136/bmjpo-2019-000547.

3. Friedel M, Aujoulat I, Dubois AC, Degryse JM. Instruments to Measure Outcomes in Paediatric Palliative Care: A Systematic Review. Paediatrics. 2019;143(1):e20182379. doi:10.1542/peds.2018-2379.

4. Downing J, Namisango E, Harding R. Outcome measurement in paediatric palliative care: lessons from the past and future developments. Ann Palliat Med. 2018;7(Suppl 3):S151-S163. doi: 10.21037/apm.2018.04.02.

5. Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS. BMJ. 1996;313(7048):29-33. doi: 10.1136/bmj.313.7048.29.

6. Cohen R, Leis AM, Kuhl D, Charbonneau C, Ritvo P, Ashbury FD. QOLLTI-F: measuring family carer quality of life. Palliat Med. 2006;20(8):755-67. doi: 10.1177/0269216306072764.

7. Friedel Friedel M, Brichard B, Boonen S, Tonon C, de Terwangne B, Bellis D, Mevisse M, Fonteyne C, Jaspard M, Schruse M, Harding R, Downing J, Degryse JM, Aujoulat I. Face and Content Validity, Acceptability, and Feasibility of the Adapted Version of the Children's Palliative Outcome Scale: A Qualitative Pilot Study [published online ahead of print, 2020 Jul 15]. J Palliat Med. 2020;10.1089/jpm.2019.0646. doi:10.1089/jpm.2019.0646 http://hdl.handle.net/2078.1/231142

8. Erhart M, Ellert U, Kurth B, Ravens-Sieberer U, Measuring adolescents' HRQoL via self and parent proxy reports: an evaluation of the psychometric properties of both versions of the KINDL-R instrument. Health and Quality of Life Outcomes. 2009; 7:77.

9. De Lavergne C. la posture du praticien-chercheur: une analyse de l'évolution de la recherche qualitative. Recherches qualitatives. Hors Série 2007,3:28-43.

10. Hamisultane S. la nécessaire distanciation du chercheur par l'analyse de son implication. Interrogations 2014 (18).

11. Pereira I, Chaar N. Immanence, dévoilement et réflexivité. Faire de la sociologie à partir d'une implication militante sur le lieu professionnel. Interrogations 2014, n° 19.

12. Duclos M. Que la relation d'enquête soit aussi d'amitié. Interrogations 2014, n° 18.

13. Bodenheimer T, Sinsky C. From triple to quadruple aim: care of the patient requires care of the provider. Ann Fam Med. 2014;12(6):573-6. doi: 10.1370/afm.1713.

14. Radbruch L, De Lima L, Knaul F, et al. Redefining Palliative Care-A New Consensus-Based Definition. J Pain Symptom Manage. 2020;S0885-3924(20)30247-5. doi:10.1016/j.jpainsymman.2020.04.027.

15. Reigada C, Arantzamendi M, Centeno C. Palliative care in its own discourse: a focused ethnography of professional messaging in palliative care. BMC Palliat Care. 2020;19, 88. https://doi.org/10.1186/s12904-020-00582-5

16. Levine DR, Mandrell BN, Sykes A, et al. Patients' and Parents' Needs, Attitudes, and Perceptions About Early Palliative Care Integration in Paediatric Oncology. JAMA Oncol. 2017;3(9):1214-1220. doi:10.1001/jamaoncol.2017.0368.

17. Dalberg T, McNinch NL, Friebert S. Perceptions of barriers and facilitators to early integration of paediatric palliative care: A national survey of paediatric oncology providers. Pediatr Blood Cancer. 2018;65(6):e26996. doi:10.1002/pbc.26996.

18. Zimmermann C, Swami N, Krzyzanowska M, et al. Perceptions of palliative care among patients with advanced cancer and their caregivers. CMAJ. 2016;188(10):E217-E227. doi:10.1503/cmaj.151171.

19.Friedrichsdorf SJ, Bruera E. Delivering Paediatric Palliative Care: From Denial, Palliphobia, Pallilalia to Palliactive. Children (Basel).2018 ;5(9):120. doi: 10.3390/children5090120.

20. Merrow NM. Palliative care most effective when invisible. CMAJ. 2016;188(13):973. doi:10.1503/cmaj.1150118

21.Bergstraesser E, Hain RD, Pereira J. The development of an instrument that can identify children with palliative care needs: the Paediatric Palliative Screening Scale (PaPaS Scale): a qualitative study approach. BMC Palliative Care 2013; 12:20

22. Bergstraesser E, Paul M, Rufibach K, Hain RD, Held L. The Paediatric Palliative Screening Scale: Further validity testing. Palliat Med. 2014 Jun;28(6):530-533.

23. Chong PH, Soo J, Yeo ZZ, Ang RQ, Ting C. Who needs and continues to need paediatric palliative care? An evaluation of utility and feasibility of the Paediatric Palliative Screening scale (PaPaS). BMC Palliat Care. 2020;19(1):18. doi: 10.1186/s12904-020-0524-4.

24. Matraka N. Critères d’identification des enfants nécessitant l’intervention de l’équipe de liaison en soins palliatifs pédiatriques selon les médecins des Cliniques Universitaires Saint-Luc : analyse qualitative et test d’un outil d’aide à la décision : la Paediatric Palliative Screening Scale. Faculté de santé publique, Université catholique de Louvain, 2018. Prom. : Aujoulat, Isabelle ; Friedel, Marie.

25. Jankovic M, De Zen L, Pellegatta F, Lazzarin P, Bertolotti M, Manfredini L, Aprea A, Memo L, Del Vecchio A, Agostiniani R, Benini F. A consensus conference report on defining the eligibility criteria for paediatric palliative care in Italy. Italian Journal of Paediatrics. 2019;45:89. doi.org/10.1186/s13052-019-0681-3.

26. Kaye E, Rubenstein J, Levine D, et al. Paediatric palliative care in the community. CA Cancer J Clin. 2015;65(4):316-33.

27. Harrop E, Edwards C. How and when to refer a child for specialist paediatric palliative care. Arch Dis Child Educ Pract Ed. 2013;0:1–7. doi:10.1136/archdischild-2012-303325.

28. Jarvis S, Fraser LK. Comparing routine inpatient data and death records as a means of identifying children and young people with life-limiting conditions. Palliat Med. 2018;32(2):543-553. doi: 10.1177/0269216317728432.

29. Nelson KE, Feinstein JA, Gerhardt CA, Rosenberg AR, Widger K, Faerber JA, Feudtner C. Emerging Methodologies in Paediatric Palliative Care Research: Six Case Studies. Children (Basel). 2018;5(3):32. doi: 10.3390/children5030032.

30. Ribbers S, Wager J, Hartenstein-Pinter A, Zernikow B, Reuther M. Core outcome domains of paediatric palliative care for children with severe neurological impairment and their families: A qualitative interview study. Palliat Med. 2020;34(3):309-318. doi:10.1177/0269216319885818.

31. Sourkes BM. Children's Experience of Symptoms: Narratives through Words and Images. Children (Basel). 2018;5(4):53. doi:10.3390/children5040053.

32. Jalmsell L, Lövgren M, Kreicbergs U, Henter JI, Frost BM. Children with cancer share their views: tell the truth but leave room for hope. Acta Paediatr. 2016;105(9):1094-1099. doi:10.1111/apa.13496.

33. Hoell JI, Weber HL, Balzer S, Danneberg M, Gagnon G, Trocan L, Borkhardt A, Janßen G, Kuhlen M. Advance care planning and outcome in paediatric palliative home care. Oncotarget. 2018;9(25):17867-17875. doi: 10.18632/oncotarget.24929.

34. Harmoney K, Mobley EM, Gilbertson-White S, Brogden NK, Benson RJ. Differences in Advance Care Planning and Circumstances of Death for Paediatric Patients Who Do and Do Not Receive Palliative Care Consults: A Single-Center Retrospective Review of All Paediatric Deaths from 2012 to 2016. J Palliat Med. 2019;22(12):1506-1514. doi:10.1089/jpm.2019.0111.

35. Friebert S, Grossoehme DH, Baker JN, et al. Congruence Gaps Between Adolescents With Cancer and Their Families Regarding Values, Goals, and Beliefs About End-of-Life Care. JAMA Netw Open. 2020;3(5):e205424. Published 2020 May 1. doi:10.1001/jamanetworkopen.2020.5424.

36. Benini F, Vecchi R, Lazzarin P, Jankovic M, Orsi L, Manfredini L, Drigo P, Sellaroli V, Gangemi M, Spizzichino M, Orzalesi M. The rights of the dying child and the duties of healthcare providers: the "Trieste Charter". Tumori. 2017;103(1):33-39. doi: 10.5301/tj.5000566.

37.Benini F, Vecchi R, Orzalesi M. A charter for the rights of the dying child, The Lancet. 2014; 383 (9928).

38. Zadeh S, Pao M, Wiener L. Opening end-of-life discussions: how to introduce Voicing My CHOiCES™, an advance care planning guide for adolescents and young adults. Palliat Support Care. 2015;13(3):591-9.

39. Wiener L, Zadeh S, Battles H, Baird K, Ballard E (2012) Allowing adolescents and young adults to plan their end-of-life care. Paediatrics ; 130(5):897-905.

40. Lotz JD, Daxer M, Jox RJ, Borasio GD, Führer M. "Hope for the best, prepare for the worst": A qualitative interview study on parents' needs and fears in paediatric advance care planning. Palliat Med. 2017;31(8):764-771. doi: 10.1177/0269216316679913.

41. Rosenberg AR, Bradford MC, Bona K, Shaffer ML, Wolfe J, Baker KS, Lau N, Yi-Frazier J. Hope, distress, and later quality of life among adolescent and young adults with cancer. J Psychosoc Oncol. 2018;36(2):137-144. doi: 10.1080/07347332.2017.1382646.

42. Sisk BA, Kang TI, Mack JW. Sources of parental hope in paediatric oncology. Pediatr Blood Cancer. 2018;65(6):e26981. doi: 10.1002/pbc.26981.

43. Rosenberg AR, Bradford MC, Barton KS, Etsekson N, McCauley E, Curtis JR, Wolfe J, Baker KS, Yi-Frazier JP. Hope and benefit finding: Results from the PRISM randomized controlled trial. Pediatr Blood Cancer. 2019;66(1):e27485. doi: 10.1002/pbc.27485.

44. Feudtner C, Carroll KW, Hexem KR, Silberman J, Kang TI, Kazak AE. Parental hopeful patterns of thinking, emotions, and paediatric palliative care decision making: a prospective cohort study. Arch Pediatr Adolesc Med. 2010;164(9):831-839. doi:10.1001/archpaediatrics.2010.146

45. Barton KS, Tate T, Lau N, Taliesin KB, Waldman ED, Rosenberg AR. "I'm Not a Spiritual Person." How Hope Might Facilitate Conversations About Spirituality Among Teens and Young Adults With Cancer. J Pain Symptom Manage. 2018;55(6):1599-1608. doi: 10.1016/j.jpainsymman.2018.02.001.

46. Hill DL, Nathanson PG, Carroll KW, Schall TE, Miller VA, Feudtner C. Changes in Parental Hopes for Seriously Ill Children. Paediatrics. 2018;141(4):e20173549. doi: 10.1542/peds.2017-3549.

47. Superdock AK, Barfield RC, Brandon DH, Docherty SL. Exploring the vagueness of Religion & Spirituality in complex paediatric decision-making: a qualitative study. BMC Palliat Care. 2018;17(1):107. doi:10.1186/s12904-018-0360-y.

48. Chong PH, De Castro Molina JA, Teo K, Tan WS. Paediatric palliative care improves patient outcomes and reduces healthcare costs: evaluation of a home-based program. BMC Palliat Care. 2018;17(1):11. doi: 10.1186/s12904-017-0267-z.

49. Craig F, Abu-Saad Huijer H, Benini F, Kuttner L, Wood C, Feraris PC, Zernikow B. [IMPaCCT: standards of paediatric palliative care]. Schmerz. 2008;22(4):401-8. doi: 10.1007/s00482-008-0690-4.

50. World Health Organization (WHO). Definition of palliative care. 1998. https://www.who.int/cancer/palliative/definition/en/ (consulted 15th June 2020).

51. World Health Organization. Definition of quality of life. https://www.who.int/healthinfo/survey/whoqol-qualityoflife/en/ (consulted 15th July 2020)

52. Harding R, Chambers L, Bluebond-Langner M. Advancing the science of outcome measurement in paediatric palliative care. Int J Palliat Nurs. 2019;25(2):72-79. doi: 10.12968/ijpn.2019.25.2.72.

53. Baker JN, Levine DR, Hinds PS, Weaver MS, Cunningham MJ, Johnson L, Anghelescu D, Mandrell B, Gibson DV, Jones B, Wolfe J, Feudtner C, Friebert S, Carter B, Kane JR. Research Priorities in Paediatric Palliative Care. J Pediatr. 2015;167(2):467-70.e3. doi: 10.1016/j.jpeds.2015.05.002.

54. Donabedian A. Explorations in Quality Assessment and Monitoring: The Definition of Quality and Approaches to Its Assessment. Volume I. Ann Arbor, MI: Health Administration Press; 1980.

55. Witt J, Murtagh FEM, de Wolf-Linder S, Higginson IJ, Daveson BA. Introducing the Outcome Assessment and Complexity Collaborative (OACC) Suite of Measures. 2015 https://www.kcl.ac.uk/cicelysaunders/attachments/Studies-OACC-Brief-Introduction-Booklet.pdf (consulted 13th July 2020).

56. Wolfe J, Orellana L, Ullrich C, et al. Symptoms and Distress in Children With Advanced Cancer: Prospective Patient-Reported Outcomes From the PediQUEST Study. J Clin Oncol. 2015;33(17):1928-1935. doi:10.1200/JCO.2014.59.1222.

57. Piette V, Beernaert K, Cohen J. et al. Healthcare interventions improving and reducing quality of life in children at the end of life: a systematic review. Pediatr Res 2020; https://doi.org/10.1038/s41390-020-1036-x

58. Taylor J, Booth A, Beresford B, Phillips B, Wright K, Fraser L. Specialist paediatric palliative care for children and young people with cancer: A mixed-methods systematic review. Palliat Med. 2020;34(6):731-775. doi:10.1177/0269216320908490

59. Yu JA, Henderson C, Cook S, Ray K. Family Caregivers of Children with Medical Complexity: Health-Related Quality of Life and Experiences of Care Coordination [published online ahead of print, 2020 Jun 26]. Acad Pediatr. 2020;S1876-2859(20)30258-8. doi:10.1016/j.acap.2020.06.014

60. Bakula DM, Sharkey CM, Perez MN, et al. Featured Article: The Relationship Between Parent and Child Distress in Paediatric Cancer: A Meta-Analysis. J Pediatr Psychol. 2019;44(10):1121-1136. doi:10.1093/jpepsy/jsz051

61. Mitchell S, Bennett K, Morris A, Slowther AM, Coad J, Dale J. Achieving beneficial outcomes for children with life-limiting and life-threatening conditions receiving palliative care and their families: A realist review. Palliat Med 2020;34(3):387-402. doi: 10.1177/0269216319870647.

62. Brimble MJ, Anstey S, Davies J. Long-term nurse-parent relationships in paediatric palliative care: a narrative literature review. Int J Palliat Nurs. 2019;25(11):542-550. doi:10.12968/ijpn.2019.25.11.542

63. Rogers SK, Gomez CF, Carpenter P, et al. Quality of life for children with life-limiting and life-threatening illnesses: description and evaluation of a regional, collaborative model for paediatric palliative care. Am J Hosp Palliat Care. 2011;28(3):161-170. doi:10.1177/1049909110380594.

64. Magliano L, Patalano M, et al. "I have got something positive out of this situation": psychological benefits of caregiving in relatives of young people with muscular dystrophy, J Neurol. 2014;261(1):188-95.

65. Antonovsky A, Sagy S. Aaron Antonovsky, the Scholar and the Man Behind Salutogenesis. 2016 Sep 3. In: Mittelmark MB, Sagy S, Eriksson M, Bauer GF, Pelikan JM, Lindström B, Espnes GA, editors. The Handbook of Salutogenesis [Internet]. Cham (CH): Springer; 2017. Chapter 3.

66. Belgian Paediatric Palliative Care group. http://bppc.be (consulted 15 Jun 2020)

67. Mypal European research project. https://cordis.europa.eu/project/id/825872 (consulted 15 Jun 2020)

68. Feudtner C, Rosenberg AR, Boss RD, et al. Challenges and Priorities for Paediatric Palliative Care Research in the U.S. and Similar Practice Settings: Report From a Paediatric Palliative Care Research Network Workshop. J Pain Symptom Manage. 2019;58(5):909-917.e3. doi:10.1016/j.jpainsymman.2019.08.011.

69. Bouffay C, Rollin A, Marcault A, Rémy C, Castaing M, Filion S, Pétillard E, Fournié F, Schell M. Indicators for clinical activity in palliative care: the experience of a regional resources team of paediatric palliative care. Médecine palliative-Soins de support-Accompagnement-Ethique. 2017;16, 161-168.

70. Palliative Care Voices. https://palliativecarevoices.org/about/ (consulted 15th July 2020)

71. Royal Belgian Academy of Medicine. Avis sur l'enseignement des soins palliatifs dans les facultés de médecine en Belgique. 2017 http://www.armb.be/index.php?eID=tx_nawsecuredl&u=0&g=0&hash=7627a33c936e4239e2cb0e438d6de9dc62347c8c&file=fileadmin/sites/armb/upload/armb_super_editor/armb_editor/pdf/Avis/2017/enseignement_des_soins_palliatifs.pdf (consulted 15 Jul 2020)

72. European Council. Resolution 2249 on palliative care provision in Europe. 2018 http://assembly.coe.int/nw/xml/XRef/Xref-DocDetails-FR.asp?FileID=25214&lang=FR (consulted 15 July 2020).

73. European Association of Palliative Care. Children's Palliative Care Education taskforce. Downing J, Ling J, Benini F, Payne S, Papadatou D. Core competencies for education in children's palliative care. 2014. https://www.icpcn.org/eapc-report-published-on-core-competencies-in-education-on-ppc/ (consulted 15 Jul 2020)

74. Haute Ecole Léonard de Vinci. Formation continue multidisciplinaire. Les soins palliatifs de la naissance à l'adolescence. http://www.parnasse-deuxalice.be/fr-be/Parnasse-Isei/Pages/FC/Les-soins-continus-et-palliatifs-de-la-naissance-a-l-adolescence.aspx (consulted 15th June 2020)

75. Nilsson S, Ohlen J, Hessman E, Brännström M. Paediatric palliative care: a systematic review. BMJ Support Palliat Care. 2020;10(2):157-163. doi:10.1136/bmjspcare-2019-001934

76. Arrêté Royal fixant les normes auxquelles la fonction "liaison pédiatrique" doit répondre pour être agréée. 2010. https://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html (consulted 15th July 2020).

CHAPTER 8. CONCLUSION

To conclude, we would like to recall our objectives :

a) first, to document the characteristics of children followed-up by paediatric liaison teams (PLT) in Belgium and their access to those services (Chapter 3);

b) second, to search for reliable instruments measuring outcomes, especially quality of life, which is the main goal of paediatric palliative care (Chapter 4);

c) third, to further develop an instrument able to evaluate children's quality of life and that of their parents by documenting its psychometric properties (Chapter 5 and Subchapter 6.1);

d) fourth, to report on the quality of life experienced by children followed-up by PLTs in Belgium (Subchapter 6.2);

e) and fifth, to understand how PLTs experienced using an outcome measurement instruments (Subchapter 6.3).

The study described in Subchapter 3.1, based on the PLT's annual reports, documented the number and profile of children followed from 2010 to 2014. In addition to the 720 children/year followed up, we were able to observe that an ever-increasing number of children require the support of the PLTs. Beside training, research and fund-raising activities, PLTs provide curative, palliative and bereavement care, demonstrating continuity of care over time and across the different care pathways. The fact that PLTs are focusing on liaison care makes it particularly difficult to make a clear distinction between the curative and palliative stages, resulting in heterogeneous data recording between teams.

