

PEDIATRIC PALLIATIVE AND HOSPICE CARE AT HOME: DEVELOPMENT OF A MEASURE
OF FAMILY-REPORTED EXPERIENCES

Jackelyn Y. Boyden

A DISSERTATION

in

Nursing

Presented to the Faculties of the University of Pennsylvania

in

Partial Fulfillment of the Requirements for the

Degree of Doctor of Philosophy

2020

Supervisor of Dissertation

Mary Ersek PhD, RN, FPCN
Professor of Palliative Care

Graduate Group Chairperson

Margo Brooks Carthon, PhD, RN, FAAN
Associate Professor, School of Nursing

Dissertation Committee:

Janet A. Deatrck, PhD, RN, FAAN
Professor Emerita of Nursing

Chris Feudtner, MD, PhD, MPH
Professor of Pediatrics, Medical Ethics and Health Policy

Kimberley Widger, RN, PhD, CHPCN(C)
Associate Professor of Nursing

PEDIATRIC PALLIATIVE AND HOSPICE CARE AT HOME: DEVELOPMENT OF A MEASURE
OF FAMILY-REPORTED EXPERIENCES

COPYRIGHT

2020

Jackelyn Y. Boyden

DEDICATION

This dissertation is dedicated to the many children with serious illnesses and their families who I have had the incredible privilege to care for as a nurse, and for all the children and families who I have the great honor to learn from in my role as a nurse researcher -- your strength and courage have inspired me in so many ways.

ACKNOWLEDGMENT

As I sit down to write this acknowledgement, I am at a loss for where to begin. There are so many people without whom I would not be where I am today. First, I express my deepest gratitude to my dissertation committee – to Dr. Mary Ersek, my dissertation chair, who has supported, encouraged, and mentored me along every step of the past nearly 6 years, who has patiently edited so many papers and grant applications and answered so many questions, and whose lessons about research, career, and life I will carry with me for many years to come. To Dr. Janet Deatrick, who has been my mentor and supporter since the very early days in my program, who has encouraged and guided me, and who has inspired me with her passion for family-centered research. To Dr. Chris Feudtner, who has inspired me with his work in pediatric palliative care since my very early days as a PICU nurse, who has helped me open so many doors in the field of pediatric palliative care, who has pushed me to think critically and creatively about every problem, and who has never passed over an opportunity, however small, to teach and to mentor. To Dr. Kim Widger, who over two years has already taught me so much and inspired me with her important research in understanding and improving the quality of pediatric palliative care. I truly would not be where I am today without this incredible team of mentors.

I also thank my readers – Dr. Kathy Bowles, who served as a reader on my dissertation proposal and who supported me in writing my F31 proposal during dissertation seminar; Dr. Karen Lasater, who served as a reader on my dissertation; and Dr. George Demiris, who served as a reader on both my proposal and my dissertation. I also thank Dr. Martha Curley, who served as my chair on my qualifying examination. My heartfelt gratitude for their time and expertise they have so generously shared with me.

I also send my deepest gratitude to the members of the Justin Michael Ingerman Center for Palliative Care at CHOP. This incredible team of providers and researchers, whose work has been my inspiration from my days as a PICU nurse, and who continues to inspire me with their

compassion and dedication to their patients and families and with their passion for the very important work that they do every day. I express my appreciation to everyone on this incredible team, but a special thanks to Ms. Karen Crew, Ms. Gwenn LaRagione, Ms. Dana Dombrowski, Dr. Wynne Morrison, and Dr. Doug Hill for their invaluable support during my program.

Thank you to the Courageous Parents Network, and to Ms. Blyth Lord especially, for their generous support of my research. The work done by this network of parents, providers, and staff is truly inspirational, and I could not have completed my project as I did without their support and participation.

Thank you to the NINR, to Dr. Jeri Miller and the Office of End-of-Life and Palliative Care Research, and especially to Dr. David Banks, who have provided critical mentorship and support through my F31 Ruth L. Kirschstein National Research Service Award Fellowship (F31NR017554) and the development of my F32 post-doctoral fellowship proposal. I also thank the University of Pennsylvania School of Nursing's Office of Nursing Research, the Independence Blue Cross Foundation's Nurses for Tomorrow Scholars Program, and Sawtooth Software's Academic Grant for their support of my research.