A second study, reported in Subchapter 3.2, based on hospital registers, identified more than 22.500 children hospitalised for chronic complex condition in the Brussels region between 2010 and 2014. Access of these children to a PLT seems to be insufficient, given that only 1.7% of children hospitalised for chronic complex condition were referred to a PLT. However, not all complex chronic conditions are life-limiting or life-threatening illnesses and do not necessarily require the support of a PLT. This study was able to highlight, on the one hand, the challenges of establishing the prevalence of children facing a life-limiting or life-threatening illness and, on the other hand, the difficulty in defining the profile of children who should be followed-up by a PLT.

The systematic literature review presented in chapter 4 sought to identify instruments that evaluate the outcomes of paediatric palliative care. The results show that most of the instruments assess children's quality of life to little or no extent, that the psychometric properties of these tools are only very partially documented, and that the outcomes instrument measurements rarely give children the opportunity to self-report.

In chapter 5, we described the pilot testing of the CPOS, which assessed its face and content validity, its acceptability by children and their parents, and its feasibility of use for teams. The CPOS was further developed to include dimensions identified by children and parents as important for their quality of life. This process resulted in the further development of the CPOS-2 with 22 items. Although the initial CPOS was thought to evaluate the quality of care, we believe -as argued in chapter 6 - that this instrument is suitable for assessing the quality of life of children and their parents in a palliative care context.

To document other psychometric properties of CPOS-2, as reported in Subchapter 6.1, we conducted a field test with the collaboration of 6 Belgian PLTs, in 2 languages, consisting of interviews with 73 families followed-up by the PLTs. Comparing it with other quality of life instruments, we found that the 22-item CPOS-2 had a reliability index (internal consistency) of 0.76. Furthermore, we found that the CPOS-2 had a satisfactory concurrent validity. This allows us to conclude that the CPOS assesses the same construct, i.e. quality of life.

In the study documented in Subchapter 6.2, we also found that the quality of life scores as reported by children were slightly higher than those reported by their parents and comparable to scores documented in other studies. Surprisingly, we did not find a statistically significant correlation between QoL scores and children's functional deficits, type of disease, or age. Finally, we found that paediatricians' intuitive estimates when assessing the degree of suffering of children or parents appear to correlate with CPOS-2 quality of life scores. However, predicting child’s life expectancy was found impossible in most cases.

The results reported in subchapter 6.3 show that PLTs perceive the use of the CPOS-2 as a useful compass/roadmap for identifying hidden needs, becoming more involved with families, addressing sensitive issues and learning from parents.

Looking at those results of these different studies, we can ask ourselves the following questions:

How and why shall we measure quality of life in a palliative context?

Does the CPOS-2 measure quality of life?

What is quality of life in a paediatric palliative context?

We were able to document the challenges inherent in measuring quality of life. These relate to the difficulty of defining the construct of quality of life, let alone quality of life from a child's point of view, when faced with a life-limiting or life-threatening illness.

It seemed essential to us to start from what the children themselves defined as the spice of their lives in order to define the dimensions that should be included in an outcome measurement instrument. On the other hand, we wanted this instrument to integrate the perspectives of both the children and their parents, in order to incorporate their perception of each other, which can be divergent and yet complementary at the same time. Moreover, this instrument must be capable of assessing both the quality of life of the children and of their parents, bearing in mind that in paediatrics the parents' well-being influences that of their child and vice versa.

Measuring the quality of life in a palliative context makes it possible to gather the experiences of children and their parents by giving them the opportunity to express their difficulties, but also to share resources that they have identified as important to live to the fullest. As our results show, to discover such resources help the teams to enrich their "toolbox" to better accompany other families. Eventually, once the CPOS-2 is fine-tuned in terms of its psychometric properties, in its graphic design or support (electronic application), it could be used at regular intervals during an accompaniment. It therefore has the potential to become a useful self-evaluation instrument, to help the teams document and support the families' quality of life.

The CPOS-2 offers the advantage of evaluating in the same document children's and parents' QoL. It assesses QoL according to dimensions that have been identified as important by the children and their parents. However, we recognize that the sample in our study was small, especially the number of children with verbal capacity. It is therefore not, at this stage, representative of all children currently receiving palliative care.

Based on the observation that quality of life is not only a highly subjective concept from one child and parent to another, but also a concept influenced by factors that may vary over time, we believe that an instrument will never be able to fully assess quality of life.

For a quality of life instrument to be truly usable with children in a palliative context, it must combine many requirements in proportionate balance: it must be reliable AND valid by containing sufficient items reflecting the many dimensions of quality of life, but also short enough to be perceived as acceptable by the children and caregivers who will use it. Derived from our observations, we found that the CPOS-2 had those characteristics. It should be hetero-administered, because the answers given to the questions and the discussion raised through the questionnaire may be helpful to open a dialogue between PLTs and families and to individualize the care plan.

It emerges from our research that the social dimension, that of interpersonal relationships, is fundamental and seems to have a highly significant impact on the perception of QoL for both children and parents. Indeed, being surrounded and loved, having friends and celebrating and feeling supported and accompanied, seem to play a key role in the quality of life of families. In some situations, this was found to compensate for progressive and rapid evolution of the disease, which can lead to the loss of the child's physical and psychological functions.

Does CPOS-2 therefore measure what makes up the quality of life or the impact of the disease on this quality of life? Based on our results, it appears that the CPOS-2 mainly measures what makes up the spice of life, independently of the burden of a disease. In other words, measuring quality of life seems to be conceptually linked to social dimensions and is not reflecting the absence of the burden of a disease.

At the very beginning of the adventure of this thesis, a sentence resonated deep within me:

To care is to connect (« *ce qui fait soin, c'est ce qui fait lien* »).

Now, at the end of writing this thesis, this sentence comes back in force and reveals, as a watermark, the major discovery of this research: Relationships are the basis of quality of life, beyond handicaps and the degradation of physical health, whilst not denying their importance.

Therefore, the essence of the PLTs is to connect, to support the relationship, to weave the thread: Between the children and their parents. Between the unsaid and the expressible. Between the curative and the palliative stage. Between all the interveners. Between the hospital and the home, sometimes the school, the day centre. Between death and mourning, even if sometimes the mourning takes place before death. Between tears and smiles.

Aiming for quality of life in paediatric palliative care is fundamentally about connecting.

EPILOGUE

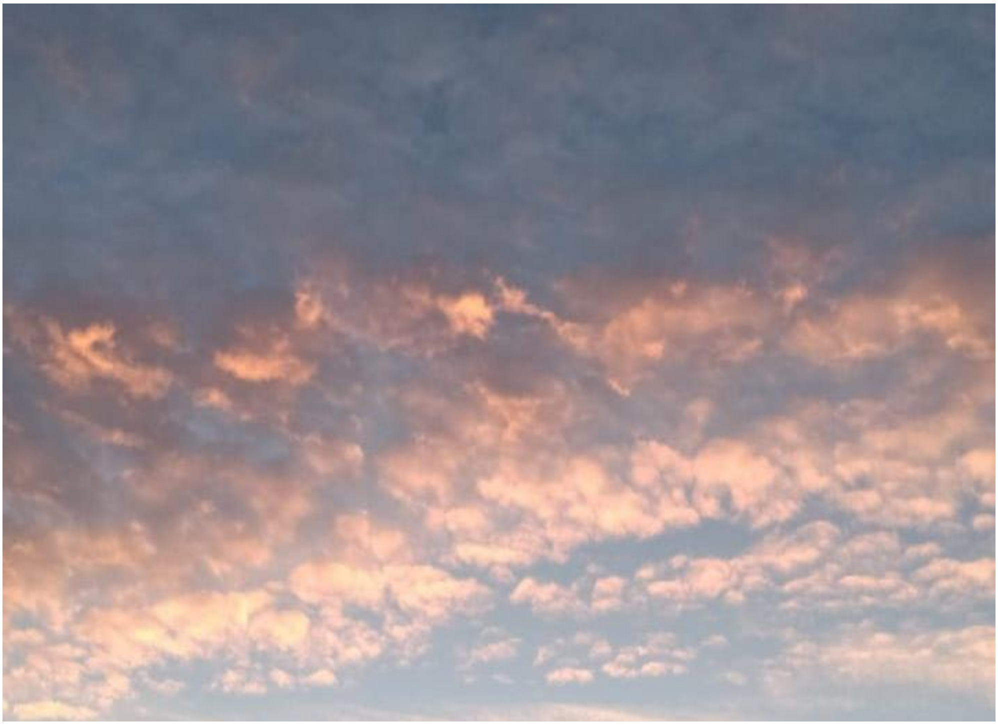
Once upon a time, there was an old man who used to go to the ocean to do his writing. He had a habit of walking on the beach every morning before he began his work. Early one morning, he was walking along the shore after a big storm had passed and found the vast beach littered with starfish as far as the eye could see, stretching in both directions. Off in the distance, the old man noticed a small boy approaching. As the boy walked, he paused every so often and, as he grew closer, the man could see that he occasionally bent down to pick up an object and throw it into the sea. The boy came closer still and the man called out, “Good morning! May I ask what it is that you are doing?” The young boy paused, looked up, and replied “Throwing starfish into the ocean. The tide has washed them up onto the beach and they can’t return to the sea by themselves,” the youth replied. “When the sun gets high, they will die, unless I throw them back into the water.” The old man replied, “But there must be tens of thousands of starfish on this beach. I’m afraid you won’t really be able to make much of a difference.” The boy bent down, picked up yet another starfish and threw it as far as he could into the ocean. Then he turned, smiled and said, “It made a difference to that one!”

adapted from The Star Thrower, by **Loren Eiseley** in The Unexpected Universe, 1969.

"As a practice, care requires more than good intentions. It requires an in-depth and thoughtful knowledge of the situation, of the situation of all the actors, as well as of their needs and skills. The implementation of ethics of care requires knowledge of the context of the care process. Those who engage in this process must make judgements: about needs, about conflicts between needs, about strategies to achieve the chosen ends, about the recipients' capacity to respond. »

Joan Tronto. Un monde vulnérable. Pour une politique du care. La Découverte, Paris, 2009.

Paediatric palliative care (PPC) is closely linked to what this young boy did with each starfish: at an individual level, trying to make a difference to one specific child, to one unique family. However, as stated by Tronto, caring is also a complex social responsibility. As researchers we must search for the best evidence in PPC; and as policymakers, opt for accessible, effective, high quality integrated PPC programs to prevent unnecessary total suffering for each child facing a life-limiting or a life-threatening condition, to live his life to the fullest.



Lockdown in Brussels, May 2020

Photo credits @ Marie Friedel

APPENDICES

Children palliative outcome scale version 2 (CPOS-2)

In French and in English

Code unique :

SECTION A: A PROPOS DE L'ENFANT indiquez 0,1,2,3,4, ou 5 ou « je ne sais pas » pour chaque question posée en pensant à ces derniers jours…

	Questions à poser à l'enfant/adolescent	**REPONSES POSSIBLES**	Score	**Questions à poser aux parents**	**REPONSES POSSIBLES**	Score
Q1.	Peux-tu me dire si tu as eu mal?	0 1 2 3 4 5 Pas mal du tout Très, très mal ☐ Je ne sais pas		Votre enfant a-t'il/elle eu mal?	0 1 2 3 4 5 Pas mal du tout Très, très mal ☐ Je ne sais pas	
Q2.	Est-ce qu'il y a des problèmes dans ton corps qui t'ont dérangé.e? *(suggérer si néc. : vomissements, diarrhée,* nausées, insomnie) ?	0 1 2 3 4 5 0 Aucun problème dans mon corps ne m'a dérangé 5 Des problèmes dans mon corps m'ont fortement dérangé ☐ Je ne sais pas		Est-ce qu'il y a des problèmes physiques qui ont dérangé votre enfant? *(suggérer si néc.:* vomissements, diarrhée, nausées, insomnie, etc.) ?	0 1 2 3 4 5 0 Aucun problème physique l'a dérangé 5 Des problèmes physiques l'ont fortement dérangé ☐ Je ne sais pas	
Q3.	Est-ce qu'il y a quelque chose qui t'a dérangé.e par rapport à ton alimentation ?	0 1 2 3 4 5 Rien du tout Oui, beaucoup ☐ Je ne sais pas		Est-ce qu'il y a quelque chose qui a dérangé votre enfant par rapport à son alimentation?	0 1 2 3 4 5 Rien du tout Oui, beaucoup ☐ Je ne sais pas	

Q4.	Peux-tu me dire, si tu as été triste?	0 1 2 3 4 5 Pas triste du tout Très triste ☐ Je ne sais pas		Votre enfant vous a-t-il/elle semblé triste?	0 1 2 3 4 5 Pas triste du tout Très triste ☐ Je ne sais pas	
Q5.	Peux-tu me dire si tu t'es senti.e heureux.se?	0 1 2 3 4 5 Pas heureux du tout Très heureux ☐ Je ne sais pas		Votre enfant vous a-t-il/elle semblé heureux ?	0 1 2 3 4 5 Pas heureux du tout Très heureux ☐ Je ne sais pas	

Q6.	As-tu pu te distraire/ jouer/ t’amuser?	0 1 2 3 4 5 Jamais Tout le temps ☐ Je ne sais pas		Votre enfant a-t ‘il/elle pu se distraire/ jouer/ s’amuser?	0 1 2 3 4 5 Jamais Tout le temps ☐ Je ne sais pas	
Q7.	Est-ce que tu as encore des questions (sur ta maladie) ?	0 1 2 3 4 5 Non, aucune Oui, plusieurs ☐ Je ne sais pas		Avez-vous l’impression que votre enfant se pose encore des questions (au sujet de sa maladie) ?	0 1 2 3 4 5 Non, aucune Oui, plusieurs ☐ Je ne sais pas	
Q8.	Est-ce que tu dors bien ?	0 1 2 3 4 5 Pas bien du tout Très bien ☐ Je ne sais pas		Est-ce que votre enfant dort bien ?	0 1 2 3 4 5 Pas bien du tout Très bien ☐ Je ne sais pas	
Q9.	Quand quelque chose te tracasse, peux-tu en parler avec quelqu’un ?	0 1 2 3 4 5 Non, jamais Oui, toujours		Quand quelque chose tracasse votre enfant, avez-vous l’impression qu’il peut le manifester/l’exprimer à quelqu’un ?	0 1 2 3 4 5 Non, jamais Oui, toujours	

		☐ Je ne sais pas			☐ Je ne sais pas	
Q10.	Est-ce que tu as des contacts avec des amis, copains autant que tu voudrais ?	0 1 2 3 4 5 Non, jamais Oui, toujours ☐ Je ne sais pas		Est-ce que votre enfant a des contacts avec des amis, copains autant qu'il/elle le voudrait ?	0 1 2 3 4 5 Non, jamais Oui, toujours ☐ Je ne sais pas	
Q11.	Si tu avais une baguette magique, est-ce que tu aimerais changer quelque chose dans ta famille? *face à un enfant anxieux, introduire prudemment cette question	0 1 2 3 4 5 Non, pas du tout Oui, absolument ☐ Je ne sais pas		Avez-vous l'impression que votre enfant souhaiterait changer quelque chose dans votre famille ?	0 1 2 3 4 5 Non, pas du tout Oui, absolument ☐ Je ne sais pas	
Q12	Te sens tu aimé(e) ? *face à un enfant anxieux, introduire prudemment cette question	0 1 2 3 4 5 Non, pas du tout Oui, absolument ☐ Je ne sais pas		Avez-vous l'impression que votre enfant se sent aimé.e ?	0 1 2 3 4 5 Non, pas du tout Oui, absolument ☐ Je ne sais pas	

<table>
<tr><td colspan="2"></td><td>Score total section A Perspective enfant</td><td></td><td></td><td>Score total section A Perspective parents</td><td></td></tr>
<tr><td colspan="6">Commentaires libres (observations enfant/adolescent) :
SECTION B. QUESTIONS A POSER uniquement AU PARENT : Ces derniers jours, …</td><td>Score</td></tr>
<tr><td>Q13.</td><td colspan="2">A quel point vos inquiétudes ont-elles un impact sur votre vie quotidienne, votre sommeil, votre travail…. ?</td><td colspan="3">0 1 2 3 4 5
Pas du tout Enormément
☐ Je ne sais pas</td><td></td></tr>
<tr><td>Q14.</td><td colspan="2">Lorsque vous le souhaitez, pouvez-vous partager avec d’autres ce que vous ressentez par rapport à la maladie de votre enfant ?</td><td colspan="3">0 1 2 3 4 5
Pas du tout fait Tout à
☐ Je ne sais pas</td><td></td></tr>
<tr><td>Q15.</td><td colspan="2">Recevez-vous, vous et votre famille, toutes les informations (au sujet de la maladie de votre enfant) dont vous avez besoin ?</td><td colspan="3">0 1 2 3 4 5
Pas du tout assez que besoin Reçu autant
☐Je ne sais pas</td><td></td></tr>
</table>

Q16.	Recevez-vous suffisamment d’aide et de conseils afin de planifier le futur de votre enfant ?	0 1 2 3 4 5 Pas du tout assez Reçu autant que besoin □Je ne sais pas	
Q17.	Vous sentez-vous en confiance par rapport aux soins à donner à votre enfant ?	0 1 2 3 4 5 Pas du tout en confiance Tout à fait en confiance □Je ne sais pas	
Q18.	Recevez-vous pour vous-même le soutien et l’accompagnement dont vous avez besoin ?	0 1 2 3 4 5 Pas du tout Tout le soutien dont nous avons besoin □Je ne sais pas	
Q19.	Les démarches administratives (matériel, traitement, soins,…) représentent-elles un poids pour vous ?	0 1 2 3 4 5 Aucun poids Poids maximal □Je ne sais pas	

Q20.	Vous arrive-t-il d’être tracassé.e par les aspects financiers liés à la maladie de votre enfant ?	0 1 2 3 4 5 Jamais Toujours ☐Je ne sais pas	
Q21.	Les soins/ médicaments que vous donnez à votre enfant représentent-ils une surcharge de travail pour vous ?	0 1 2 3 4 5 Aucune Poids maximal ☐Je ne sais pas	
Q22.	Comment évaluez-vous votre qualité de vie actuellement ?	0 1 2 3 4 5 Très mauvaise Très bonne ☐Je ne sais pas	
	SCORE TOTAL section B :		
	Commentaires libres (observations parents) :		

Adapted 22-items CPOS-2 in English

SECTION A: regarding the CHILD. Please indicate 0, 1, 2, 3, 4 or 5 or « I don’t know » for every question asked, thinking about the last few days.

	Question	**Answers**	Score	**Question**	**Answers**
Q1.	Can you tell me if you have had any pain?	0 1 2 3 4 5 No pain at all A lot of pain ○ I don’t know		Has your child had any pain?	0 1 2 3 4 5 no pain at all yes, a lot ● I don’t know
Q2.	Have you experienced any problems in (with?) your body that have been bothering you? *(Suggest if necessary: vomiting, diarrhea, nausea, insomnia*)	0 1 2 3 4 5 0 No problems in my body 5 Problems in my body are bothering me ● I don’t know		Are they any physical problems that have been bothering your child? *(Suggest if necessary: vomiting, diarrhea, nausea, insomnia*)	0 1 2 3 4 5 0 No physical problems bothered him/her 5 Yes, a lot of physical problems bothered my child ● I don’t know
Q3.	Is there anything about food that has been bothering you?	0 1 2 3 4 5 Nothing Yes, a lot ● I don’t know		Is there anything about food that has been bothering your child?	0 1 2 3 4 5 Nothing Yes, a lot ● I don’t know
Q4.	Can you tell me if you have been sad?	0 1 2 3 4 5 Not at all yes, very sad		According to you, has your child been sad?	0 1 2 3 4 5 Not sad at all Very sad

		- I don't know			- I don't know
Q5.	Can you tell me if you have been happy?	0 1 2 3 4 5 Not happy at all Very happy - I don't know		According to you, has your child been happy?	0 1 2 3 4 5 Not happy at all Very happy - I don't know
Q6.	Have you had fun? Have you been playing?	0 1 2 3 4 5 Never Always - I don't know		Has your child had fun? Has he/she been playing?	0 1 2 3 4 5 Never Always - I don't know
Q7.	Do you still have any questions (about your illness)?	0 1 2 3 4 5 No, none Yes, a lot - I don't know		Do you feel that your child still has some unanswered questions (about his or her illness)?	0 1 2 3 4 5 No, none Yes, a lot - I don't know
Q8.	Do you sleep well?	0 1 2 3 4 5 Not at all Very good - I don't know		Does your child sleep well?	0 1 2 3 4 5 Not at all Very good - I don't know

Q9.	When something bothers you, can you talk to someone about it?	0 1 2 3 4 5 No, never Yes, always • I don't know		When something is bothering your child, do you feel that he or she can express it to someone? (talk to someone about it?)	0 1 2 3 4 5 No, never Yes, always • I don't know
Q10.	Do you have as much contact with friends as you would like?	0 1 2 3 4 5 No, never Yes, always • I don't know		Do you feel that your child has as many contacts with his/her friends/peers as he/she would like?	0 1 2 3 4 5 No never Yes, always • I don't know
Q11.	If you had a magic wand, is there something you would you like to change in your family?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know		Do you feel that your child would like to change something in your family?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know
Q12	Do you feel loved? *this question is to be introduced cautiously, in the case the child is anxious	0 1 2 3 4 5 No, not at all Yes, absolutely • I don't know		Do you feel that your child feels loved?	0 1 2 3 4 5 Not at all Yes, absolutely • I don't know

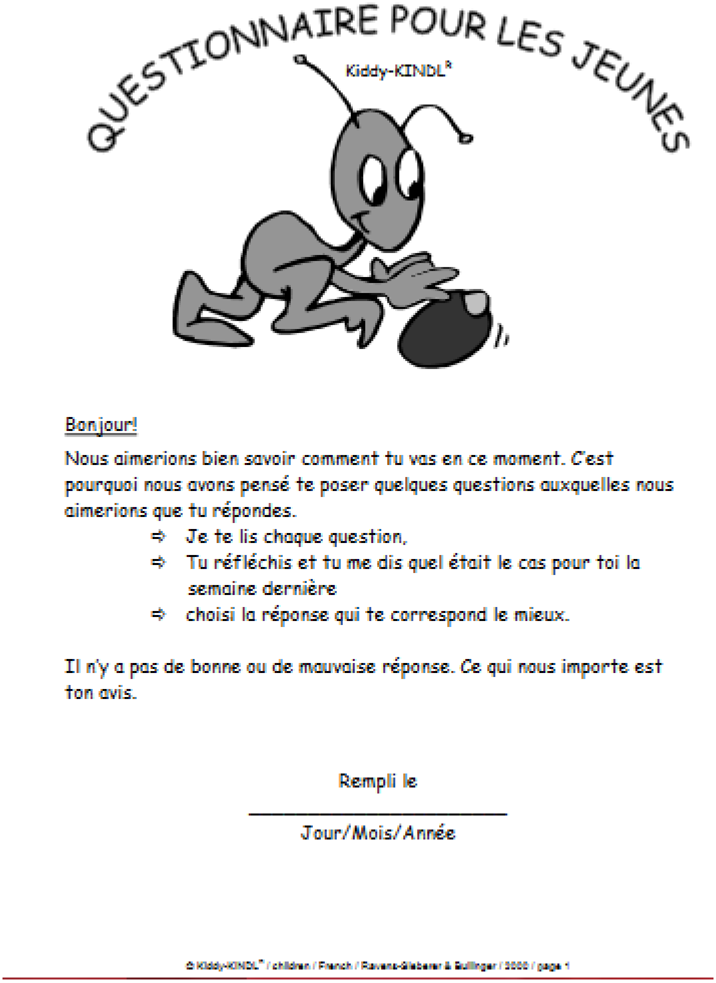
Comments/observations :

SECTION B. QUESTIONS to address only to the **PARENT** **« In the last few days, …**		
Q13.	To what extent do your concerns (worries?) have an impact on your daily life, sleep, work...?	0 1 2 3 4 5 Not at all A lot • I don’t know
Q14.	To what extent can you share with others how you feel about your child's illness?	0 1 2 3 4 5 Not at all A lot • I don’t know
Q15.	Do you and your family receive as much information as needed (about your child's illness)?	0 1 2 3 4 5 Not enough at all Receive as much as needed • I don’t know
Q16.	To what extent do you receive enough help and advice to plan for your child's future?	0 1 2 3 4 5 Not enough at all Receive as much as needed • I don’t know
Q17.	To what extent do you feel confident about caring for your child?	0 1 2 3 4 5

		Not confident at all Completely confident • I don’t know
Q18.	To what extent do you receive the support and guidance you need for yourself?	0 1 2 3 4 5 Not at all All the support needed • I don’t know
Q19.	To what extent do administrative procedures (equipment, treatment, care) represent a burden for you?	0 1 2 3 4 5 Not at all A lot • I don’t know
Q20.	Do you ever worry about the financial aspects linked to your child's illness?	0 1 2 3 4 5 Not at all A lot • I don’t know

Q21.	To what extent does the medical care you provide to your child represent an overload of work for you?	0 1 2 3 4 5 Not at all A lot ● I don’t know
Q22.	How do you currently evaluate your own quality of life?	0 1 2 3 4 5 Very bad very good ● I don’t know

KINDL *4-6 years*



QUESTIONNAIRE POUR LES JEUNES

Kiddy-KINDL®

<u>Bonjour!</u>

Nous aimerions bien savoir comment tu vas en ce moment. C'est pourquoi nous avons pensé te poser quelques questions auxquelles nous aimerions que tu répondes.

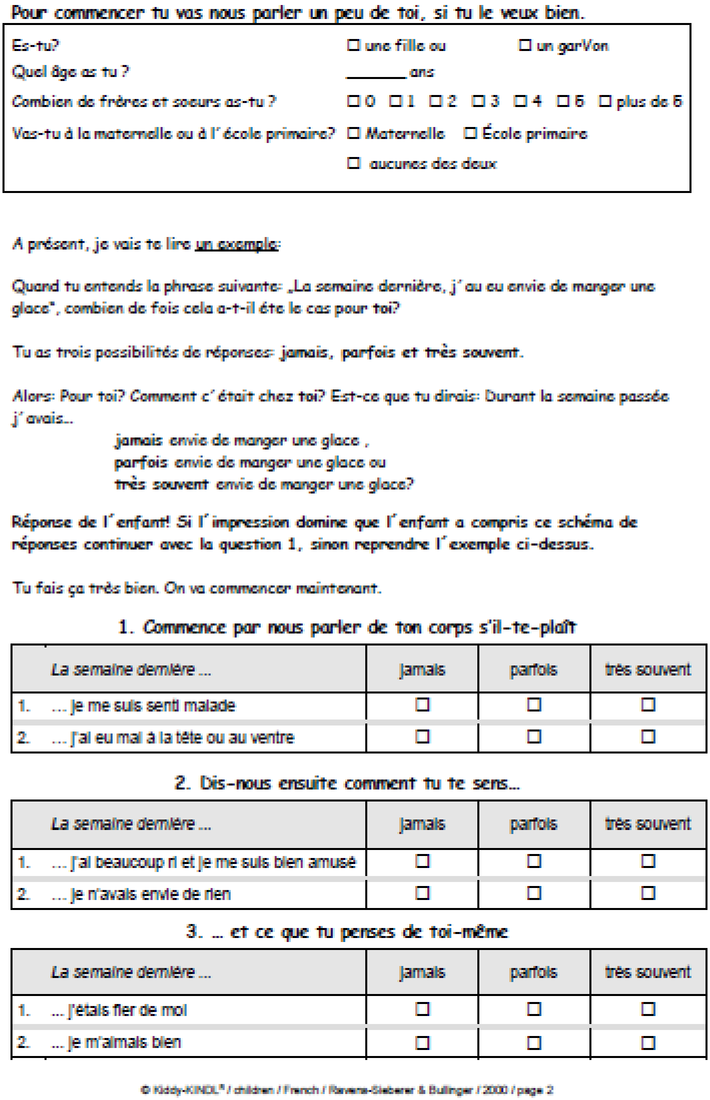
- ⇨ Je te lis chaque question,
- ⇨ Tu réfléchis et tu me dis quel était le cas pour toi la semaine dernière
- ⇨ choisi la réponse qui te correspond le mieux.

Il n'y a pas de bonne ou de mauvaise réponse. Ce qui nous importe est ton avis.

Rempli le

____

Jour/Mois/Année



Pour commencer tu vas nous parler un peu de toi, si tu le veux bien.

Es-tu?	☐ une fille ou	☐ un garVon
Quel âge as tu ?	_______ ans	
Combien de frères et soeurs as-tu ?	☐ 0 ☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5 ☐ plus de 5	
Vas-tu à la maternelle ou à l'école primaire?	☐ Maternelle	☐ École primaire
	☐ aucunes des deux	

A présent, je vais te lire un exemple:

Quand tu entends la phrase suivante: „La semaine dernière, j' au eu envie de manger une glace", combien de fois cela a-t-il éte le cas pour toi?

Tu as trois possibilités de réponses: **jamais**, **parfois** et **très souvent**.

Alors: Pour toi? Comment c'était chez toi? Est-ce que tu dirais: Durant la semaine passée j' avais..

jamais envie de manger une glace ,
parfois envie de manger une glace ou
très souvent envie de manger une glace?

Réponse de l'enfant! Si l'impression domine que l'enfant a compris ce schéma de réponses continuer avec la question 1, sinon reprendre l'exemple ci-dessus.

Tu fais ça très bien. On va commencer maintenant.

1. Commence par nous parler de ton corps s'il-te-plaît

	La semaine dernière ...	jamais	parfois	très souvent
1.	... je me suis senti malade	☐	☐	☐
2.	... j'ai eu mal à la tête ou au ventre	☐	☐	☐

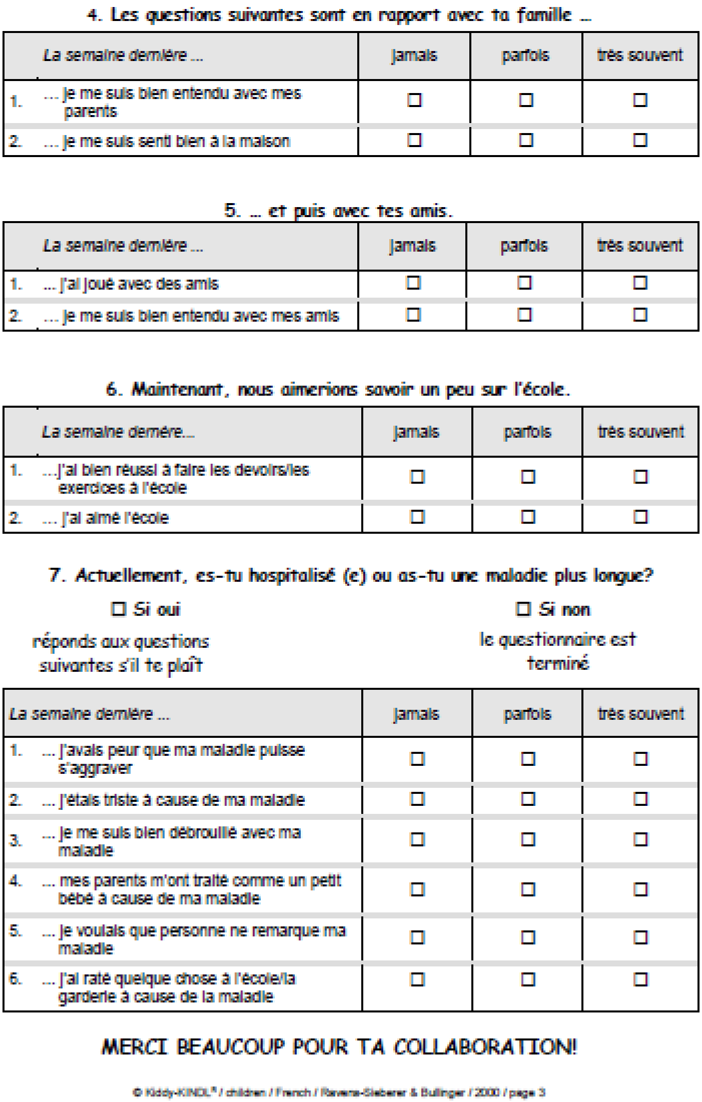
2. Dis-nous ensuite comment tu te sens...

	La semaine dernière ...	jamais	parfois	très souvent
1.	... j'ai beaucoup ri et je me suis bien amusé	☐	☐	☐
2.	... je n'avais envie de rien	☐	☐	☐

3. ... et ce que tu penses de toi-même

	La semaine dernière ...	jamais	parfois	très souvent
1.	... j'étais fier de moi	☐	☐	☐
2.	... je m'aimais bien	☐	☐	☐

© Kiddy-KINDL® / children / French / Ravens-Sieberer & Bullinger / 2000 / page 2



4. Les questions suivantes sont en rapport avec ta famille …

La semaine dernière …	jamais	parfois	très souvent
1. … je me suis bien entendu avec mes parents	☐	☐	☐
2. … je me suis senti bien à la maison	☐	☐	☐

5. … et puis avec tes amis.

La semaine dernière …	jamais	parfois	très souvent
1. … j'ai joué avec des amis	☐	☐	☐
2. … je me suis bien entendu avec mes amis	☐	☐	☐

6. Maintenant, nous aimerions savoir un peu sur l'école.

La semaine dernière…	jamais	parfois	très souvent
1. …j'ai bien réussi à faire les devoirs/les exercices à l'école	☐	☐	☐
2. … j'ai aimé l'école	☐	☐	☐

7. Actuellement, es-tu hospitalisé (e) ou as-tu une maladie plus longue?

☐ Si oui
réponds aux questions suivantes s'il te plaît

☐ Si non
le questionnaire est terminé

La semaine dernière …	jamais	parfois	très souvent
1. … j'avais peur que ma maladie puisse s'aggraver	☐	☐	☐
2. … j'étais triste à cause de ma maladie	☐	☐	☐
3. … je me suis bien débrouillé avec ma maladie	☐	☐	☐
4. … mes parents m'ont traité comme un petit bébé à cause de ma maladie	☐	☐	☐
5. … je voulais que personne ne remarque ma maladie	☐	☐	☐
6. … j'ai raté quelque chose à l'école/la garderie à cause de la maladie	☐	☐	☐

MERCI BEAUCOUP POUR TA COLLABORATION!

© Kiddy-KINDL® / children / French / Ravens-Sieberer & Bullinger / 2000 / page 3

KINDL 7-14 years

ID: ______

QUESTIONNAIRE POUR LES JEUNES

Kid-KINDL®

<u>Bonjour!</u>

Nous aimerions bien savoir comment tu vas en ce moment. C'est pourquoi nous avons imaginé quelques questions auxquelles nous te prions de répondre.

- ⇨ lis s'il-te-plaît chaque question en entier,
- ⇨ réfléchis comment c'était pendant la période indiquée,
- ⇨ coche la réponse qui te correspond le mieux,
- ⇨ ne coche s'il-te-plaît qu'une seule case pour chaque question.

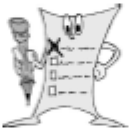
Il n'y a pas de bonne ou de mauvaise réponse. Ce qui nous importe est ton avis.

<u>Un exemple</u> :	jamais	rare-ment	parfois	souvent	toujours
La semaine dernière, j'ai eu du plaisir à écouter de la musique	☐	☐	☐	☒	☐

Rempli le

____
Jour/Mois/Année

Pour commencer, tu vas nous dire des choses à ton sujet, si tu le veux bien. *Coche une case ou écris ta réponse!*



Es-tu ☐ une fille ☐ un garçon

Quel âge as-tu ? _______ ans

Combien de frères et sœurs as-tu ? ☐ 0 ☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5 ☐ plus de 5

Quelle école fréquentes-tu?
- ☐ Ecole primaire
- ☐ Ecole de fin de scolarité
- ☐ Lycée
- ☐ Etablissement d'enseignement primaire et d'enseignement secondaire
- ☐ Collège d'enseignement secondaire
- ☐ Etablissement d'éducation spécialisée

1. *Commence par nous parler de ton corps s'il-te-plaît*

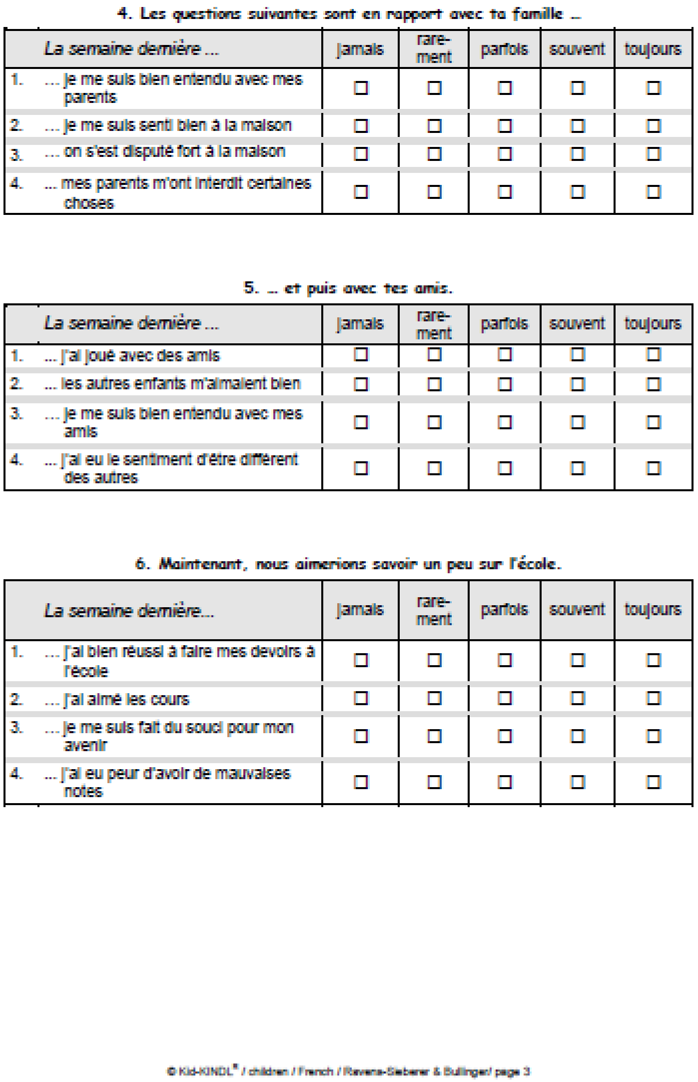
	La semaine dernière ...	jamais	rarement	parfois	souvent	toujours
1.	... je me suis senti malade	☐	☐	☐	☐	☐
2.	... j'ai eu mal à la tête ou au ventre	☐	☐	☐	☐	☐
3.	... j'étais fatigué et abattu	☐	☐	☐	☐	☐
4.	... j'avais beaucoup de force et j'avais du fond	☐	☐	☐	☐	☐

2. Dis-nous ensuite comment tu te sens...

	La semaine dernière ...	jamais	rarement	parfois	souvent	toujours
1.	... j'ai beaucoup ri et je me suis bien amusé	☐	☐	☐	☐	☐
2.	... je me suis embêté	☐	☐	☐	☐	☐
3.	... je me suis senti seul	☐	☐	☐	☐	☐
4.	... j'ai eu peur	☐	☐	☐	☐	☐

3. ... et ce que tu penses de toi-même

	La semaine dernière ...	jamais	rarement	parfois	souvent	toujours
1.	... j'étais fier de moi	☐	☐	☐	☐	☐
2.	... je me trouvais bien	☐	☐	☐	☐	☐
3.	... je m'aimais bien	☐	☐	☐	☐	☐
4.	... j'ai eu plein de bonnes idées	☐	☐	☐	☐	☐



4. Les questions suivantes sont en rapport avec ta famille ...

La semaine dernière ...	jamais	rare-ment	parfois	souvent	toujours
1. ... je me suis bien entendu avec mes parents	☐	☐	☐	☐	☐
2. ... je me suis senti bien à la maison	☐	☐	☐	☐	☐
3. ... on s'est disputé fort à la maison	☐	☐	☐	☐	☐
4. ... mes parents m'ont interdit certaines choses	☐	☐	☐	☐	☐

5. ... et puis avec tes amis.