I send a special thanks to all of my PhD colleagues and friends – it truly takes a village, and I could not have made it through this program without all of them! Thank you especially to Dr. Patty Pawlow and Dr. Jennifer Morone for being a part of this journey with me.

Thank you also to the Penn School of Nursing Palliative Care Scholars Group – I learned so much from this amazing group of dedicated and talented palliative care scholars!

To my family and friends – especially my parents, Georgina and Denny Ng, my siblings Brian and Claudia, and my in-laws Mark and Tina, who encouraged me and supported me through 6 (long!) years. To my mom especially, who on so many occasions dropped everything and flew up to

Philadelphia to care for my children so that I could get my work done. To my husband Brew, who has been my rock and my biggest cheerleader – for his love, encouragement, patience, and enthusiasm for everything (from my course assignments to my dissertation project!). He deserves every bit of recognition for this accomplishment – his constant support through a PhD program (and the birth of two babies during the program!) was invaluable. And to my beautiful daughters, Emi and Maggie, who have been my inspiration and my reason to keep pushing and who supported me with their love and patience, even when Mommy was “always doing work” these last few months. I hope that you will never lose your endless sense of curiosity, your love of learning new things, and your incredible creativity.

And finally, to all of the providers, researchers, and especially parents who participated in this project -- who, in the midst of their incredibly busy days, generously and graciously shared their time, energy, and experiences with me -- thank you for allowing me to learn from you and your families. I am deeply and truly grateful to all of you.

ABSTRACT

PEDIATRIC PALLIATIVE AND HOSPICE CARE AT HOME: DEVELOPMENT OF A MEASURE OF FAMILY-REPORTED EXPERIENCES

Jackelyn Y. Boyden

Mary Ersek

While children with serious life-threatening illnesses (SLTIs) are increasingly being cared for primarily at home by their families, an estimated 5,000 U.S. children are living within the last six months of their lives on any given day, many with significant, medically-complex care needs. Pediatric palliative and hospice care at home (PPHC@Home) plays a critical role in supporting children with SLTIs and their families. Yet, experts are concerned that these children and families are not receiving consistently high-quality PPHC@Home. Providing consistent, high-quality care that meets children's and families' most important needs and priorities requires the systematic measurement of care, although no previous family-reported experience instruments exist to specifically evaluate PPHC@Home in the U.S. Using a multi-method, multi-stakeholder approach, the purpose of this dissertation project was to develop a measure of families' experiences with PPHC@Home.

In **Aim 1**, I explored parents' priorities for PPHC@Home using a discrete choice experiment with parents whose children have received PPHC@Home. In **Aims 2 and 3**, I followed a phased instrument development process, including item selection, item prioritization and reduction, and cognitive interviewing. Starting with a pool of 100 items derived from existing outcomes instruments in PPC, standards of practice, and quality guidelines, I developed the EXPERIENCE@Home (**Ex**periences of **P**alliative and Hospice Care for **C**hildren and **C**aregivers at Home) Measure, which contains 22 of the highest-valued items, as rated by PPC providers, researchers, parent advocates, bereaved parents, and parents currently caring for their child. In

Aim 4, I propose next steps to evaluate the validity, reliability, acceptability, feasibility, and clinical utility of the EXPERIENCE@Home Measure from the perspective of parents and PPHC providers in a post-doctoral fellowship application.

Through this dissertation study, I have developed the first known instrument to measure families' experiences with PPHC@Home, and proposed next steps for the evaluation of this newly developed instrument. Only by measuring patient and family care experiences can we begin to understand how to improve the care we provide to children with SLTIs and their families living at home.

TABLE OF CONTENTS

ACKNOWLEDGMENT	IV
ABSTRACT	VII
LIST OF TABLES	XI
LIST OF FIGURES	XII
CHAPTER 1: GENERAL INTRODUCTION	1
BACKGROUND	2
PURPOSE, SPECIFIC AIMS, FRAMEWORK, AND METHODS	9
ORGANIZATION OF THIS DISSERTATION	13
SUMMARY AND SIGNIFICANCE	14
BIBLIOGRAPHY	16
TABLES AND FIGURES	22
CHAPTER 2: WHAT DO PARENTS VALUE REGARDING PEDIATRIC PALLIATIVE AND HOSPICE CARE IN THE HOME SETTING? (PAPER 1)	26
ABSTRACT	27
INTRODUCTION	30
METHODS	31
RESULTS	35
DISCUSSION	37
CONCLUSION	42
BIBLIOGRAPHY	44
TABLES AND FIGURES	50