La semaine dernière ...	jamais	rare-ment	parfois	souvent	toujours
1. ... j'ai joué avec des amis	☐	☐	☐	☐	☐
2. ... les autres enfants m'aimaient bien	☐	☐	☐	☐	☐
3. ... je me suis bien entendu avec mes amis	☐	☐	☐	☐	☐
4. ... j'ai eu le sentiment d'être différent des autres	☐	☐	☐	☐	☐

6. Maintenant, nous aimerions savoir un peu sur l'école.

La semaine dernière...	jamais	rare-ment	parfois	souvent	toujours
1. ... j'ai bien réussi à faire mes devoirs à l'école	☐	☐	☐	☐	☐
2. ... j'ai aimé les cours	☐	☐	☐	☐	☐
3. ... je me suis fait du souci pour mon avenir	☐	☐	☐	☐	☐
4. ... j'ai eu peur d'avoir de mauvaises notes	☐	☐	☐	☐	☐

7. Actuellement, es-tu hospitalisé (e) ou as-tu une maladie plus longue?

☐ Si oui — réponds aux questions suivantes s'il te plaît

☐ Si non — le questionnaire est terminé

La semaine dernière ...	jamais	rare-ment	parfois	souvent	toujours
1. ... j'avais peur que ma maladie puisse s'aggraver	☐	☐	☐	☐	☐
2. ... j'étais triste à cause de ma maladie	☐	☐	☐	☐	☐
3. ... je me suis bien débrouillé avec ma maladie	☐	☐	☐	☐	☐
4. ... mes parents m'ont traité comme un petit enfant à cause de ma maladie	☐	☐	☐	☐	☐
5. ... je voulais que personne ne remarque ma maladie	☐	☐	☐	☐	☐
6. ... j'ai raté quelque chose à l'école à cause de la maladie	☐	☐	☐	☐	☐

MERCI BEAUCOUP POUR TA COLLABORATION!

QOLLTI-F : Qualité de vie au cours d’une maladie grave: proches aidant: version 2

Instructions : Dans ce questionnaire, les questions se présentent de la façon suivante : il y a d’abord un énoncé et, ensuite, suivent deux réponses à l’opposé l’une de l’autre.

Des chiffres de 0 à 10 s’échelonnent entre les deux extrêmes. Veuillez encercler le chiffre entre 0 et 10 qui correspond le mieux à ce qui est vrai pour vous. Il n’y a pas de bonne ou de mauvaise réponse.

Des réponses honnêtes nous seront des plus utiles. EXEMPLE

J’ai faim :

Pas du tout	1	2	3	4	5	6	7	8	9	10	extrêmement

- Si vous n’avez même pas une toute petite faim, vous pouvez encercler 0.
- Si vous avez une petite faim (vous venez de terminer votre repas et il vous reste de la place pour un dessert), vous pouvez encercler 1,2 ou 3.
- Si vous avez modérément faim (car l’heure du repas approche), vous pouvez encercler 4,5 ou 6.
- Si vous avez très faim (car vous n’avez rien mangé de toute la journée), vous pouvez encercler 7,8 ou 9.
- Si vous avez extrêmement faim, vous pouvez encercler 10.

COMMENCER ICI

Veuillez répondre à toutes les questions en vous rappelant qu’il s’agit de comment vous vous êtes senti(e) AU COURS DES DEUX DERNIERS JOURS SEULEMENT

Partie A : Qualité de vie globale

A. En tenant compte de tous les aspects de ma vie (physique, émotionnel, social, spirituel et financier), ma qualité de vie au cours des deux derniers jours (48h) a été :

Très faible	1	2	3	4	5	6	7	8	9	10	excellentes

PARTIE B : Environnement

1. Au cours des deux derniers jours (48h), j'ai été satisfait(e) de l'endroit où le membre de ma famille/l'ami(e) dont je prends soin se trouvait maison, hôpital ou autre) :

Pas du tout	1	2	3	4	5	6	7	8	9	10	Tout à fait

2. Au cours des deux derniers jours (48 heures), j'ai pu bénéficier de l'intimité que je souhaitais avoir :

Pas du tout	1	2	3	4	5	6	7	8	9	10	Tout à fait

PARTIE C : Etat du patient

3. Au cours des deux derniers jours (48 heures), l'état du membre de ma famille/de l'ami(e) dont je prends soin m'a affligé(e) :

Pas souvent	1	2	3	4	5	6	7	8	9	10	Toujours

PARTIE D : Votre propre état

4. Au cours des deux derniers jours (48 heures), le niveau de contrôle que j'ai eu sur ma vie :

N'a pas été un problème	1	2	3	4	5	6	7	8	9	10	A été un problème majeur

5. Au cours des deux derniers jours (48 heures), j'ai eu le temps de m'occuper de moi-même :

Jamais	1	2	3	4	5	6	7	8	9	10	Toujours

6. Au cours des deux derniers jours (48 heures), j'ai été capable de penser clairement :

Pas souvent	1	2	3	4	5	6	7	8	9	10	toujours

7. Au cours des deux derniers jours (48 heures), je me suis senti(e) physiquement :

Extrêmement mal	1	2	3	4	5	6	7	8	9	10	Extrêmement bien

8. Au cours des deux derniers jours (48 heures), sur le plan émotionnel, je me suis senti(e) :

Extrêmement mal	1	2	3	4	5	6	7	8	9	10	Extrêmement bien

PARTIE E : Votre perspective

9. Au cours des deux derniers jours (48 heures), je me suis senti(e) bien lorsque j'ai pris soin du membre de ma famille/de mon ami(e) ou que je lui ai tenu compagnie :

Rarement ou jamais	1	2	3	4	5	6	7	8	9	10	Toujours

10. Au cours des deux derniers jours (48 heures), ma perspective sur la vie, ma foi ou ma spiritualité m'a réconforté(e) :

Pas du tout	1	2	3	4	5	6	7	8	9	10	Tout à fait

11. Présentement, j'ai le sentiment que ma vie a un sens :

Très peu de sens	1	2	3	4	5	6	7	8	9	10	Beaucoup de sens

PARTIE F : Qualité des soins

Pour les questions de cette section (12 à 14), si vous n'avez pas pris de décisions importantes ou si vous n'avez pas eu besoin de soins de santé durant les deux derniers jours, veuillez répondre en pensant aux dernières fois où cela s'est passé.

12. Au cours des deux derniers jours (48 heures), je me suis senti(e) à l'aise avec la manière dont les décisions ont été prises en ce qui concerne le membre de ma famille/l'ami(e) dont je prends soin :

Pas du tout	1	2	3	4	5	6	7	8	9	10	Tout à fait

13. Au cours des deux derniers jours (48 heures), nous avons reçu des soins lorsque nous en avons eu besoin :

Jamais	1	2	3	4	5	6	7	8	9	10	Toujours

14. Au cours des deux derniers jours (48 heures), la qualité des soins que nous avons reçus a été :

Insatisfaisante	1	2	3	4	5	6	7	8	9	10	Excellente

PARTIE G : Relations

15. Au cours des deux derniers jours (48 heures), j'ai senti que ma relation avec le membre de ma famille/l'ami(e) dont je prends soin était :

Très détendue	1	2	3	4	5	6	7	8	9	10	Tendue

16. Au cours des deux derniers jours (48 heures), j'ai senti que, dans l'ensemble, ma relation avec les autres personnes qui me sont chères a été :

Très détendue	1	2	3	4	5	6	7	8	9	10	Tendue

PARTIE H : Préoccupations financières

17. Au cours des deux derniers jours (48 heures), j'ai été stressé(e) en raison de ma situation financière :

Pas du tout	1	2	3	4	5	6	7	8	9	10	Tout à fait

Manuel d'utilisation du SEIQoL-dw (basé sur Hickey et al, BMJ 1996)

Le SEIQoL-dw doit être utilisé au cours d'un entretien semi-dirigé auprès d'un enfant/adolescent entre 6-18 ans, qui comprend, parle le français et n'a pas de déficience cognitive majeure. Prévoir un environnement calme, la présente fiche, un bic, l'échelle visuelle analogique et le disque rotatif coloré. L'entretien dure environ 20 min.

1. Compléter les questions générales relatives **à l'enfant et à son environnement** au verso.
2. **Evaluer le score douleur** à l'aide de l'échelle visuelle analogue (EVA) et notez le score.
3. Poser la question à l'enfant/adolescent :
   « Peux-tu **citer 5 domaines** qui contribuent à ta qualité de vie? La qualité de vie, c'est quand tu te sens bien avec toi et avec les autres, que tu es heureux, content. Quelles sont pour toi les domaines qui font que ta vie est bonne? Qu'est-ce qui est vraiment important pour toi ?» Si l'enfant n'a pas d'idées, vous pouvez énumérer les domaines suivants : la famille, la santé, les loisirs, les amis, l'école, les animaux domestiques, la foi/spiritualité, les mouvements de jeunesse … Notez ses réponses au verso de cette feuille.

4. « Pour chacun des domaines énoncés, peux-tu m'indiquer à l'aide de l'échelle visuelle analogique, ton **niveau de satisfaction** ? Indique un chiffre entre 0 et 10. 0 signifie que tu n'es pas satisfait du tout et 10 si tu es très satisfait par rapport à ce domaine ». Divisez la réponse par 10. Notez sur le verso de cette feuille les réponses de l'enfant pour chacun des domaines cités.

5. « Pour chacun des domaines énoncés, à l'aide du disque rotatif coloré, peux-tu m'indiquer le **degré d'importance »** ? (entre 0 et 100). Attribuer une couleur à chaque domaine cité par l'enfant. Montrez à l'enfant comment répartir les domaines selon les couleurs sur le disque rotatif. Notez ses réponses (les proportions) sur le verso de cette feuille.

6. **Effectuer le calcul de l'index SEIQoL** en additionnant les produits du niveau de satisfaction avec le degré d'importance. C'est-à-dire, l'index SEIQoL est la somme du niveau de satisfaction pour chaque domaine cité multiplié par son degré d'importance. Plus l'index SeiQoL est élevé, plus il exprime une perception individuelle élevée de qualité de vie.
   EXEMPLE :

Domaines	Niveau satisfaction (tel qu'indiqué par l'enfant sur l'EVA)	Degré d'importance (tel qu'indiqué par l'enfant sur le disque rotatif)	Multiplication	Produit
Famille	8 (divisé par 10) : 0,8	35	0,8x35	28
Loisirs	5 (divisé par 10) : 0,5	25	0,5x25	12,5
Amis	6 (divisé par 10) : 0,6	20	0,6x20	12
Ecole	6 (divisé par 10) : 0,6	10	0,6x10	6
Santé	7 (divisé par 10) : 0,7	10	0,7x10	7

Addition de tous les produits : 28+12,5+12+6+7= 65,5 => Index SEIQoL-dw

UCLouvain

Initiale Nom et Prénom
enfant/ado :……………………………………………………………………………………
Date naissance :…………………………………………………………………………….
Pathologies/Diagnostic :……………………………………………………………………
Situation familiale/fratrie :
……………………………………………………………………………………………………
……………………………………………………………………………………………………
Date entretien :……………………………………………………………………………..
Lieu entretien :……………………………………………………………………………….
Durée entretien :…………………………………………………………………………..
Personnes présentes pendant entretien :……………………………………….
Score EVA douleur juste avant de commencer entretien :………………

Domaines	Niveau satisfaction à l'aide de l'échelle EVA (0-10)	Degré d'importance à l'aide du disque rotatif (0-100)	Multiplication	Produit
	:10=		X	
	:10=		X	
	:10=		X	
	:10=		X	
	:10=		X	
Observations importantes à signaler :			Addition= Index SEIQoL-dw	

The Trieste Charter

1. To be considered as a ‘person’ until death regardless of age, location, condition and care setting.
2. To receive effective treatment, through qualified, comprehensive and continuous care, for pain and for other physical and psychological symptoms causing suffering.
3. To be listened to and properly informed about his/her illness with due consideration to his/her wishes, age and ability to understand.
4. To participate, on the basis of his/her abilities, values and wishes, in care choices regarding his/her life, illness and death.
5. To express his/her feelings, wishes and expectations and have these taken into consideration.
6. To have his/her cultural, spiritual and religious beliefs respected and receive spiritual care and support in accordance with his/her wishes and choices.
7. To have a social and relational life suitable to his/her age, conditions and expectations.
8. To be surrounded by family members and loved ones who are empowered in the organization and provision of the child’s care and who are supported in the management of the emotional and financial burdens that arise from the child’s condition.
9. To be cared for in a setting appropriate for his/her age, needs and wishes, and that enables the family to be close and involved.
10. To have access to child-specific palliative care services that respect the child’s best interest and avoid both futile or excessively burdensome practices and therapeutic abandonment.

Sources:https://www.fondazionemaruzza.org/wp/wp-content/uploads/2016/12/CartaDiTrieste200x240_ENG_REV_15_APR_2016_03.pdf

Benini F, Vecchi R, Orzalesi M. A charter for the rights of the dying child. Lancet. 2014;383(9928):1547-1548. doi:10.1016/S0140-6736(14)60746-7

Articles and book chapters

Here are presented some articles and book chapters related to paediatric palliative care and written by Marie Friedel during her PhD.

The first one explains the specificities of the paediatric liaison model in Belgium and how it ensures continuity of care for children with life-limiting conditions. It is complementary to the chapter describing the facts and figures of paediatric liaison care in Belgium (published as a book chapter in French in Manuel de soins palliatifs, De Broucker, Jacquemin (eds), Dunod, 2020.

The second one focuses on anticipatory grief and bereavement care for children facing life-limiting conditions and their families and is co-authored with a social worker, consultant in bereavement care in Zimbabwe and a paediatrician in palliative care in India. (published as a book chapter in Children's Palliative Care: an international case-based Manual, Downing J (ed), Springer, 2020.

The third article explores the perceptions and experiences of paediatric liaison teams in regard of the Belgian euthanasia law extended to minors since 2014. (Friedel et al. Int J Palliat Nurs 2018).

La liaison pédiatrique. Un modèle original de soins palliatifs pédiatriques transmuraux

This article was published as a book chapter under the title

La liaison pédiatrique. Un modèle original de soins palliatifs pédiatriques transmuraux. Friedel M. in Manuel de soins palliatifs. De Broucker D, Jacquemin (Eds.) 5ème édition. Ouvrage coordonné par R. De Berre. Dunod 2020. 1248 p. ISBN 2100712365.

Les soins palliatifs pédiatriques sont des soins actifs, holistiques, compatibles avec des soins curatifs destinés à des enfants ayant des maladies limitant ou menaçant leur vie, prodigués en équipe interdisciplinaire et ayant comme objectif essentiel de promouvoir la qualité de vie des enfants dans une approche centrée sur tous les membres de la famille (OMS, 1998 ; Abu-Saad Huijer, 2007). Les enjeux éthiques et organisationnels sont multiples de par l'hétérogénéité des maladies, des groupes d'âges concernés, mais également liés à l'impact émotionnel que représentent la maladie grave et la mort des enfants dans notre société. En tant que discipline relativement jeune, les soins palliatifs pédiatriques sont encore largement restreints à la fin de vie, peu documentés dans les publications scientifiques et dont l'impact n'est que très partiellement évalué (Friedel, 2018). Pourtant, pour atteindre les recommandations internationales (World Health Assembly, 2014) qui visent à intégrer les soins palliatifs pédiatriques dans les systèmes de soins existants, un virage est à prendre en termes de sensibilisation du grand public, de formation à des compétences spécifiques (Downing, 2013) et de recherche ciblée sur le vécu des enfants et les effets des soins (Baker, 2015).

Les modèles de soins palliatifs pédiatriques : une perspective internationale

La manière de prodiguer les soins palliatifs pédiatriques varie selon les pays et les structures de sante existantes pour adultes. Néanmoins des spécificités propres aux enfants sont observées. Une tendance générale est de favoriser au maximum les soins dans le lieu de vie familial habituel de l'enfant.

Une revue systématique a recensé au niveau international l'offre de services en soins palliatifs pédiatriques. Les auteurs montrent que ce sont les régions où les besoins de soins palliatifs pédiatriques sont les plus importants, que l'on retrouvait le moins de services. Pour soixante-cinq pour cent des pays inclus dans l'étude, aucun service de soins palliatifs pédiatriques n'était recensé (Knapp, 2011). Une étude cross-sectionnelle publiée en 2017 estime qu'au niveau mondial 21 millions d'enfants devraient bénéficier d'une approche palliative tandis que 8 millions d'entre eux auraient besoin de recevoir des soins palliatifs spécialisés (Connor, 2017).

Trois niveaux (cfr Fig. 1) décrivent le degré de spécialisation des compétences, des offres et des prestations de soins palliatifs : la sensibilisation aux soins palliatifs (au sein de la communauté, via les médecins généralistes ou maisons médicales), les soins palliatifs généraux (prodigués par exemple en unités hospitalières de pédiatrie) et les soins palliatifs spécialisés (au travers d'équipes exclusivement dédiées aux soins palliatifs pédiatriques).

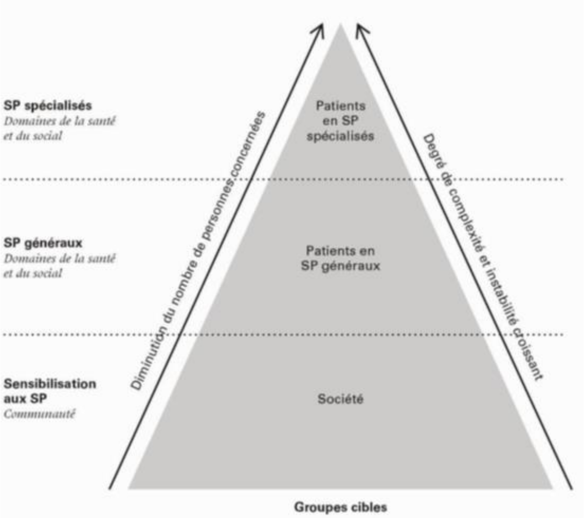


Fig. 1.1 Niveaux de soins palliatifs (https://www.palliative.ch/)

Force est de constater qu'il manque des modèles d'organisation de soins palliatifs pédiatriques clairement décrits, explicités et évalués (Virdun, 2015 ; Harding, 2014), qui permettraient de comprendre comment ces 3 niveaux s'articulent entre eux.

Néanmoins plusieurs formes d'organisation de soins palliatifs s'observent selon les contextes et pays et sont décrits brièvement ci-après (Kaye, 2015):

Soins palliatifs pédiatriques : Services interdisciplinaires qui répondent aux besoins des enfants, adolescents et jeunes adultes qui ont des conditions limitant/menaçant leur vie ainsi qu'à ceux de leurs parents. Le traitement vise à soulager la souffrance physique, psychosociale et/ou spirituelle. Le but est d'améliorer la qualité de vie de l'enfant, accompagner la famille au travers de la maladie et du deuil, facilitant les prises de décision et aidant à la coordination des soins. (Feudtner, 2013)

Soins palliatifs pédiatriques communautaires : Ce sont des soins offerts en dehors de l'hôpital en proposant des ressources et services dans des structures privées, structures de soins de santé primaires, structures de répit. Ces soins peuvent être gérés au travers de soins à domicile, de médecins traitants, avec un soutien offert au travers de personnes, par téléphone, par correspondance électronique. Ces programmes permettent une continuité des soins pour les patients qui circulent entre les soins intra et extra-hospitaliers pour améliorer la qualité de vie des enfants au travers d'un continuum de soins (Meyers, 2014).

Hospice care : Il s'agit d'une forme de soins palliatifs dans lesquels des structures d'hospices lient des services médicaux, psycho-sociaux, spirituels et de bénévolat tout en offrant un équipement médical à long terme, ainsi que des interventions diagnostiques et thérapeutiques qui correspondent aux objectifs de soins de l'enfants et de sa famille. Ces services sont habituellement financés par un forfait journalier (*all-inclusive*) et peuvent être proposés soit à la maison, soit dans une structure d'hospice séparée (Feudtner, 2013).