CHAPTER 3: DEVELOPING A FAMILY-REPORTED MEASURE OF EXPERIENCES WITH HOME-BASED PEDIATRIC PALLIATIVE AND HOSPICE CARE: A MULTI-METHOD, MULTI-STAKEHOLDER APPROACH (PAPER 2)	62
ABSTRACT	63
BACKGROUND.....	64
METHODS AND RESULTS	65
DISCUSSION	75
BIBLIOGRAPHY	84
TABLES AND FIGURES.....	89
CHAPTER 4: EXPERIENCES OF PALLIATIVE AND HOSPICE CARE FOR CHILDREN AND CAREGIVERS AT HOME (EXPERIENCE@HOME) PROJECT (PAPER 3)	117
SPECIFIC AIMS	118
RESEARCH STRATEGY	120
PROTECTION OF HUMAN SUBJECTS	128
BIBLIOGRAPHY	136
TABLES AND FIGURES.....	142
APPENDIX	146
CHAPTER 5: SUMMARY AND FUTURE DIRECTIONS	156
SUMMARY OF FINDINGS	157
DISCUSSION	161
DIRECTIONS FOR FUTURE RESEARCH.....	167
CONCLUSION.....	168
BIBLIOGRAPHY	170

LIST OF TABLES

Chapter 1	Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home	Page 23
Chapter 2	Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care in the Home	Page 51
	Table 2: Demographic and Clinical Characteristics of Parents and Children	Page 53
	Table 3: Sub-group Domain Score Comparisons: 2-Sample Independent t-tests	Page 56
Chapter 3	Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home	Page 90
	Table 2: Characteristics of Professional Participants (Phase 2)	Page 92
	Table 3: Demographic and Clinical Characteristics of Parent Participants and Their Children (Phase 3)	Page 93
	Table 4: Items Prioritized by Parents (Phase 3)	Page 96
	Table 5: Final EXPERIENCE@Home Measure	Page 101
	Supplemental Table 1: Items Prioritized by PPHC Professionals (Phase 2)	Page 107
	Supplemental Table 2: Phase 4 -- Demographic and Clinical Characteristics of Parent Participants and Their Children	Page 115
Chapter 4	Table 1: Example EXPERIENCE@Home Measure Items and Weights	Page 143
	Table 2: Additional Study Measures	Page 144

LIST OF FIGURES

Chapter 1	Figure 1: EXPERIENCE@Home Measure Development Process	Page 25
Chapter 2	Figure 1: Average Importance Scores Across Domains	Page 60
	Figure 2: Individual-level Variation Across Domain Importance Scores	Page 61
Chapter 3	Figure 1: Item Selection and Reduction Process	Page 105
	Figure 2: Sample DCE Choice Set	Page 106
Chapter 4	Figure 1: Domains of Quality PPHC@Home	Page 145

CHAPTER 1: GENERAL INTRODUCTION

BACKGROUND

Approximately 13 to 18 percent of U.S. children have special health care needs.(1) A sub-group of these children are living with life-shortening serious illnesses, which may or may not have potential curative or life-prolonging treatments, but all-too-often result in death.(2) In part due to medical advancements over recent decades, these children are generally living longer (3-6), and many are primarily cared for at home by their families.(4-7) Yet, an estimated 5,000 U.S. children are living within the last six months of their lives on any given day (8), many with significant, medically-complex care needs (6, 9), particularly toward the end of life.

These children and families are often supported by pediatric palliative and hospice care in the home, hereafter referred to as PPHC@Home, which is “patient and family-centered care that optimizes quality of life by anticipating, preventing, and treating suffering” that can be provided by home hospice, home health care, or hospital-based pediatric palliative care (PPC) programs that also conduct home visits.(5) PPHC@Home is provided primarily outside of the hospital and integrates a complex network of home, community, outpatient, hospital, and hospice care providers across medical and social service settings and institutions.(5, 6, 10) While the total number of PPHC@Home providers is unknown, only approximately 30% of U.S. children’s hospitals offer any type of home visitation service.(11) Additionally, of the nearly 78% of surveyed hospice organizations who reportedly serve pediatric patients, only about 36.6% have a formal pediatric program in place.(5)

Disparities in PPHC@Home Quality and Outcomes

Children and families deserve high-quality PPHC@Home. The provision of PPHC@Home has been descriptively associated with numerous positive child and family outcomes, including improved child quality of life (12-17), greater concordance between preferred and actual location of death (12, 16, 17), family quality of life (14, 15), better pain and symptom management (13, 18, 19), and family satisfaction with care.(13, 15, 16, 20) Additionally, enrollment in a PPHC@Home program has been found to be associated with substantial reductions in hospital utilization.(14, 15, 21) One study found that children with non-cancer diagnoses enrolled in a PPHC@Home program had an average decreased length of hospital stay of 38 days.(21)

Experts are concerned, however, that children with serious illnesses and their families are not receiving consistently high-quality care.(10, 18, 22-24) No standard model for PPHC@Home exists in the U.S., and the composition of and services provided by individual PPHC@Home programs vary considerably.(25) In an integrative review I published in 2018¹, I found that numerous multilevel factors, including a child's age and diagnosis, family support, access to 24-7 medical and nursing care, coordination of services, financial and funding arrangements, geography, and state and national regulations such as concurrent care, together influence the use of PPHC@Home.(25) The provision of PPHC@Home therefore varies widely, and PPHC@Home outcomes, including avoidable hospitalizations (19, 26), pain and symptom management (19, 27),

¹ In my qualifying exam paper, I explored the multi-level factors associated with the increased use of community-based pediatric palliative and hospice care. This paper was published in 2018 in the *Journal of Pain and Symptom Management*. (1. Boyden JY, Curley MAQ, Deatrick JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage* 2018;55:117-131. doi: 10.1016/j.jpainsymman.2017.04.017)

and parental mental health (28), also likely vary across programs and geographic areas.(9, 10, 24, 25)

Outcomes measurement in PPHC@Home

Providing consistent, high-quality PPHC@Home and ensuring that care teams are meeting children's and families' most important needs and priorities require the systematic measurement of outcomes and experiences with care. Indeed, palliative care experts have argued that "a fundamental barrier" to improving PPHC@Home for children with serious illnesses and their families is our inability to effectively measure perceptions of quality of care and care outcomes.(29) In particular, measures of patient and family perceptions of, and experiences with, the care they receive would provide critical information regarding the care provided, including if services meet patient and family priorities and expectations, as well as areas of unmet need and potential improvement for individual patients and families. These measures may be particularly valuable for a comprehensive assessment of care quality (30), and could ultimately facilitate care improvement efforts within and across programs, institutions, and geographic areas.(30-33)

In order to effectively measure PPHC@Home experiences, instruments should possess, at a minimum, 4 characteristics.(31, 34-36) First, instruments should be important and relevant; that is, informed by patients' and families' priorities and goals for care and aligned with health system priorities.(36) Second, instruments should be usable and actionable; that is, the instrument should provide vital and actionable data on child and family experiences and other outcomes that could help providers improve care

delivery.(33, 37) Third, instruments should be feasible; that is, the instrument should be easy to integrate into routine clinical care, should not place undue burden on patients and families to complete, and the benefits of the knowledge gained should outweigh the cost and burden of measurement.(33, 37, 38) For example, briefer instruments may be more easily integrated into clinical care routines, may lower respondent burden, and may lead to higher response rates and data quality.(39-41) And finally, instruments should be scientifically acceptable; that is, instruments should provide valid and reliable data about the PPHC@Home setting and be responsive to changes in care quality over time.(33, 42)

Gaps in current PPHC@Home outcomes measurement

Recent systematic reviews of instruments to measure PPC outcomes have found that, across the field, no consensus exists on what aspects of care to measure because dimensions of care that “matter the most for children in PPC are not currently known.”(43) Additionally, many instruments are limited to specific diseases or conditions, and many are predominantly used retrospectively to explore parental perspectives about past events and satisfaction with the care received, potentially limiting reliability of results.(30, 43) Finally, many studies do not report the psychometric properties of the instrument used.(30, 43) Another recently published scoping review of indicators to assess the impact of PPC identified 82 different process and outcome indicators, with little agreement on the definitions and importance of these indicators.(44)

Many of these same issues affect the measurement of PPHC@Home:

Importance and relevance. Previous research, conducted almost exclusively with inpatient-based samples of parents and providers, has identified several factors that influence parents' perceptions of the quality of care; these include excellent pain and symptom management (27, 45-48); child- and family-centered care and decision-making (13, 45, 46, 49); inclusion of siblings in care processes (27, 45); consistent, high quality communication between family and providers (13, 27, 45, 47, 49); family education and preparation for the end of life (47); psychosocial and spiritual care (46, 49); a comfortable death (45, 48, 49); care coordination and management (13, 46, 49); and bereavement care.(27) While there is evidence that these care components are as important in the home setting (10, 24, 25, 27, 50), we do not currently know which of these aspects of care are the most valuable for children and families at home, and, thus, most important to measure.