Cette idée de continuum de soins pour les soins palliatifs, est soulignée par l'Assemblée mondiale de la santé en janvier 2014 (WHA, 2014), mais aussi par la chambre belge des représentants dans sa loi 2016 (Ministère belge de la Justice, 2016) lorsqu'elle élargit la notion de soins palliatifs au-delà des seuls soins de fin de vie.

Les caractéristiques et critères de qualité de ces services assurant une continuité des soins « sans couture » (*seamless*) sont les suivantes (Muller, 2015 ; Kaye, 2015 ; Lichtenthal, 2015 ; Mastro, 2015 ; Weaver, 2015 ; Niswander, 2014) :

-Gestion optimale de la douleur et des autres symptômes

-Présence et activation d'une équipe interdisciplinaire

-Possibilité pour les familles d'accéder à un service 24h/24 et 7 jours sur 7

-Soins centrés sur les besoins de tous les membres de la famille

-Soins holistiques (qui répondent aux besoins bio-psycho-sociaux et spirituels)

-Communication transparente avec les membres de la famille permettant d'établir une relation de confiance et un réel partenariat dans les processus décisionnels (au sujet du traitement, du lieu des soins)

-Service de soins palliatifs pédiatriques gratuit pour les familles (intégration dans le système de sécurité sociale)

-Mise en place de matériel et équipement médicaux selon état de santé et besoins de l'enfant

-Elaboration de protocoles participant à la planification anticipée des soins (*l'advanced care planning*) :formulaire de planification préalable de niveau de soins, projet thérapeutique, protocole de non-réanimation, protocole de détresse, ordonnances automatisées, lettre de recommandation pour un retour rapide à l'hôpital si nécessaire.

-Mise en place de soins à domicile et/ou d'organisation du décès de l'enfant à domicile si tel est le souhait de la famille

-Possibilité d'accès à des structures de soins de répit

-Coordination structurée entre les différents services au travers d'outils de communication informatisée, de réunions de concertation

-Accès à des services d'accompagnement au deuil (assuré par les mêmes équipes ou leur relai)

En synthèse, les soins palliatifs pédiatriques de qualité doivent inclure l'accessibilité, la coordination, la flexibilité et l'individualisation des soins à l'enfant et à sa famille. Ce sont ces attributs qui sont supposés soutenir la qualité de vie de l'enfant et celle des membres de sa famille.

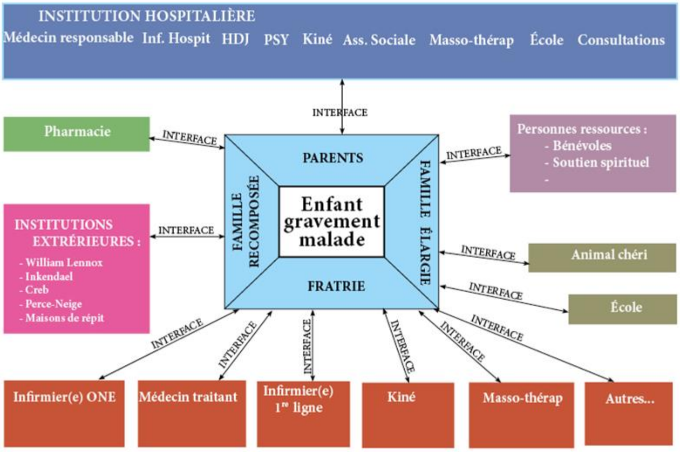
L'organisation des soins palliatifs pédiatriques en Belgique

Le modèle des équipes de liaison pédiatrique

En Belgique, les soins palliatifs pédiatriques sont organisés au moyen d'équipes de liaison qui visent à garantir la continuité des soins quels que soient le degré d'avancement de la maladie de l'enfant et le lieu de vie où celui-ci est soigné et accompagné.

> « Quand je parle de complexité, je me réfère au sens latin élémentaire du mot "complexus", "ce qui est tissé ensemble". Les constituants sont différents, mais il faut voir comme dans une tapisserie la figure d'ensemble. Le vrai problème (de réforme de pensée) c'est que nous avons trop bien appris à séparer. Il vaut mieux apprendre à relier. Relier, c'est-à-dire ne pas seulement établir bout-à-bout une connexion, mais établir une connexion qui se fasse en boucle. » (Morin, 1995)

Cette citation de Morin illustre bien le défi auquel sont confrontées les équipes de liaison, à savoir celui d'assurer la continuité des soins des enfants gravement malades, quel que soit leur âge, leur pathologie et leur lieu de vie au travers d'un maillage avec de nombreux professionnels en intra et en extra hospitalier. (Telle qu'illustrée par la Fig. 2.1.1)



Brigitte de Terwangne - SPP 2014

Fig. 2.1.1 Coordination entre les différents intervenants (de Terwangne B., 2014)

Les soins palliatifs pédiatriques ont vu le jour en Flandres, Belgique dans les années 80, à Gand (Verlooy, 2010) puis à Leuven (Renard, 2013), en partant des unités d'oncologie pédiatrique qui ont voulu créer une extension des services ambulatoires en proposant des soins curatifs (prises de sang, …) aux enfants.
C'est ainsi que, de fil en aiguille, des familles ayant un enfant qui se révélait incurable ont demandé qu'il puisse être soigné chez eux jusqu'à son décès.
Les équipes palliatives pédiatriques, appelées « de liaison », ont vu leur définition de fonction naître en 1992 sous l'impulsion de Sonja Develter aux Cliniques universitaires st Luc, à Bruxelles. Ces équipes sont caractérisées par une grande flexibilité, une disponibilité 7 jours sur 7 /24h sur 24 et une collaboration avec les équipes de première ligne (médecin traitant, infirmiers à domicile).

Les équipes de liaison pédiatrique ont été officiellement reconnues par l'Arrêté Royal du 30 novembre 2010 qui fixe les conditions auxquelles elles doivent répondre pour être agréées. (Ministère belge de la justice, 2010). Il existe 5 équipes de liaison pédiatrique en Belgique : Kites Team à l'UZ Leuven, Koester Team à l'UZ Gand, Globul'home à l'Hôpital universitaire des enfants Reine Fabiola à Bruxelles, l'Equipe Soins continus pédiatriques du CHR Citadelle a Liège, et l'équipe Interface pédiatrique aux Cliniques universitaires St-Luc à Bruxelles.

La fonction s'adresse aux jeunes patients atteints d'une pathologie chronique lourde qui s'est déclarée avant l'âge de 18 ans et a une fonction transmurale (volet interne en soutien aux équipes hospitalières et externe à l'hôpital).

Les enfants suivis peuvent se trouver en phase :

- curative, en rapport avec une affection pour laquelle un traitement curatif est possible ;
- palliative, en rapport avec une affection pour laquelle il n’y a pas ou plus de traitement curatif possible ; ou
- terminale dès l’instant où l’affection ne permet plus qu’un accompagnement de la fin de vie.

La fonction est créée dans un hôpital traitant des jeunes patients atteints d'une pathologie chronique lourde, dont au moins 50 nouveaux patients par an de moins de 16 ans présentent des affections hémato-oncologiques ou hématologiques sévères non oncologiques pouvant nécessiter une prise en charge complexe telle qu'entre autres une transplantation de cellules souches

L’équipe interdisciplinaire doit comporter : 0,5 équivalent-temps plein (ETP) pédiatre expérimenté en traitement de la douleur, 4 ETP infirmiers (dont minimum 1 doit être spécialisé en pédiatrie), 0,5 ETP psychologue et 0,5 ETP personne remplissant des fonctions administratives.

Ces personnes doivent avoir suivi une formation spécifique en SP, en particulier en ce qui concerne les jeunes enfants et un infirmier et un médecin doivent être joignables 7 jours sur 7.

Les missions de l’équipe de liaison pédiatrique sont définies comme suit:

-promouvoir la communication entre, d'une part l'équipe hospitalière et d'autre part, les acteurs de 1ère ligne ;

-promouvoir la continuité du traitement hospitalier lorsque le jeune patient quitte l'hôpital pour poursuivre le traitement à son domicile ou inversement ;

-fournir des informations sur la fonction "liaison pédiatrique" auprès des patients et des prestataires de soins;

-formuler des avis sur la liaison pédiatrique aux prestataires de soins ainsi qu'à la direction de l'hôpital en vue de la politique à mener en la matière.

L’analyse quantitative et qualitative des rapports annuels des 5 équipes de liaison pédiatrique entre 2010 et 2014 montre qu’en moyenne 720 enfants/adolescents sont suivis chaque année par les équipes de liaison en Belgique, 85 d’entre eux décèdent chaque année, dont la moitié à domicile (Friedel a), 2018).

Leurs domaines d'activités principales sont repris dans la Fig. 2.1.2 ci-après (Friedel 2016, Friedel 2018).

Fig.2.1.2 Activités principales des équipes de liaison pédiatrique (Friedel, 2018)

La coordination et la communication avec les différents partenaires de la première ligne (médecin traitant, infirmières et pédiatre du domicile) ainsi qu'avec les acteurs de soins hospitaliers prennent une part prépondérante.

Les soins palliatifs sont intégrés aux soins curatifs. Différencier les deux soins est d'ailleurs parfois difficile. Le terme soins de liaison semble plus approprié et conforme à la réalité clinique.

Le modèle de liaison pédiatrique entraîne un changement de paradigme au niveau des soins palliatifs pédiatriques. Il offre l'opportunité et la structure organisationnelle pour proposer les soins palliatifs pédiatriques non plus uniquement lorsque l'enfant est en fin de vie, mais dès le diagnostic de maladie limitant ou menaçant la vie, rejoignant ainsi les recommandations internationales (WHA 2014)

L'évolution des définitions des soins palliatifs pédiatriques peut être illustrée par la Fig. 2.3 qui présente l'évolution de l'intégration des soins palliatifs aux soins curatifs. Le modèle a) juxtapose les soins palliatifs aux soins curatifs. Tandis que le modèle b) propose une introduction progressive des soins palliatifs au fur et à mesure que les soins curatifs se rarifient. Enfin, le modèle c) suggère une intégration des soins palliatifs entendus avant tout comme des soins qui guérissent (*healing*) même en l'absence de succès thérapeutique. Il est à noter que le processus de deuil est à accompagner bien avant le moment du décès. (Milstein 2005).

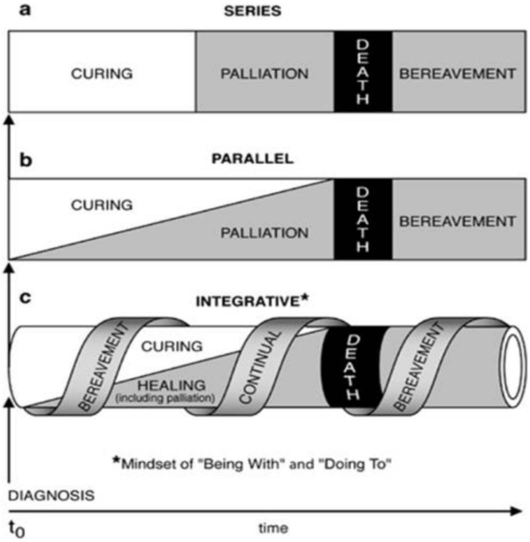


Fig. 2.1.3 Les soins palliatifs intégrés

Cette continuité des soins se réalise sur trois plans : sur un plan géographique (que l'enfant soit soigné au domicile, à l'hôpital, dans une structure de répit ou dans une institution médico-sociale), un plan temporel (de l'annonce de la maladie grave à la phase palliative et à l'accompagnement au deuil) et un plan relationnel (canaux de communication efficaces et établissement de relation de confiance). Cette triple continuité contribue à des soins palliatifs pédiatriques de qualité (Friedel, 2014).

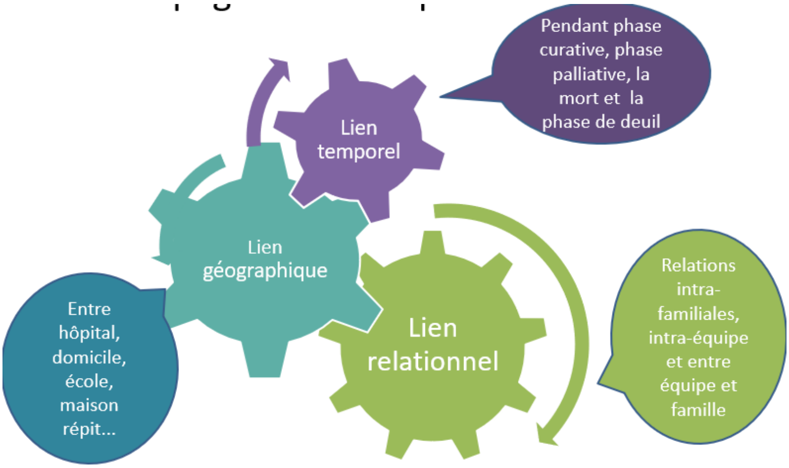


Fig. 2.1.4 Triple continuité de la liaison pédiatrique (Friedel, 2014)

Etre parents d'un enfant gravement malade et le soigner au long cours risque d'entraîner un état d'épuisement physique et/ou psychique. Dans le passé, il arrivait que certains enfants doivent être hospitalisés pour permettre aux parents de souffler. Suite à ce constat et l'absence de structures d'accueil satisfaisantes, trois maisons de répit pédiatrique ont été créées en Belgique. Cinq groupes d'enfants peuvent bénéficier d'un séjour en maison de répit:

1. Les enfants présentant une maladie qui peut entraîner une mort prématurée et dont le traitement intensif prolongé peut échouer (cancer, atteinte cardiaque, atteinte rénale…)

2. Les enfants dont la maladie entraîne inévitablement une mort prématurée. Ces enfants peuvent avoir besoin de longues périodes de traitements intensifs destinés à prolonger leur vie et à leur permettre de participer à des activités normales (mucoviscidose, dystrophie musculaire…)

3. Les enfants présentant une maladie progressive sans espoir de guérison. Les traitements sont palliatifs et peuvent s'étendre sur plusieurs années (maladie de Batten, mucopolysaccharidose…)
4. Les enfants présentant une maladie non progressive accentuant leur vulnérabilité et accroissant les risques de complication non prévisibles avec détérioration sévère de leur état (accidents avec atteintes neurologiques, paralysie cérébrale grave,…)

5. Les nouveau-nés dont la survie est fortement menacée.

Ces 5 groupes concernent les groupes d'enfants repris dans la définition des soins palliatifs pédiatriques publiée en 2007 par l'Association Européenne de Soins Palliatifs (Abu-Saad Huijer, 2007). Chaque enfant entrant dans un des 5 catégories reprises ci-dessus peut bénéficier de 32 jours de répit/an. Le financement des séjours est majoritairement pris en charge par le gouvernement fédéral. Une faible participation aux frais est néanmoins demandée aux familles. Dubois et al. décrivent le processus que les parents parcourent pour dépasser les sentiments de culpabilité, avant d'accepter de confier leur enfant à une équipe interdisciplinaire travaillant en maison de répit (Dubois, 2018).

De plus, plusieurs études (Lichtenthal, 2015 ; Hendrickson, 2009 ; Vander Geest, 2014 ; Rosenberg, 2015) ont montré que le décès d'un enfant représentait un risque important de compromettre la santé mentale des différents membres de la famille. Par conséquent, les soins palliatifs pédiatriques ne devraient pas s'arrêter au moment du décès de l'enfant mais doivent inclure l'accompagnement au deuil des membres de la famille : parents, fratrie, grands-parents, voire même le petit-ami.

En effet, pendant la phase de la maladie de l'enfant, les familles sont souvent en contact avec de nombreux professionnels de la santé. Le risque de se sentir abandonnée après la mort de leur enfant doit être prévenu par des contacts entre l'équipe de liaison et celle-ci au moins à 1 mois, 3 mois, 6 mois et 12 mois. Une adaptation flexible de cet accompagnement au deuil se fait en respectant les besoins et demandes de chaque famille.

Une rencontre post-décès entre la famille et le médecin pédiatre spécialiste devrait être toujours possible et est souvent appréciée par les familles lorsqu'elles ont encore des questions médicales en suspens, susceptibles d'entraver leur processus de deuil.

Enjeux éthiques des soins palliatifs pédiatrique dans le contexte de la loi belge sur l'euthanasie étendue aux mineurs d'âge

En février 2014, la Belgique a été le premier pays au niveau mondial à étendre la loi dépénalisant l'euthanasie aux enfants, sans aucune limite d'âge (Clément de Cléty, 2014, 2016).

Alors qu'en 2002, la loi belge sur l'euthanasie concernant les adultes dépénalisait celle-ci pour des personnes ayant une maladie incurable et une douleur physique ou psychique accompagnée d'une souffrance insupportable, l'extension de cette loi aux mineurs d'âge ajoute cependant des restrictions supplémentaires (Service Public Fédéral, Justice, 2014) :

-Il doit s'agir de la phase terminale d'une maladie accidentelle ou pathologique avec la survenue probable du décès à brève échéance (Article 2)
-La douleur physique doit être constante, insupportable, réfractaire aux traitements. Le critère de douleur psychologique est exclu pour les mineurs d'âge. (Article 2c)
-L'enfant doit être conscient et faire une demande écrite. (Article 2f)
-Le consentement écrit des deux parents doit être obtenu. (Article 2f)
-La capacité de discernement doit être évaluée par un psychiatre ou un psychologue. (Article 2d)
-Une information sur les possibilités de recevoir un soutien psychologique doit être donnée aux familles et aux professionnels. (2g)
-Un deuxième médecin doit être consulté pour avis.

L'euthanasie en Belgique est définie comme « l'acte (médical), pratiqué par un tiers (médecin), qui met intentionnellement fin à la vie d'une personne à la demande de celle-ci. » (Service Public Fédéral, Santé publique, Sécurité de la chaîne alimentaire et Environnement, 2018).

En 25 ans d'existence, c'est-à-dire entre 1989 et 2014, les 5 équipes de liaison pédiatrique affirment ne jamais avoir été confronté à des demandes d'euthanasie venant d'enfants ou d'adolescents (Renard et al., 2018).

Pourtant, en deux ans, entre le 1er janvier 2016 et le 31 décembre 2017, parmi 4437 euthanasies déclarées et enregistrées auprès de la Commission fédérale de contrôle et d'évaluation de l'euthanasie, trois d'entre elles ont concernées des mineurs d'âges. Il s'agissait d'enfants de 9, 11 et 17 ans, dont le décès était prévisible à brève échéance et qui souffraient soit d'une mucoviscidose, d'un glioblastome ou de la maladie de Duchenne. (Commission fédérale de contrôle et d'évaluation de l'euthanasie, 2018).
Ces situations suscitent des questionnements éthiques à plus d'un titre (Friedel, 2014, 2018 b) :
Comment appréhender le processus fixé par la loi au regard des interrelations faites de loyauté et de protection qui existent entre l'enfant et sa famille ?
Comment évaluer le fait que la procédure d'euthanasie cause moins de tort à la famille qu'une sédation continue jusqu'au décès ?
Quelle part de responsabilité pèse sur l'enfant dans la prise de décision d'euthanasie ?
Et quelles en sont les conséquences à court et à moyen terme sur le deuil parental et celui des fratries ?

La procédure belge d'euthanasie, si elle est décryptée sous la loupe du concept de l'autonomie relationnelle, pose question. En effet, il est possible qu'elle ne tienne pas suffisamment compte des relations interpersonnelles complexes, entre l'enfant et ses parents, lorsqu'elles sont mises à l'épreuve de la maladie grave et de la fin de vie.

Dans un processus d'aide à la décision clinique éthique, Hain propose de soupeser chaque geste clinique en se posant préalablement les 3 questions suivantes (Hain, 2016):
Quelle est l'intention explicite et implicite du geste envisagé ?
Quelle est l'action envisagée ?
Quelles en seront les conséquences ?

Face à la nécessité d'impliquer les adolescents dans les prises de décision le concernant, et sans pour autant lui incomber la responsabilité que représente la décision d'euthanasie, plusieurs outils ont été développés dans les pays anglo-saxons : my quality (Harris, 2016), 5 wishes (Lyon, 2014), le SEIQoL (Hickey, 1996). Ils permettent aux enfants/adolescents d'exprimer ce qui est important pour eux et de faire part de leur souhait en matière d'options thérapeutiques, de retour à domicile ou de tout ce qui permet d'améliorer leur qualité de vie selon leur perspective.