From a broader systems perspective, improving PPHC@Home is increasingly recognized as a priority by health care leaders such as the American Academy of Pediatrics (51), the Institute of Medicine (22), and the National Quality Forum.(52) Improving our knowledge of PPHC@Home access, quality, and outcomes through research is also recognized as a priority by the National Institute of Nursing Research (53, 54) and leading experts in the field of PPC.(55, 56) There are, however, no unified recommendations on which specific dimensions of PPHC@Home to measure and to focus improvement efforts.

Usable/actionable. While several family-reported measures have been developed to evaluate PPC provided in hospital and community settings for children, these instruments were developed for populations outside the U.S.(57-59) These

instruments will likely not produce useful or actionable information for the population of PPHC@Home patients and families for several reasons. First, there are significant differences in health care system structures, funding mechanisms, cultural norms, social policies, and provider practices between the U.S. and these non-U.S. populations, limiting the applicability of these instruments.(60) Additionally, two instruments were not specific to home-based care, and included items specific to hospital-based care (e.g., affordability of hospital food), which would again not produce useful information for evaluating PPHC@Home.(58, 59) And finally, all three instruments were developed exclusively for the retrospective completion by bereaved parents (57-59) (although one instrument has more recently been tested for use across the illness trajectory in the oncology setting (61)). This singular focus on past care experiences is similar to tools developed for other program-specific evaluation studies in the U.S.(12-16, 20) While these tools can provide us with information about the effectiveness of programs that can be used to improve care for future children and families, they cannot provide us with specific, clinically-actionable information about the often rapidly-evolving needs of children and families currently receiving care.(30) This limits opportunities to improve care and influence care outcomes for individual children and families at the time is provided.

Feasible. Existing multi-dimensional instruments are comprehensive yet lengthy, ranging from 56 (57) to 136 (58) items. This may place an undue burden on families to complete, particularly while families are currently providing care for their children. Lengthy instruments may also limit response rates (39-41) and may be difficult to integrate into busy clinical care routines.

Scientifically acceptable. In general, few published PPC outcomes instruments report psychometric properties.(30, 43) In the PPHC@Home context specifically, many instruments have been developed for individual programs and have not been psychometrically evaluated (12-16, 20), particularly for use across programs. Additionally, as noted above, many existing instruments survey bereaved parents. In general, retrospectively-administered outcome measures have the potential to introduce recall bias and limit the reliability of the instrument.(30, 43, 62)

While measures have been developed and validated for adult palliative care and hospice populations, these measures have not been evaluated for use in pediatric practice.(23, 46, 63) For example, the content validity of adult instruments may change when applied to pediatric populations due to fundamental differences between adult and pediatric palliative care (e.g., role of parents in decision-making, unique psychosocial and emotional needs of terminally-ill children, siblings, and parents).(50, 51, 63) For example, location of death has traditionally been used as a quality indicator in adult palliative care, which is not supported as an indicator of PPC quality.(51, 63) There is a growing understanding that “the infrastructure to measure and improve pediatric end-of-life care quality cannot simply mimic that in the adult world” (p. e875).(63) Another challenge is adapting existing, validated generic pediatric measures for use in a palliative care context. Generic pediatric measures may contain items that may not be appropriate for pediatric populations with serious illnesses (30, 64), again affecting content validity. To illustrate, one study of Medicaid-enrolled children with life-limiting illnesses examined the psychometric properties of the PedsQL instrument, which is one of the most commonly-used instruments to measure pediatric quality of life. The authors

concluded that the PedsQL does not have valid psychometric properties for measuring health-related quality of life in the pediatric palliative care setting. For example, the authors conducted a confirmatory factor analysis, which did not support the construct validity of the PedsQL for use in the population of children with life-limiting illness, and the authors hypothesized that the health-related quality of life structures between children with life-limiting illnesses and other populations of children may be sufficiently different.(64) These generic pediatric measures may miss important domains or items to comprehensively assess PPC outcomes, and should be further evaluated before use in a PPC context.