Plaidoyer pour des soins palliatifs pédiatriques durables

Les soins palliatifs pédiatriques durables sont des soins qui ont démontré leur efficacité et qui tiennent compte à long terme des ressources humaines, matérielles et organisationnelles.
Cette réflexion autour de ce que sont des soins palliatifs durables nous amène à détailler deux points.

Le premier point d'attention a trait au critère d'efficacité. Qu'entend-on par des soins palliatifs efficaces, c'est-à-dire qui ont fait leur preuve ? Selon la définition des soins palliatifs, ce sont des soins qui améliorent, ou du moins soutiennent la qualité de vie des enfants. Mais comment évaluer la qualité de vie lorsqu'un enfant se trouve en phase palliative ?
Plusieurs études soulignent le fait qu'il n'y a actuellement pas d'instruments capables de mesurer la qualité de vie des enfants en contexte palliatif (Coombes, 2016 ; Huang, 2010)
De nombreux défis se posent en effet qui ont trait à l'hétérogénéité des maladies rencontrées, des spécificités liées aux tranches d'âges, de la nécessité de combiner la perspective parentale avec celle de l'enfant et enfin aux difficultés d'impliquer des enfants vulnérables dans des protocoles de recherche (Friedel, 2019). Pourtant, l'évaluation de l'impact des soins palliatifs est nécessaire non seulement pour améliorer les soins individualisés aux enfants, mais aussi pour rendre crédible cette discipline encore souvent restreinte à un art intuitif et réduite à la phase de fin de vie. Evaluer des soins les rend plus crédibles et peut ainsi contribuer à assurer les financements à plus long terme des équipes de soins palliatifs pédiatriques.

Le deuxième point d'attention a trait à la nouvelle définition des soins palliatifs proposée tout récemment par l'International Association of Hospice and Palliative Care (IAHPC, 2018), qui va plus loin que les définitions antérieures en visant désormais non seulement la qualité de vie du patient, celle de sa famille mais également, et cela est nouveau, celle des soignants.

Cette définition rejoint les propos de Bodenheimer et al. (Bodenheimer, 2014) qui complète les trois objectifs décrits précédemment par Berwick (Berwick, 2008) en matière de soins de santé : qualité de vie du patient, qualité et efficience des soins, en y ajoutant la qualité de vie des soignants.

Les équipes de soins palliatifs pédiatriques exerçant une discipline relativement jeune, à la croisée entre la pédiatrie et la médecine palliative sont donc appelées à se définir davantage, à documenter les activités réalisées et à participer à des recherches visant à évaluer l'impact de leurs services non seulement sur la satisfaction des familles mais sur la qualité de vie de celles-ci. Ce processus continu d'évaluation ne pourra se faire sans une attention constante à ce qui favorise le bien-être des équipes, au contact quotidien avec la souffrance des enfants et de leurs familles. L'accès à des supervisions régulières et systématiques ou de tout autre dispositif préventif permettant la mise en parole des émotions et expériences humaines intenses vécues par les membres des équipes représente une condition indispensable à la qualité des soins.

Bibliographie :

Abu-Saad Huijer H., Benini F., Craig F., H. Kuttner L., Wood C., Feraris P., Zernikow B. (2007), IMPaCCT: Standards for paediatric palliative care in Europe. *Eur J Palliat Care*,14,2-7.

Arrêté Royal du 15 novembre 2010 fixant les normes auxquelles la fonction liaison pédiatrique doit répondre pour être agréée. http://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html

Berwick D., Nolan T., Whittington J. (2008), « The Triple Aim: care, health, and cost », *Health Aff (Millwood),*27,3,759-769.

Bodenheimer T., Sinsky C. (2014), « From triple to quadruple aim: care of the patient requires care of the provider », *Ann Fam Med*,12,6,573-576.

Clément de Cléty S, Friedel M, Verhagen AA, Lantos JD, Carter BS. (2016), «Please Do Whatever It Takes to End Our Daughter's Suffering! » Paediatrics,137,1.
Connor S., Downing J., Marston J (2017), « Estimating the Global Need for Palliative Care for Children: A Cross-sectional Analysis », *Pain Symptom Manage*, 53,2,171-177.

Commission fédérale de contrôle et d'évaluation de l'euthanasie (2018). Commission euthanasie 8ème rapport.
https://organesdeconcertation.sante.belgique.be/fr/documents/cfcee-rapport-euthanasie-2018

Coombes L., Wiseman T., Lucas G., Sangha A., Murtagh FE. (2016), « Health-related quality-of-life outcome measures in paediatric palliative care: A systematic review of psychometric properties and feasibility of use », *Palliat Med* ,30,10,935-949.

Downing J., Ling J., Benini F., Payne S., Papadatou D. (2013), Report of the EAPC Children's Palliative Care Education Taskforce. « Core Competencies for Education in Paediatric Palliative Care ».

Dubois A-C, Cuvelier S., Murinni C., Aujoulat I. « Avoir recours au répit: regards sur la prise de décision de parents d'enfants gravement malades », *Pratiques Psychologiques*,24,3,257-275.

European Association of Palliative Care (2007), « IMPaCCT: des recommandations pour des soins palliatifs pédiatriques en Europe » *European Journal of Palliative Care*,14,3,109-114. http://www.sfap.org/system/files/impacct-recos-sp-pediatriques-europe.pdf

European Association of Palliative Care, Fondazione Maruzza (2009), « Les soins palliatifs pour les nourrissons, les enfants et les adolescents. Les faits ».
http://www.fondazionemaruzza.org/wp/wp-content/uploads/2016/10/fatti-fra.pdf

Feudtner C., Friebert S., Jewell J. (2013), « Paediatric palliative care and hospice care commitments, guidelines, and recommendations. Section on hospice and palliative Medicine and committee on Hospital Care », *Paediatrics*, 132:966-972.

Friedel M. (a), Brichard B., Fonteyne C, Renard M, Misson JP, Vandecruys E, Tonon C, Verfaillie F, Hendrijckx G, Andersson N, Ruysseveldt I, Moens K, Degryse JM, Aujoulat I. (2018), « Building Bridges, Paediatric Palliative Care in Belgium: A secondary data analysis of annual paediatric liaison team reports from 2010 to 2014», *BMC Palliat Care*, 22, 17,1,77. https://bmcpalliatcare.biomedcentral.com/track/pdf/10.1186/s12904-018-0324-2

Friedel M. (b), Terwangne B., Brichard B., Ruysseveldt I., Renard M. (2018), « The Belgian euthanasia law and its impact on the practises of Belgian paediatric palliative care teams. » *Int J Palliat Nurs*, 2, 24,7, 333-337.

Friedel M., Brichard B., Fonteyne C., Misson JP, Renard M., Vandecruys E., Verfaillie F. , Ruysseveldt I. , Tonon C., Hendrijckx G., Aujoulat I., THE BELGIAN LIAISON MODEL OF PAEDIATRIC PALLIATIVE CARE. First findings of a retrospective study based on a national database. Presentation at the 2nd Conference of the International Children's Palliative Care Network, Argentinia, May 2016.

Friedel M., Aujoulat I., Dubois A.- C., Degryse J.-M. (2019). «Instruments to measure outcomes in paediatric palliative care », *Paediatrics,*143,1.

Friedel M. (2014), « Does the Belgian law legalising euthanasia for minors really address the needs of life-limited children? » *Int J Palliat Nurs*, 20,6, 265-267.

Friedel M. (2014), « Soins palliatifs en pédiatrie. Ethique et relation au patient »
Soins Pediatr Pueric, 281, 42-46.

Friedel M. (2014), « D'un passage à l'autre. Analogies entre l'accouchement et la mort. » *Soins Pediatr Pueric,* 37, 289, 37-41.

Hain R. (2016), Oral conference, 2nd International Children's Palliative Care Network ICPCN Conference, Buenos Aires, Argentinia.

Harding R., Albertyn R. et al. (2014), « Paediatric palliative care in sub-saharan Africa : a systematic review of the evidence for care models, interventions and outcomes », *Journal of Pain and Symptom Management*, 47,3,642-651.

Harris N, Beringer A, Fletcher M. (2016), « Families' priorities in life-limiting illness: improving quality with online empowerment », *Arch Dis Child*,101,3,247-252.

Hendrickson KC. (2009) « Morbidity, mortality, and parental grief: a review of the literature on the relationship between the death of a child and the subsequent health of parents », *Palliat Support Care*,7,1,109-119.

Hickey AM, Bury G, O'Boyle CA, Bradley F, O'Kelly FD, Shannon W. (1996), «A new short form individual quality of life measure (SEIQoL-DW): application in a cohort of individuals with HIV/AIDS », *BMJ*,313,7048,29-33.

Huang IC, Shenkman EA, Madden VL, Vadaparampil S, Quinn G, Knapp CA. (2010), « Measuring quality of life in paediatric palliative care: challenges and potential solutions », *Palliat Med*, 24,2,175-82.

Kaye E, Rubenstein J, Levine D, et al. (2015), « Paediatric palliative care in the community », *CA Cancer J Clin*, 65(4):316-33.

Knapp C., Woodworth L., Wright M. et al. (2011), « Paediatric palliative care provision around the world: a systematic review », *Paediatric Blood & Cancer*, 57,361-368.

Lichtenthal W., Sweeney C. (2015), « Bereavement follow-up after the death of a child as a standard of care in paediatric oncology », *Paediatric Blood & Cancer*, 62,5, S 834-869.

Lyon ME, Jacobs S, Briggs L, et al. (2014), «A longitudinal, randomized, controlled trial of advance care planning for teens with cancer: anxiety, depression, quality of life, advance directives, spirituality », *J Adolesc Health*, 54,6,710-717.

Mastro K., Johnson J. et al. (2015), « The benefits of a nurse-driven, patient- and family-centered Paediatric Palliative Care Program », *J Nurs Adm*, 45,9,423-428.

Meyers K., Kerr K, Cassel JB (2014), Up close : a field Guide to Community-based Palliative Care in California. Sacramento, CA: California Health Care foundation.

Milstein J. (2005), « A paradigm of integrative care. Healing with curing throughout life » *Journal of Perinatology*,25,563–568.

Ministère belge de la Justice (2016), 21 JUILLET 2016- Loi modifiant la loi du 14 juin 2002 relative aux soins palliatifs en vue d'élargir la définition des soins palliatifs (M.B. du 29.08.2016) LOI 2002/06/14_MOD2016/07/21

Morin E. (1995), « La stratégie de reliance pour l'intelligence de la complexité », *Revue Internationale de Systémique*, 9, 2.

Müller E., Levy C. et al. (2015), « Paediatric Palliative Care: Current evidence and evidence gaps », *Journal of Paediatrics*,166,6,1536-1540.

Niswander L., Cromwell P. et al. (2014), « End-of-life Care for children enrolled in a community-based Paediatric Palliative Care Program », *Journal of Palliative Medicine*,17,5,589-591.

Renard M.,Van den Eynde G., Van Ruysseveldt I. Paediatric palliative care as a transmural liaison model : experience of 10 years of palliative care for all children. Oral Presentation at Maruzza Congress 2013.
http://www.maruzza.org/new/wpcontent/uploads/2013/02/05-Renard.pdf

Rosenberg AR, Postier A, Osenga K, Kreicbergs U, Neville B, Dussel V, Wolfe J. (2015), «Long-term psychosocial outcomes among bereaved siblings of children with cancer », *J Pain Symptom Manage*, 49,1,55-65.

Service Public Fédéral, Santé Publique, Sécurité de la chaîne alimentaire et Environnement (2018), consultable sur https://www.health.belgium.be/fr/sante/prenez-soin-de-vous/debut-et-fin-de-vie/euthanasie

Service Public Fédéral, Justice (2014), 28 février 2014. Loi modifiant la loi du 28 mai 2002 relative à l'euthanasie, en vue d'étendre l'euthanasie aux mineurs, consultable sur https://www.health.belgium.be/sites/default/files/uploads/fields/fpshealth_theme_file/loi20140228_frnl.pdf

Van der Geest I., Darlington A-S, Streng I., Michiels E., Pieters R., van den Heuvel-Eibrink M (2014), «Parents' experiences of paediatric palliative care and the impact on long-term parental grief », *J Pain Symptom Manage*, 47,6,1043-1053.

Verlooy J., Mattheeuws, K., De Clercq, M. J., De Porre, J. (2010), « Nearly twenty years experience of palliative and curative home-care for paediatric hematology/oncology patients », *Paediatric Blood & Cancer*,55,5,957.

Virdun C, Brown N et al (2015), «Elements of optimal paediatric palliative care for children and young people: an integrative review using a systematic approach. » *Collegian*, 22,421-431.

Weaver, Heinze et al. (2015), « Palliative care as standard in Paediatric Oncology », Paediatric Blood &Cancer 2015, 62(5): S829-833.

World Health Organization. (2018), Integrating palliative care and symptom relief into paediatrics. A WHO guide for health care planners, implementers and managers. http://apps.who.int/iris/bitstream/handle/10665/274561/9789241514453-eng.pdf?ua=1

World Health Assembly (WHA), Strengthening of palliative care as a component of integrated treatment within the continuum of care. 2014. 134th session EB134.R7.

Quelques références incontournables:

Abu-Saad Huijer H., Benini F., Craig F., H. Kuttner L., Wood C., Feraris P., Zernikow B. (2007), IMPaCCT: Standards for paediatric palliative care in Europe. Eur J Palliat Care,14,2-7. http://www.eapcnet.eu/LinkClick.aspx?fileticket=ShMQyZuTfqU%3D

American Academy of Paediatrics (2013). Paediatric Palliative Care and Hospice Care Commitments, Guidelines, and Recommendations. SECTION ON HOSPICE AND PALLIATIVE MEDICINE AND COMMITTEE ON HOSPITAL CARE. Paediatrics. VOLUME 132 / ISSUE 5. https://paediatrics.aappublications.org/content/paediatrics/132/5/966.full.pdf

Amery J. (2016), *A really practical handbook of children's palliative care for doctors and nurses anywhere in the world*. http://www.icpcn.org/wp-content/uploads/2016/04/A-REALLY-PRACTICAL-Handbook-of-CPC.pdf

Benini F., Vecchi R., Lazzarin P., Jankovic M., Orsi L., Manfredini L., Drigo P., Sellaroli V., Gangemi M., Spizzichino M., Orzalesi M. (2017), « The rights of the dying child and the duties of healthcare providers: the Trieste Charter », *Tumori*, 21,103,1,33-39. http://www.fondazionemaruzza.org/en/charter-of-the-rights-of-the-dying-child-the-trieste-charter/

Canadian Hospice Palliative Care Association (2006), Association canadienne de soins palliatifs (ACSP) and Canadian Network of Palliative Care for Children, réseau canadien de soins palliatifs pour enfants. Paediatric Hospice Palliative Care. Guiding Principles and Norms of Practice. http://www.chpca.net/media/7841/Paediatric_Norms_of_Practice_March_31_2006_English.pdf

European Association of Palliative Care, Fondazione Maruzza (2009), « Les soins palliatifs pour les nourrissons, les enfants et les adolescents. Les faits ».

http://www.fondazionemaruzza.org/wp/wp-content/uploads/2016/10/fatti-fra.pdf

Frache S., Etourneau F., Lecaudey S. (2018), Soins palliatifs pédiatriques. Des repères interdisciplinaires pour une pratique soignante, Coll. Soigner et Accompagner, Paris, Ed. Lamarre.

National Institute for Health and Care Excellence/NICE (2016). End-of-life care for infants, children and young people. https://www.nice.org.uk/guidance/indevelopment/gid-cgwave0730
https://www.nice.org.uk/guidance/ng61/evidence/full-guidance-pdf-2728081261

National Hospice and Palliative Care Organization (2015). Facts for Practice for Paediatric Palliative Care and Hospice.
https://www.nhpco.org/sites/default/files/public/quality/Paediatric_Facts-Figures.pdf

Together for short lives (2012), A guide to end-of-life care of children and young people before death, at the time of death and after death. http://www.togetherforshortlives.org.uk/assets/0000/1855/TfSL_A_Guide_to_End_of_Life_Care_5_FINAL_VERSION.pdf

Anticipatory grief and bereavement in children

This article was published as a book chapter under the title : Anticipatory grief and bereavement in children. Hunt J. Talawadekar P, Friedel M. pp.201-210 in Children's Palliative Care: An International Case-Based Manual. Downing J. (ed), Springer. 2020. 290 p. DOI: 10.1007/978-3-030-27375-0_16   ISBN 978-3-030-27375-0

Key Learning Points

1. Both anticipatory grief and bereavement of children demand honesty, reassurance and safe expression of emotions
2. Anticipatory grief is triggered by disclosure of diagnosis of a life-threatening condition and prepares the child and family for dying and death
3. Bereavement includes a wide range of emotions, experiences, thoughts, physical symptoms, social and spiritual changes over a long period of time, at differing times and with varying intensities and combinations
4. Bereaved children and young people need to be listened to with empathy, permission and facilitation to grieve in their unique way, assurance that there is no right or wrong way to grieve, and given time and support for feelings to be expressed
5. Grief reactions of children and young people will change over time and maturity, and vary according to gender, culture, age, previous losses and experiences, family support, the relationship to the deceased and the nature of the death

Introduction

In this chapter we take the position that there is no one correct way to think about dying, death and bereavement. There is a general lack of evidence regarding which type of interventions are the most effective in bereavement care, but a recently published Norwegian study among cancer bereaved parents identified some factors associated with a resilience process: perception of self, social resources, family cohesion and importance of relations within a "system" (family, village) (Vegsund HK et al 2019).

This holistic approach informs our chapter with thoughts that may assist during the illness trajectory of the dying child, and for the bereaved family after his death. In a departure from other texts that focus on typical child developmental approaches to dying and death, we encourage adopting a non-evaluative stance and remaining curious about the child patient's, siblings' and parents' meanings and stories about life and death. While the impact of a child's death is known to be associated with distress, anxiety, depression and sometimes post-traumatic stress disorder (Wikman A et al 2018, Ljungman L et al 2015, Ljungman et al 2016), we propose that much of managing anticipatory grief and the grief after the death of a child is not necessarily the domain of professionals. By helping families to better listen to, interact and communicate with children, we can support them in not avoiding the child, but remaining close and present, bearing the pain, fear and sadness that comes with this devastating news (Bluebond-Langner 1978). When the child has received formal care, ongoing relationships between providers and bereaved families appear significant (Snaman JM et al 2016).

Amos is a 15 year old boy diagnosed with acute lymphocytic leukaemia. He lives with his parents Sybil and John, and his younger sister Pretty (10). His older brother Elijah died in hospital from the same condition 5 years ago, a distressing and frightening time for all the family.

The palliative care team meets Amos who is suffering a relapse of his condition having received treatment, and his distressed family at their simple home. They ask "what do you think is happening with Amos?" Sybil describes similar signs of deterioration that Elijah manifested before he died, but refuses to acknowledge that Amos will die, preferring to only talk of hope and future plans. She admits to spoiling him with the few luxuries she can afford. She is angry with John who drinks heavily and spends hours out of the house, despite being unemployed. No one seems to know where Pretty is until one of the team finds her hiding with her doll in a tree in the garden.

When Amos is asked how he is feeling he stays quiet. His father asks "Are you angry? Are you angry with us? With me?" He refuses to say anything. After a while he looks anxiously at both his parents before asking "Is what happened to Elijah going to happen to me?" Sybil angrily accuses the team of upsetting her son but calms down when she notices that Amos appears eager to talk. Pretty comes quietly into the room accompanied by one of the team, and John asks her to play outside. The team gently asks John's permission to invite her to join them. They answer Amos' questions until he fully understands that his illness will not improve and that he will receive all necessary care until he dies, emphasising how Elijah's care was different. One of the team asks "what do I need to know about you right now so that I can best help you?" Amos cries and responds that he wants his bicycle with him when he dies, the same one that Elijah used to ride, so that the brothers can still enjoy it together.