In order to develop an instrument that is important to children and families and relevant to their experiences of PPHC@Home (importance/relevance), that produces clinically-actionable information about care outcomes for clinical teams (usable/actionable), that families can complete without undue burden and that can be integrated into routine clinical care (feasible), and that produces consistent and accurate measurements in the PPHC@Home setting (scientifically acceptable), a multi-method, multi-stakeholder process for instrument development is necessary.

PURPOSE, SPECIFIC AIMS, FRAMEWORK, AND METHODS

Purpose

The purpose of this dissertation project was to develop and evaluate a family-reported instrument to measure experiences with PPHC@Home provided in the U.S. The specific aims were as follows:

Specific aims for this dissertation project

Aim 1: Explore how parents rate and prioritize domains of PPHC@Home using a discrete choice experiment with parents whose children are receiving or who have received PPHC@Home services.

Aim 2: Winnow an item pool of approximately 75 measures of PPHC@Home, drawn from existing measures, practice standards and guidelines, and a literature review, to approximately 15 to 20 items based on stakeholder judgments of importance using discrete choice experiments with 1) a sample of national PPHC professionals, and 2) a sample of at least 30 parents whose children are receiving or who have received PPHC@Home services.

Aim 3: Perform cognitive interviews to evaluate the content validity and clarity of the newly developed EXPERIENCE@Home measure in a sample of at least 10 parents whose children are receiving or who have received PPHC@Home services.

Aim 4: Propose next steps in the evaluation and development of the EXPERIENCE@Home Measure, including 1) an evaluation of the validity, reliability, feasibility, and acceptability of the instrument with a sample of parents of children currently receiving PPHC@Home services, and 2) an evaluation of the perceived clinical utility of the instrument with an interdisciplinary group of PPHC stakeholders.

Conceptual framework

The National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) are widely-accepted standards for high-quality palliative care.(52) While these guidelines were developed for all people living with serious illness, regardless of

diagnosis, age, or setting, I sought a framework specific to high-quality care for children with serious illnesses and their families living in the home setting because of the unique attributes of pediatric palliative and end-of-life care, as well as the unique needs of children living with serious illness and their families (5, 22, 46, 50, 63, 65), particularly in the home setting. I further adapted the NCP Guidelines using pediatric palliative care specific practice guidelines (22, 51) and standards of care (65), peer-reviewed literature (66), and informed by a panel of PPHC stakeholders (providers and parent advocates). The adapted framework includes 20 PPHC@Home-specific domains and sub-domains (Table 1).

Overview of methods

This study utilized a **multi-method, mixed-stakeholder approach** for instrument development. My instrument development process included four completed phases (Dissertation Aims 1, 2, 3): First, item identification and development using an integrative review of the literature, practice guidelines, and standards of care, Second, initial item prioritization and reduction using a discrete choice experiment with PPHC providers, researchers, and parent advocates; third, final item prioritization and reduction using a discrete choice experiment with bereaved parents and parents currently receiving care for their child at home; and fourth, cognitive interviewing with parents (Figure 1). Phase five, proposed for completion during my post-doctoral fellowship (Dissertation Aim 4), will include further psychometric evaluation and field testing of the instrument (Figure 1).

In terms of multi-methods, this study used a discrete choice experiment (DCE) methodology, which comes from a family of choice-based methods rooted in the fields of

econometrics and marketing.(67) The DCE approach allowed for the quantitative measurement of participants' stated preferences for items that they feel are most important and salient to include in a PPHC@Home measure. In particular, I used a DCE with maximum difference (MaxDiff) scaling, which allowed me to obtain quantitative estimates of the relative importance of specific items and domains, and to rank order and winnow items. I employed a traditional MaxDiff in Aim 1, which allowed participants to rate all 20 domains. I then employed a "Bandit" MaxDiff approach in Aim 2 to rate items, which is a MaxDiff approach that: first, oversamples top-rated items to increase the precision of estimates of these items; second, minimizes sample size requirements; and third, decreases the cognitive and time burden placed on participants by allowing each participant to rate a sub-set of the overall item pool.(68,69)

Additionally, this study used cognitive interviewing to strengthen the content validity and reduce potential response error of the proposed instrument (Aim 3). Cognitive interviewing is a family of diagnostic techniques used to pre-test survey instruments, providing information about the relevance and clarity of instrument items from the parents' perspective.(70, 71) And finally, the proposed next steps in the instrument development process will leverage the experience-based co-design approach (Aim 4). Experience-based co-design is a design process that incorporates users' experience – leading service design alongside patient, family, or staff "users" – to improve the user's experience.(72)

From the multi-stakeholder perspective, I developed this instrument with the participation of national PPHC experts (Aims 1, 2) and parents of children who are receiving or who have received PPHC@Home services (Aims 1, 2, 3), as well as with the support of my

research mentors who have expertise in outcomes measurement, instrument development, DCE, and cognitive interviewing methods.