Over the next few months the team assists Sybil, John and Pretty to help Elijah find a way to achieve his dream. Pretty offers to draw a picture of the bicycle that Amos can take with him when he meets Elijah, and Amos agrees this is perhaps the answer. John promises Amos that he will continue to maintain the bicycle in perfect order after he has died and Amos gives permission for Pretty to 'inherit' his prized possession. Amos dies peacefully at home some months later surrounded by all his family and his bicycle.

Question 1. What do we mean by anticipatory grief and bereavement?

Both anticipatory grief and bereavement of children demand honesty, reassurance and safe expression of emotions. Death shatters a child's illusion that the world is safe and predictable, where bad things happen only to bad people, where if you are good there is reward, and that a God can make things better. After disclosure of a child's life-threatening illness by a clinician, one of the hardest things a parent will ever need to do is continue this discussion with their unwell child and other children in the family. This disclosure and conversation trigger *anticipatory grief*, an important and natural process that occurs during the rest of the child's life, and which prepares him and the family for dying and death. After death, the experiences of grief and the process of bereavement heal those that mourn the deceased child. *Bereavement* is a process whereby the bereaved find ways to take the deceased with them into their futures 'not moving on without him, but a moving forward with him' (McInerny 2019, Klass and Walter 2001) to ensure he is never forgotten and that future children and grandchildren will 'know' him. Bereavement moreover helps in coming to terms with the reality of a painful world, and is a valuable survival skill.

Question 2. How do we manage disclosure to Amos that he is dying?

How to manage disclosure to a child such as Amos, of his life-threatening diagnosis is often informed by theories of developmental psychology (Fredman 1997 p.127 and McKissock 1998 p.19-29). Young children are often thought to be lacking in knowledge in this area, and most adults will go to great lengths to prevent and protect children and young people of all ages suffering either physical or emotional distress. This often results in parents not wanting, or not knowing how to share the information. Although child developmental understandings of death are useful guides, they may ignore culture, religion, race, class and gender, and may make assumptions about what a child or young person may be experiencing.

Literature on dying and death generally encourages an honest explanation about the diagnosis and for family members to talk about the impending death rather than indulge in 'mutual pretence', a co-created structure designed to protect but that denies the dying process, 'as if' Amos will grow into an adult. This is often associated with a poor dying outcome, and complicated bereavement (Kreicsberg et al. 2004, Jalmsell 2015). At the same time most adults find it very difficult to talk openly about death, hence a contradiction of 'must talk but can't talk' (Fredman 1997). Sybil exemplifies the parent that fears that talk about death kills hope; that it's dangerous as it might welcome or invite death, and that it might go away if we ignore it. While health professionals often describe patients and family members who are struggling to talk as 'in denial' after diagnosis, we suggest this is rather more a persisting for the time being with a version of the information that they can currently manage, rather than resisting the 'truth' (Fredman 1997). Children and young people may prefer to talk with friends rather than professionals at times. Failure to converse comfortably about death does not always reflect an inability to do so. Amos' parents may hold a strong belief that it is not good, right or appropriate at that time, and possibly hold strong views that Pretty is 'too young' to hear the conversation. This needs to be respected while sensitively working towards a full and honest disclosure where emotions can be safely expressed.

Question 3. How can we talk to Amos about what is happening to him?

Joining the child or young person’s language will enable them to describe what they are experiencing, rather than have others attempt to describe their emotions for them, as in Amos being asked “are you angry?” Simple, clear questions can explore what matters to Amos right now (the bicycle for example.) It is vital furthermore to acknowledge Amos and his family’s expertise of their own lives and experiences by exploring who knows what, and who wants who to know what? This family is an obvious illustration of a family’s previous confrontation with the death of a child and the grief that was associated with that. Resilience and coping strategies of all family members will constantly change and grow, but always an inquiry into their knowledge and experience will provide guidance for the palliative care team. For example it may be helpful to ask Sybil “What would you want to protect Amos from?” “What does Amos need to know so that he can feel protected?” “Do you think Amos is trying to protect anyone?”

Question 4. How can we support the family in their loss of future dreams and help them focus on what is happening now?

Childhood development presupposes a future for Amos, comprising dreams, goals and fantasies of what a child or young person may become, so this diagnosis presents confusion for all the family as to their roles and relationships with a child who will not reach adulthood. As difficult as it may be, Pretty and her parents need to move away from a future framework and provide a present focus for interaction while Amos lives. Children such as Amos, who have witnessed the deaths of others from the same or similar conditions (as in HIV or inherited conditions), may have specific fears about dying driven by their personal experience. Recognition and accommodation of these fears is a prerequisite before providing reassurance that all that can be done will be done throughout the illness progression, including living as normally as possible. This includes school attendance and play with peers where possible.

Family stability can be maintained as far as possible by continuing routines, clear rules and boundaries (McKissock 1998). Integral to this is creating a safe environment in which the child feels free to express emotions and talk about preoccupations, with those around able to explore different possible meanings. For example when Amos asks “Is what happened to Elijah going to happen to me?” the team member can respond: “Are you asking if your illness is the same as Elijah’s? Are you also asking if you will die in the same way as Elijah?” Responses to these kinds of questions can clarify meanings as well as generate new questions and guide the team and family towards exploring aspects of anticipatory grief and meeting the needs of the child. Siblings may feel invisible, neglected or abandoned. When we meet Pretty she is on her own, with her own thoughts and feelings and no adult actually knew where she was. Some siblings may feel angry that the sick child receives so much attention, followed by intense guilt for acknowledging these feelings.

Question 5. What losses might Amos be grieving?

Dying children and adolescents such as Amos may grieve many losses during this time. There may be the immediate shift in health and independence, attractiveness and inclusion in the lives of their peers. Longer term dreams of future romances, sexual experiences, career, travel, marriage and children may also be identified with sensitive inquiry and support (Amery 2016). By providing a context of safety and acceptance a dying child such as Amos can use his anticipatory grief to construct a uniquely individualised knowledge about his death in relationship with his family, culture, community and hospital. It is helpful during this time for the patient and family to consciously capture memories through development of memory books and boxes, special mementoes, and sharing of important conversations. Legacy-making interventions can be powerfully healing, such as Pretty giving Amos a drawing of his bicycle and Amos leaving his bicycle to Pretty as a special inheritance (Akard et al, 2015).

> *When Amos dies the family follow their cultural tradition of burying their son next to his brother in the family burial plot. Because Pretty has been able to spend many hours talking and drawing with Amos while he was unwell, she knows that he wanted her picture of his bicycle to be buried with him, and the family fulfils this promise. Sybil notices that Pretty sits at the grave almost every day, and hears her crying and talking to Elijah and Amos. Sybil wonders if this is normal but when she asks Pretty how she is doing, her daughter refuses to talk, sometimes claiming she has nothing to say and at other times she runs off and climbs a tree. There are also times when Pretty cannot sleep and she seems increasingly anxious. She insists on sleeping in her parents' bed. At times she refuses to leave the house and John has heard from her teachers that she has been reprimanded at school for fighting with her friends.*
>
> *Several months after Amos died John feels depressed and indicates that he finds life meaningless, and sometimes thinks of killing himself. He and Sybil argue a lot and do not mention their sons' names at all. Sybil continues to work with difficulty but feels isolated and alone with her grief, finding it hard to put her agony into words. Pretty becomes increasingly angry and in an outburst, blames her parents for giving this illness to both her brothers. She says she wishes she could die.*

Question 6. What emotions might Amos' family be experiencing?

A child's death feels wrong, out of time, and leads to a liminal space, without anchor, floating 'in between' anything that has been previously known (Carter, 2017). Members of a grieving family will experience a wide range of emotions, experiences, thoughts, physical symptoms, social and spiritual changes and other reactions over a long period of time, at differing times and with varying intensities and combinations. In a palliative care context, exposure to the dying process provides valuable preparation for bereavement, with the patient, parents and siblings encouraged to benefit from the healing power of conversations. This can promote resilience and help to prevent post-traumatic stress (Lichtenthal et al 2015a, Weaver et al 2015, Wiener et al 2018, Jaaniste et al 2017, Waugh et al 2018). Involvement in this process may result in fewer regrets, knowing that family members have done all they can. When a child dies of a life-threatening illness, as in the case of Amos, the family has the chance to discuss and achieve goals, enjoy living to the best of their ability and to say goodbye (Wallin, 2019).

Many emotions are experienced in bereavement, not in any particular order, some repeatedly and others a sharp and sudden insight. Grief and its reactions are not completed in stages, but rather as a lengthy, tumultuous roller coaster. While it is commonly believed that the hardest part of grief is when the death occurs, it can take some weeks and months before the body and mind allows the bereaved to fully absorb the reality of the loss. It may be confusing and frightening for Sybil, John and Pretty to feel a lot worse about 6 months after Amos died, despite their anticipatory grief during his illness. It can be some months after the loss that true despair sets in, a reactive and constructive depression to the loss of a loved one, when there is reflection on what the future holds in the context of unbearable pain. It is usual for the bereaved to allude to or consider suicide at times; this is a measure of how painful grief is for them, and it helps to be able to share these thoughts without judgment or panic.

Question 7. What are the specific challenges for 10 year old Pretty?

For Pretty, there are many important aspects to consider when helping her manage her reactions. Bereaved children in particular need simple, factual information to explain what is happening. They need to be listened to with empathy, permission and facilitation to grieve in their unique way, assurance that there is no right or wrong way to grieve, and they need to be given time and support for feelings to be shared or expressed. A simple recognition and explanation of the many emotions experienced, including sadness, may help bereaved children to construct for themselves, their emotional reactions to and the meaning of this life-changing event (Weaver et al 2018).

Grief reactions of children such as Pretty, will change over time and maturity, and will vary according to gender, culture, age, previous losses and experiences, family support, the relationship to the deceased and the nature of the death. It may surprise and confuse parents how quickly young children may switch between emotions, one moment crying for the deceased to return, followed rapidly by laughter and play with friends. Indeed, an initial response may be startlingly practical. Perhaps Pretty asked “can I have his bicycle? Who will have his toys? Will I still be able to go to school?” This basic need to ensure the child’s own continuity possibly assists in making the environment safe enough to allow grief to occur.

Fear is commonly experienced by a grieving child. The death may raise concerns that they caused their loved one to die, (perhaps having had a fight or said things in anger such as 'I wish you would die'), that they too may die, or someone they love may die. Fear may manifest in regression to earlier behaviours such as becoming clingy, bedwetting or thumb-sucking which require understanding and sensitive management. Checking on the whereabouts of family members, insisting on not sleeping alone, and not wanting to go to school may all indicate separation anxiety.

Sadness is inevitable in grief. Crying can help and afterwards the child may feel tired but peaceful. Modelling of sadness by parents and adults is necessary to normalise this reaction and validate that they too are important and would also be missed if they died. Sadness says 'I hurt" and when it's expressed, elicits compassion, comfort and empathy. Watching sad movies or reading a sad book during their bereavement may cause more crying than usual, and provides 'an excuse' for more emotional release. When expression of sadness is difficult or restricted however, children may show physical, behavioural or social symptoms, such as anger and aggression. Sybil's sense of being unable to share her grief with her family may mean Amos is seldom talked of. Sybil may find it hard to know how to help Pretty and may hope that as a child she will 'just get on with things'. Sybil's lack of engagement with Pretty and inability for the moment to demonstrate healthy grieving will make it difficult for Pretty to process the impact of Amos' death on her family. One way she can express her pain and confusion will probably be in her behaviour and angry outbursts. Anger can often be understood as a means of pleading for the situation or the pain to stop, and enquiring as to whether this is what Pretty is feeling, may open up further conversations. In this case we hear that Pretty has been fighting with her school friends. Children, like adults, tend to move away, shut down or become withdrawn when angry to avoid hurting self and others. Understanding this behaviour assists adults in helping bereaved families to distinguish between anger and aggression, and to find safe ways to express the pain of grief. "Grief is an explanation, not an excuse" (McKissock 1998; 149).

Bereaved children may gain understanding through play, enacting death scenes, burial and digging up items to check what's happened to a 'body'. They may cover themselves in blankets and pillows to experience what it may feel like to be buried. In cultures where cremation is common, preoccupation with burning, what's left in ashes and imagining what happens to the body can be expressed and worked through by drawing or enacting relevant scenes. They may experience vivid dreams and nightmares. Ask them to describe/draw what they remember, what feelings they are left with upon waking and what they understand by them. The family belief system needs to be addressed by exploring the impact of grief upon it. It may be helpful to ask "How does this death change what you believe about life and death?" Integrating spiritual conversations into the bereavement process is known to positively assist bereaved siblings (Lövgren et al 2017, Eilertson et al 2018).

Guilt is commonly experienced during this process, by parents and siblings, for a variety of reasons either real or perceived. Amos was the second child in this family to die from this kind of cancer, although this cancer is seldom an inherited condition, and despite different treatment interventions. It will be helpful for the family to explore their perspectives of guilt, powerful or magical thinking to validate their emotions and help them uncover at their own pace the reassurance that this was not their fault. It is generally unhelpful however to simply repeat that they are not guilty when this is what they feel. Where death results from a non-communicable disease siblings may need to be reassured that the illness is not contagious.

To talk or not to talk about the deceased is an individual choice that the child can make in a context where sharing about the deceased is permitted, encouraged and respected. It is relevant to explain to a bereaved child that not mentioning the deceased is to deny his existence, as though he never mattered (Fredman 1997). As we all carry different messages about talking depending on contexts, relationships, timing and culture this will vary over time and situation. Care should therefore be taken not to label a child's expression of grief according to an adult's expectation of what grief should look like.

Question 8. What support can we give to Pretty to enable her to grieve healthily?

There are many activities that can ensure a safe and nurturing environment for a child to grieve healthily. Where possible, display photographs and memorabilia of the deceased, allocating these to siblings as desired. Inheriting Amos' bicycle will be a tangible connection between Pretty and both her deceased brothers, something of theirs that she can take into the future. Memory books and boxes that contain special messages, belongings, anecdotes and pictures help to maintain the relationship with deceased after death (Akard et al 2015). Special celebrations need to be acknowledged by all in the family, together with events that mark important milestones, memories, future plans and goals.

Parents and other significant adults need to take the initiative to include children of all ages in developing rituals that help to make sense of their loss. By so doing, children such as Pretty feel valued and integrated in the changed family (McKissock 1998). Through a process of sensitive curiosity and compassionate support, children and others in the family can be helped to revisit old selves and evolve new selves by rewriting their personal stories to answer questions such as "Who am I now? Am I still a sister?" These opportunities can also act as active preparation for future death events, including their own. Family sharing helps coordinate memories that enable the bereaved Pretty to create a context from which she can carry on, by incorporating her deceased siblings (Wallin, 2019). The bereaved Pretty may find that well informed teachers and the opportunity to interact with peers when returning to school helps maintain continuity and routine (Howard Sharp KM et al 2018). Where available bereaved siblings and parents can benefit from the camaraderie and sharing that is offered in support groups (Lichtenthal, 2015b).

Conclusion

A child's or young person's experience of loss, whether of their own future life or in response to the death of a loved one, is painful on many levels. Recognising that the resulting grief reactions are a healthy and necessary response in order to heal over time is an important step for everyone to take. By honouring the uniqueness of each individual through listening without judgment, providing honest communication and reassurance, children and young people can be enabled to express their grief as they need to over time.

References:

Akard TF, Dietrich MS, Friedman DL et al (2015) Digital storytelling: an innovative legacy-making intervention for children with cancer. Pediatr Blood Cancer 62(4):658-665.

Amery J (2016) A Really Practical Handbook of Children's Palliative Care. Lulu Publishing Services, London

Bluebond-Langner M (1978) The Private Worlds of Dying Children. Princeton University Press, Princeton

Carter BS (2017) Liminality in Paediatric Palliative Care. Am J Hosp Palliat Care 34(4):297-300.

Eilertsen MB, Lövgren M, Wallin AE et al (2018) Cancer-bereaved siblings' positive and negative memories and experiences of illness and death: A nationwide follow-up. Palliat Support Care 16(4):406-413.

Fredman G (1997) Death Talk. Conversations with Children and Families. Karnac Books, London

Howard Sharp KM, Russell C, Keim M et al (2018) Grief and growth in bereaved siblings: Interactions between different sources of social support. Sch Psychol Q 33(3):363-371.

Jaaniste T, Coombs S, Donnelly TJ et al (2017) Risk and Resilience Factors Related to Parental Bereavement Following the Death of a Child with a Life-Limiting Condition. Children (Basel).

Jalmsell L, Kontio T, Stein M et al (2015) On the Child's Own Initiative: Parents Communicate with Their Dying Child About Death. Death Stud 39(1-5):111-117.

Klass D, Walter T (2001) Processes of Grieving: How Bonds are Continued. In: Stroebe M et al (eds) Handbook of Bereavement Research. American Psychological Association, Washington DC

Kreicbergs U, Valdimarsdóttir U, Onelöv E, et al (2004) Talking about death with children who have severe malignant disease. N Engl J Med 16;351(12):1175-1186.

Lichtenthal WG, Sweeney CR, Robert KE et al (2015a) Bereavement follow-up after the death of a child as a standard of care in Paediatric Oncology, Pediatr Blood Cancer 62(Suppl 5):S834-S869.

Lichtenthal WG, Corner GW, Sweeney CR et al (2015b) Mental health services for parents who lost a child to cancer: If we build them, will they come? J Clin Oncol 33(20):2246-2253.

Ljungman L, Hovén E, Ljungman G et al (2015) Does time heal all wounds? A longitudinal study of the development of posttraumatic stress symptoms in parents of survivors of childhood cancer and bereaved parents. Psychooncology 24(12):1792-1798.

Ljungman L, Boger M, Ander M et al (2016) Impressions That Last: Particularly Negative and Positive Experiences Reported by Parents Five Years after the End of a Child's Successful Cancer Treatment or Death. PLoSOne. 11(6) e0157076

Lövgren M, Sveen J, Steineck G et al (2019) Spirituality and religious coping are related to cancer-bereaved siblings' long-term grief. Palliat Support Care 17:138-142. https://doi.org/10.1017/ S1478951517001146

McInerny N (2019) 'We don't move on with grief; we move forward with it'. TED Women 2018. Accessed 13 Apr 2019

McKissock D (1998) The Grief of our Children. ABC Books, Sydney

Snaman JM, Kaye EC, Torres C et al (2016) Helping parents live with the hole in their heart: The role of health care providers and institutions in the bereaved parents' grief journeys. Cancer 1;122(17):2757-2765.

Vegsund HK, Reinfjell T, Moksnes UK et al (2019) Resilience as a predictive factor towards a healthy adjustment to grief after the loss of a child to cancer. PLoS One. 14(3): e0214138

Wallin AE, Udo C, Kreicbergs U et al (2019) Cancer-bereaved siblings' advice to peers - A nationwide follow-up survey. Death Stud.

Waugh A, Kiemle G, Slade P (2018) What aspects of post-traumatic growth are experienced by bereaved parents? A systematic review. Eur J Psychotraumatol. 9(1):1506230.

Weaver MS, Heinze KE, Kelly KP et al (2015) Palliative Care as a Standard of Care in Paediatric Oncology. Pediatr Blood Cancer 62(Suppl 5): S829–S833.

Weaver MS, Lichtenthal WG, Larson K, Wiener L (2019) How I approach expressing condolences and longitudinal remembering to a family after the death of a child. Pediatr Blood Cancer. 66(2):e27489

Wiener L, Rosenberg AR, Lichtenthal WG, Tager J, Weaver MS (2018) Personalized and yet standardized: An informed approach to the integration of bereavement care in paediatric oncology settings. Palliat Support Care, 16(6):706-711.

Wikman A, Mattsson E, von Essen L et al (2018) Prevalence and predictors of symptoms of anxiety and depression, and comorbid symptoms of distress in parents of childhood cancer survivors and bereaved parents five years after end of treatment or a child's death. Acta Oncol 57(7):950-957.