ORGANIZATION OF THIS DISSERTATION

The present chapter, **Chapter One: General Introduction**, has presented an overview of this dissertation project’s background, significance, and specific aims. The next two chapters (**Chapters Two & Three**) contain manuscripts for publication (see table below).

Chapter Four is a grant application for the National Institute of Nursing Research (NINR) Ruth L. Kirschstein National Research Service Award (NRSA) Post-doctoral Fellowship (Parent F32) I submitted in December 2019. **Chapter Five: Summary and Future Directions** provides a synthesis of the research findings, and a description and interpretation of the significance of the overall dissertation project results. It also identifies implications for future research, practice, and policy.

Paper Title	Brief Description	Submission Details
* Paper 1: What do parents value regarding pediatric palliative and hospice care in the home setting? (Chapter 2)	This paper describes a quantitative, choice-based approach to measuring and prioritizing what parents value the most regarding pediatric palliative and hospice care in the home setting (Aim 1).	Accepted for publication in the <i>Journal of Pain and Symptom Management</i> (July 2020)
* Paper 2: Developing a family-reported measure of experiences with pediatric palliative and hospice care in the home setting: A multi-method, multi-stakeholder approach (Chapter 3)	This paper presents a multi-method (quantitative choice-based method; qualitative cognitive interviewing), multi-stakeholder (providers, researchers, parents) approach to developing an instrument to measure family-reported experiences with pediatric palliative and hospice	Submitted to <i>BMC Palliative Care</i> (July 2020)

	care in the home setting (Aims 1, 2, 3).	
Paper 3: Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home) Project (F32 proposal) (Chapter 4)	In this F32 application, we propose to evaluate the newly developed EXPERIENCE@Home Measure from the perspective of parents whose children are currently receiving palliative and hospice care in the home setting, as well as from the perspective of palliative and hospice care providers who will ultimately be implementing this tool in clinical practice. To accomplish this goal, we propose three specific aims: 1) Evaluate evidence for score validity and reliability of the EXPERIENCE@Home Measure from parents of children with serious life-threatening illnesses currently receiving palliative or hospice care at home; 2) Evaluate the feasibility and acceptability of using the EXPERIENCE@Home Measure in real-time in parents of children with serious life-threatening illnesses currently receiving palliative or hospice care at home; and 3) Evaluate the perceived clinical utility of the EXPERIENCE@Home Measure with an interdisciplinary group of pediatric palliative and hospice care stakeholders (Aim 4).	Submitted in December 2019 to the NINR and received an overall impact score of 27. This application was funded July 2020.

* = data-based paper

SUMMARY AND SIGNIFICANCE

From this dissertation project, we have a clearer understanding of what parents prioritize regarding the domains of PPHC@Home. Additionally, the EXPERIENCE@Home Measure is the first known comprehensive, family-reported measure of families' experiences with PPHC@Home developed for children and families receiving care in the

U.S. To my knowledge, my project was also the first time the DCE methodology was used for instrument development in health care. Additionally, it is the first known measure developed with the participation of both bereaved parents and parents currently caring for their child at home, which provides a unique and comprehensive perspective of experiences along the illness trajectory and through the end of life. The EXPERIENCE@Home Measure is also among the first instruments to measure real-time experiences with PPHC@Home. While there is debate as to the optimal timing of survey administration in patient-reported outcomes measurement (73), surveying parents whose child is currently receiving services may produce the most complete and accurate information about care experiences (30, 74, 75) and allow providers to respond, in real-time, to patient and family needs and perceived gaps in care.(31, 76-78)

My approach to instrument development has allowed me to leverage the unique perspectives and expertise of diverse stakeholders and the strengths of unique yet complementary methods to develop a comprehensive, meaningful, and practical instrument for measuring PPHC@Home experiences. I will continue to leverage the perspectives and expertise of diverse PPHC stakeholders in my proposed next steps to evaluate the validity, reliability, feasibility, acceptability, and clinical utility of this instrument in a broader sample of parents currently receiving care for their children. The current project and proposed next steps align closely with the National Institute of Nursing Research's strategic research plan (54) and funding priorities (53) to advance high-quality, evidence-based community-focused palliative and hospice care, particularly for children and families.