Perceptions of paediatric liaison teams on the Belgian Euthanasia Law extended to minors

This article was published under the title: What impact has the Belgian euthanasia law extended to minors in 2014 on our practice? Perspectives from Belgian paediatric palliative care teams. Friedel M, Terwangne B, Brichard B, Ruysseveldt I, Renard M. Int J Palliat Nurs. 2018;24(7):333-337. doi: 10.12968/ijpn.2018.24.7.333.

Discrepancy between clinical practice of paediatric palliative care and the Belgian Euthanasia Act extended to minors in 2014

In 25 years of clinical practice, Belgian paediatric palliative care teams express they were never confronted to a child requesting euthanasia (1). If parental requests of euthanasia were observed, they were often caused by intense despair or a feeling of being overburdened. Once the intense physical pain of the child was managed and the parent's fears listened to, those requests generally disappeared.

However, an existing euthanasia law for adults was extended in February 2014 to minors. Values such as compassion, humanity and auto determinism were raised by the defenders to justify the need to extend the law. Interestingly, no restriction of age was specified, placing Belgium as the only state in the world allowing euthanasia for minors, apart from the Netherlands where euthanasia is possible from 12 years on. This law decriminalizes, under very strict conditions, euthanasia, defined in Belgium as the "medical act to intentionally end life, at patients request" (2). These conditions are namely: the child must suffer from an incurable disease, death expectancy must be near, he must be in a state of constant suffering, experiencing unbearable physical pain, his capacity must have been assessed by a psychologist or psychiatrist and both parents must have given their consent to it. Two independent physicians must assess that all criteria are fulfilled (3). Excluded from those conditions are proxy-requests (parental requests), neonates and severely cognitive impaired children who wouldn't have a capacity-making to make a request for themselves (4).

Further analysis of the whole process which led to the law was reported by Friedel M. (5) and showed that the specialized Paediatric Palliative Care (PPC) teams, called in Belgium paediatric liaison teams (6) were not invited by the Senate to discuss the proposition of the law. Nevertheless, in the north Flemish part of Belgium, many discussions on the legal, ethical and psychological issues raised by the law were observed, whereas in the south-French-speaking part of Belgium, awareness seemed to be less present.

Since the law was enacted also for children, from 2014 to 2017, euthanasia was granted for 2 adolescents in the north part of Belgium. They had first benefitted from palliative care in their last life stage.

In Belgium, euthanasia and palliative care are considered by some authors as mutually complementary, synthesised even as the "Belgian model of integral end-of-life care"(7,8).

For them, a rare request for euthanasia could be made possible, even for a child, although good palliative care has been provided, recognizing the limits of palliative care in some cases and giving a voice to a child's choice. Additionally, nurses may believe the framework provided by the euthanasia law would eventually prevent abuses made by physicians in ending lives without the patient's request and moreover the law would trigger patients, families and health care teams to discuss in a more open way sensitive topics such as unbearable suffering, despair and fear of dying. Cultural differences in perceptions and practices are also recognized in the White Paper from the European Association of Palliative Care (EAPC), which argue that "the provision of euthanasia and physician-assisted suicide should not be included into the practice of palliative care" (9). The same arguments were argued by the International Association of Palliative Care and Hospice (IAPCH) in 2017: "no country or state should consider the legalization of euthanasia or Physician-assisted suicide until it ensures universal access to palliative care services and to appropriate medications (10)." This statement does not specify if it applies only for adults.

Powerlessness and hope

At the contrary, very often, PPC liaison teams are facing situations where parents ask medical teams to prolong their child's life, to desperately try other types of active curative treatments, even if those are not recognized by the official protocols, are expensive and sometimes offered in a country which is at the opposite side of the world. Facing the death of a son/daughter is certainly the most difficult situation experienced by a parent. The legitimate request addressed to physicians to prolong the life of their beloved or to end his life might be linked to hope or powerlessness. Recent studies on the important role of hope in paediatric palliative care, give us pathways on how it can be maintain throughout the tough parental journey. The objective is to shift smoothly, in a compassionate and subtle way, the hope to cure to the hope to be cared for adequately (11-15).

This is in line with Waldman and Wolfe (16), who suggests 5 cardinal questions to enhance communication between physician and parents and to explore the domains of parental hope: "Tell us about your child? What is your understanding of your child's illness? What are you hoping for? What are your worries? Where do you find your strength?"

Taking the time to listen to parents and children and exploring with them meaning-making, might reduce their intense suffering.

The inherent paradox in paediatric palliative care: promoting quality of life in the face of death

The World Health Organization states that the principal aim of PPC is to improve quality of life of the child and of the family members (17). This may seem paradoxical: promoting quality of life while death is approaching. Denis Vasse distils this idea in the following nutshell: "I want to live but I will die."(18) Quality PPC is offered moment by moment, never assuming that children and their families will remain constant in their desire for the child to live. Relations are made of complex interactions where professionals can only represent "one side" and do their best to accompany the difficult journey of those children and their families.

Nevertheless, several studies do demonstrate that children's quality of life was improved through PPC (19-22). Those results were achieved through different interventions: excellent pain and symptom management, good communication with the child and the family members, continuity and coordination of care, availability 24/7 and offering respite care services (23).

Management of complex pain situations

Belgian PPC liaison teams recognize that pain and other refractory symptoms are not completely managed for all children. Studies confirm that in some situations -mostly for children with advanced cancer-, pain may not always be under control (24).

In Belgium, management of complex pain symptoms is discussed during interdisciplinary weekly rounds, and several treatment options shared with parents and the child. In some case this results in palliative sedation, when all others pain medications failed. For other children, withholding or withdrawal of treatments is leading to death.

Law defenders argue that professionals should accept the limits of paediatric palliative care and recognizing euthanasia as a possible end-of life decision if unbearable, constant suffering cannot be managed.

Relational autonomy and involvement of children in decision-making

In our society, the concept of autonomy, in which personal choice are highly valued, determines "moral agents as rational, independent, self-sufficient decision-makers"(25). The Belgian euthanasia law relies on this perception. At, the contrary, other perspectives (26) focus on relational autonomy and "understands moral agents as rational, emotional, creative and interdependent (27)".

In this last perspective, both children and parents are influenced during the illness. Parents are influenced by what the physician tells them about the illness and the options of treatment, advanced care planning or end-of life care options (28). Children are influenced by their parent's experience, or expectations towards them and their desire to keep their love (29).

"Choices are only choices if they are really choices" (30). In the perspective of relational autonomy, access and quality of paediatric palliative care depends on several factors, namely: PPC education of caregivers, early referrals to PPC teams, social acceptance of PPC services, and adequate symptom management. Dussel underpins the risk that insufficient symptom management alone could lead to parental request for hastening death of their child (31).

The absence of any of these may diminish choice.

Children should be involved in decision-making in an age-appropriate and sensitive way. This is underlined by the Charter of Trieste in 2014 (32,33). Guidance (34) exists on how to initiate difficult discussions, particularly on advanced care planning (35,36), but little is known nor used in Belgium. However, advanced care planning might improve end of life care and reduce euthanasia requests.

An euthanasia procedure might represent an additional burden at end-of-life, at a stage where many emotions are already difficult to manage. Furthermore, it is not known how a this procedure might impact parental grief. It is very common that children in a palliative context tend to protect their parents. The Belgian euthanasia procedure, in which children have to express their request, might disrupt those relations, who are often very strong, loyal or even fusional.

Protecting one another, called as mutual pretense, might be in opposition with the euthanasia procedure. Bluebond-Langner defines “mutual pretense, where each party in the interaction knows what is going to happen, but does not acknowledge it openly to one another, and becomes the dominant mode of interaction between parents and children with life-limiting illnesses, especially when cure is not likely ”(37). This concept was first described by Glaser and Strauss under the name “level of awareness of death” (38).

Paediatric Palliative care needs and provision in Belgium

Whereas Fraser (39) found in the UK, the prevalence of children living with life-limiting conditions being 32/10.000, no similar prevalence is known for the Belgian paediatric population.

However, national statistics on mortality shows that, for the year 2014, 796 children and young adults (from 0-19 years) died, from which 26% (n=209) of those died from congenital abnormalities or neuro-metabolic diseases and 8 % (n=61) from cancer (40).

Furthermore, a retrospective study conducted by Friedel et al. documented for the first time the number of 700 children cared for each year by the paediatric liaison teams. On an annual basis, 85 children died (12%), and for half of them in their own home (41). Additionally, this study pointed some barriers to access PPC suggesting there is room for improvement. First, governmental funding of PPC services is partially lacking, obliging PPC teams to dedicate time for fundraising. Second, although PPC competences are much needed for home care nurses, courses couldn’t be launched due to insufficient number of applicants (42). Third, the term palliative care is commonly understood restrictively as end-of life care, which frightens people and hinders them to access palliative care.

In light of these facts, doubts might be raised about societal awareness and adequate provision of PPC in Belgium.

Nevertheless, an excellent initiative has been the creation of the Belgian Paediatric Palliative Care Group, assembling since 2012, more than 70 PPC actors from all over the country to collaborate in the development of guidelines for PPC in Belgium (43,44). Moreover, a website sharing PPC information was created in 2016 (45). Perhaps most significant has been the inclusion of a PPC representative in the Belgian Federal Commission responsible for the evaluation of palliative care which has an advisory function to the Belgian Federal Ministry of Health (46).

Conclusion

Since the euthanasia law was extended to minors, PPC liaison teams must be prepared to deal in the future with potential euthanasia requests expressed by children suffering unbearable suffering at end of life. This new policy triggers PPC teams to engage with children and their parents open conversations about end-of-life issues, but also to assess quality of care provided and to improve societal awareness about the added value PPC may offer.

It is a societal and especially a health profession issue to prevent situations where a child or a young adult would experience a situation leading him to request euthanasia.

A metaphor illustrating this challenge could be the choreography of the German artist Pina Bausch entitled “Café Müller” in which a dancer – with her eyes closed- slowly walks, without seeing chairs scattered all over the room. Another dancer carefully removes the chairs, one after another with the aim to free the passage and prevent her falling. Maybe paediatric palliative care aims to do something similar? Trying by all means, early enough, and step by step, through a multidisciplinary team, in a holistic way, centred on both the experience of the child and his family, to take away the multiple obstacles such as intractable pain, despair, loneliness, in the aim to free the passage.

References

1. Renard M, Ruysseveldt I (2017) l’euthanasie pour les mineurs d’âge. L’extension de la loi relative à l’euthanasie a-t-elle apportée des changements ? Percentile; 22(4):20-23.
2. Belgian law on euthanasia in 2002 which was extended to minors in 2014: http://www.ejustice.just.fgov.be/cgi_loi/change_lg.pl?language=fr&la=F&cn=2002052837&table_name=loi (accessed on 23th November 2016)
3. Dan B, Fonteyne C, Clément de Cléty S (2014) Self-requested euthanasia for children in Belgium. Lancet 22;383(9918):671-2.
4. Clément de Cléty S, Friedel M, Verhagen AA, Lantos JD, Carter BS (2016) Please Do Whatever It Takes to End Our Daughter's Suffering! Paediatrics Jan;137(1).
5. Friedel M (2014) Does the Belgian law legalising euthanasia for minors really address the needs of life-limited children? Int J Palliat Nurs ;20(6):265-7.

6. Royal Decree on paediatric liaison teams http://www.etaamb.be/fr/arrete-royal-du-15-novembre-2010_n2010024459.html
(accessed on 23th November 2016)
7. Bernheim JL, Raus K (2017) Euthanasia embedded in palliative care. Responses to essentialistic criticisms of the Belgian model of integral end-of-life care. J Med Ethics. Jan 6. pii: medethics-2016-103511. doi: 10.1136/medethics-2016-103511.
8. Bernheim JL, Distelmans W, Mullie A, Ashby MA (2014)Questions and answers on the Belgian model of integral end-of-life care: experiment? Prototype? : "Eu-euthanasia": the close historical, and evidently synergistic, relationship between palliative care and euthanasia in Belgium: an interview with a doctor involved in the early development of both and two of his successors. J Bioeth Inq ;11(4):507-29.
9. Radbruch L, Leget C, Bahr P, Müller-Busch C, Ellershaw J, Conno de F, Vanden Berghe P (2015) Euthanasia and Physician-assisted suicide: A white paper from the European Association for Palliative Care. Palliative Medicine
10. De Lima L, Woodruff R, Pettus K, et al.: International association for hospice and palliative care position statement: Euthanasia and physician-assisted suicide. J Palliat Med 2017;20:8–14.
11. Rosenberg A, Feudtner C (2016) What else are you hoping for? Fostering hope in paediatric serious illness. Acta Paediatr ;105(9):1004-5.
12. Hill D, Miller V, Walter J, Carroll K, Morrison W, Munson D, Kang T, Hinds P, Feudtner C. (2014) Regoaling: a conceptual model of how parents of children with serious illness change medical care goals. BMC Palliat Care; 13;13(1):9.
13. Feudtner C, Carroll K, Hexem K, Silberman J, Kang I, Kazak A (2010) Parental Hopeful Patterns of Thinking, Emotions, and PPC Decision-making: A Prospective Cohort Study. Arch Pediatr Adolesc Med; 164(9):831-9.
14. Feudtner C (2009)The Breadth of Hopes. N Engl J Med ; 361:2306-2307.
15. Samson A, Tomiak E, Dimillo J, Lavigne R, Miles S, Choquette M, Chakraborty P, Jacob P (2009)The lived experience of hope among parents of a child with Duchenne muscular dystrophy: perceiving the human being beyond the illness. Chronic Illn ;5(2):103-14.
16. Waldman E, Wolfe J (2013) Palliative Care for children with Cancer. Nat Rev Clin Oncol ;10(2):100-7.
17. WHO 1998 Definition of Palliative Care for children : http://www.who.int/cancer/palliative/definition/en/ (accessed on 24th November 2016)
18. Vasse D. (2008) Le poids du réel, la souffrance, Ed Du Seuil.

19. Groh G, Borasio G, Nickolay C, Bender H, von Luttichau I, Fuhrer M (2013) Specialized paediatric palliative home care: a prospective evaluation. J Palliat Med ; 16(12):1588-1594.
20. O'Quinn L, Giambra B (2014), Evidence of improved quality of life with paediatric palliative care. Pediatr Nurs; 40(6):284-288, 296.
21. Schmidt P, Otto M, Hechler T, Metzing S, Wolfe J, Zernikow B (2013) Did increased availability of paediatric palliative care lead to improved palliative care outcomes in children with cancer? J Palliat Med ; 16(9):1034-1039.
22. Vollenbroich R, Duroux A, Grasser M, Brandstatter M, Borasio G, Führer M (2012) Effectiveness of a paediatric palliative home care team as experienced by parents and health care professionals. J Palliat Med; 15(3):294-300.
23. In Belgium 3 paediatric respite care services are funded by national health insurance: Villa Rozerood, Villa Limerick and Villa Indigo.
24. Wolfe J, Orellana L, Ullrich C (2015) Symptoms and Distress in Children With Advanced Cancer: Prospective Patient-Reported Outcomes From the PediQUEST Study. J Clin Oncol ; 10;33(17):1928-35.
25. Walter J, Ross L (2014) Relational autonomy: moving beyond the limits of isolated individualism. Paediatrics;133 Suppl 1:S16-23.
26. Ells C,Hunt M, Chambers-Evans J (2011) Relational autonomy as an essential component of patient-centered care. International Journal of Feminist Approaches to Bioethics; 4(2):79-101.
27. Haward M, Murphy R, Lorenz J (2012) Default options and neonatal resuscitation decisions. Journal of Medical Ethics; 38(12):713-718.
28. Bluebond-Langner M (1978) The private worlds of dying children. Princetown University Press.
29. Hinds PS, Schum L, Baker J, Wolfe J (2005) Key factors affecting dying children and their families. Journal of Palliative Medicine;8(Suppl. 1):70–78.
30. Baker J, 3rd Maruzza Conference on Paediatric Palliative Care. 16-19 Nov 2016. Rome.
31. Dussel V (2010) Considerations about hastening death among parents of children who die of cancer. Arch Pediatr Adolesc Med;164(3): 231-237.
32. Benini F, Vecchi R, Orzalesi M (2014) The rights of the dying child. The Lancet; 383(9928): 1547–1548.
33. Benini F, Vecchi R, Lazzarin P, Jankovic M, Orsi L, Manfredini L, Drigo P, Sellaroli V, Gangemi M, Spizzichino M, Orzalesi M (2016) The rights of the dying child and the duties of healthcare providers: the "Trieste Charter".Tumori; Oct 8:0. doi: 10.5301/tj.5000566. [Epub ahead of print]

34. Weaver M, Baker J, Gattuso J, Gibson D, Sykes A, Hinds P (2015) Adolescents' preferences for treatment decisional involvement during their cancer. Cancer ; 124(24):4416-4424.
35. Zadeh S, Pao M, Wiener L (2015) Opening end-of-life discussions: how to introduce Voicing My CHOiCES™, an advance care planning guide for adolescents and young adults. Palliative and Support Care; 13(3):591-599.
36. Wiener L, Zadeh S, Battles H, Baird K, Ballard E (2012) Allowing adolescents and young adults to plan their end-of-life care. Paediatrics ; 130(5):897-905.
37. Bluebond-Langner M, DeCicco A, Belasco J. Involving Children with Life-shortening Illnesses in Decisions About Participation in Clinical research: A proposal for shuttle
Diplomacy and Negotiation (Chapt. 18), in Kodish E. (ed). Ethics and Research with Children. Oxford University Press. 2005:333.
38. Glaser B, Strauss A (1965). Awareness of dying. Aldine Publishing Company.
39. Fraser, L.K., Miller, M., Hain, R., Norman, P., Aldridge, J., McKinney, P.A. & Parslow, R.C. (2012). Rising national prevalence of life-limiting conditions in children in England. Paediatrics, 129(4), 923-929.
40. Belgian Data Statistics. Statbel 2014.
41. Friedel M., Brichard B., Renard M., Fonteyne Ch., Vandecruys E., Misson J-P, Degryse J-M., Aujoulat I. Hidden part of the iceberg : Paediatric Palliative Care in Belgium. Poster presented during the symposium of the research network IN-supC, European Association of Palliative Care (EAPC), 30th sept 2016, Brussels, Belgium.
42. Friedel M. Botbol-Baum M., Brichard B.,Clément de Cléty S, Develter S., Jacquemin D. How does the Belgian law on child euthanasia impact a Belgian Paediatric Palliative Care Course? Oral Presentation at the 2nd Maruzza Congress on Paediatric Palliative Care. 19-21th November 2014, Rome.
43. Guidelines in 5 major domains are still elaborated: definition of PPC, Pain and symptom-management in PPC, Decision-making and advanced care planning in PPC, Organization of PPC in Belgium, end-of-life care and ethics.
44. Renard M, Brichard B, Francotte N, Fonteyne C, Misson J.-P, Faingnaert M, Mattheeuws K, Ruysseveldt I. Belgian Paediatric Palliative Care: Spreading the Ideas of Paediatric Palliative Care in the Different Fields of Paediatric Medicine. Poster presentation at 3rd Maruzza Congress on Paediatric Palliative Care, 16-19 November 2016.
45. http://www.palliatieve-zorg-en-kinderen.be/template.asp?f=start.htm (accessed the 5th October 2016)
46. http://www.soinspalliatifs.be/images/Rapport-CESP-2014-FR_FINAL.pdf (accessed the 5th October 2016)

Marie Friedel trained as a paediatric nurse (1996, Haute Ecole Léonard de Vinci, Belgium) and holds a Master in Public Health (2000, UCLouvain, Belgium) and a Diploma in acute/chronic Paediatric Pain and Paediatric Palliative Care (2016, University of Lyon I, France). She worked for several years in a mobile paediatric liaison team in Belgium, ensuring continuity of care for children with life-limiting conditions. Since 2000, she has been a Lecturer at a Nursing and Midwifery High School (Haute Ecole Léonard de Vinci, Belgium). Passionate about making paediatric palliative care more visible, she started a PhD in Public Health in 2016 at the Institute of Health and Society (IRSS), exploring children's quality of life in a family-centred context through a collaborative approach with Belgian paediatric liaison teams. Spouse and mother of 3 teenagers/young adults, she loves sowing seeds to build bridges between people, countries and disciplines.