BIBLIOGRAPHY

1. Bethell CD, Read D, Blumberg SJ, Newacheck PW. What is the prevalence of children with special health care needs? Toward an understanding of variations in findings and methods across three national surveys. *Mat Child Health J* 2008;12:1-14. doi: 10.1007/s10995-007-0220-5
2. Fraser LK, Miller M, Aldridge J, McKinney PA, Parslow RC. Life-limiting and life-threatening conditions in children and young people in the United Kingdom; National and regional prevalence in relation to socioeconomic status and ethnicity. University of Leeds, 2011. Available from: <https://www.togetherforshortlives.org.uk/wp-content/uploads/2018/01/ExRes-Childrens-Hospices-Ethnicity-Report-Leeds-Uni.pdf>. Accessed June 26, 2020.
3. Feudtner C, Feinstein JA, Satchell M, Zhao H, Kang TI. Shifting place of death among children with complex chronic conditions in the United States, 1989-2003. *JAMA* 2007;297:2725-2732. doi: 10.1001/jama.297.24.2725
4. Feudtner C, Kang TI, Hexem KR, et al. Pediatric palliative care patients: A prospective multicenter cohort study. *Pediatr* 2011;127:1094-1101. doi: 10.1542/peds.2010-3225
5. Friebert S, Williams C. NHPCO'S Facts and Figures: Pediatric Palliative and Hospice Care in America. Alexandria, VA: NHPCO, 2015. Available from: https://www.nhpc.org/wp-content/uploads/2019/04/Pediatric_Facts-Figures-1.pdf. Accessed June 26, 2020.
6. Nageswaran S, Radulovic A, Anania A. Transitions to and from the acute inpatient care setting for children with life-threatening illness. *Pediatr Clin North Am* 2014;61:761-783. doi: 10.1016/j.pcl.2014.04.008
7. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980-1998. *Pediatr* 2002;109:656-660. doi: 10.1542/peds.109.4.656
8. Feudtner C, Hays RM, Haynes G, et al. Deaths attributed to pediatric complex chronic conditions: National trends and implications for supportive care services. *Pediatr* 2001;107:E99. doi: 10.1542/peds.107.6.e99
9. Lindley LC, Lyon ME. A profile of children with complex chronic conditions at end of life among Medicaid beneficiaries: Implications for health care reform. *J Palliat Med* 2013;16:1388-1393. doi: 10.1089/jpm.2013.0099
10. Kaye E, Rubenstein J, Levine D, et al. Pediatric palliative care in the community. *CA Cancer J Clin* 2015;65:315-333. doi: 10.3322/caac.21280
11. Feudtner C, Womer J, Augustin R, et al. Pediatric palliative care programs in children's hospitals: A cross-sectional national survey. *Pediatr* 2013;132:1063-1070.
12. 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;18:143-150. doi: 10.1089/jpm.2014.0285
13. Hays RM, Valentine J, Haynes G, et al. The Seattle Pediatric Palliative Care Project: Effects on family satisfaction and health-related quality of life. *J Palliat Med* 2006;9:716-28. doi: 10.1089/jpm.2006.9.716
14. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based pediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. *BMC Palliat Care* 2016;15:73. doi: 10.1186/s12904-016-0138-z

15. Gans D, Kominski GF, Roby DH, et al. Better outcomes, lower costs: Palliative care program reduces stress, costs of care for children with life-threatening conditions. UCLA Center for Health Policy Research 2012. Available from: <https://escholarship.org/uc/item/7244h6wq>. Accessed June 26, 2020.
16. Bona K, Bates J, Wolfe J. Massachusetts' Pediatric Palliative Care Network: Successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med* 2011;14:1217-1223. doi: 10.1089/jpm.2011.0070
17. Niswander LM, Cromwell P, Chirico J, Gupton A, Korones DN. End-of-life care for children enrolled in a community-based pediatric palliative care program. *J Palliat Med* 2014;17:589-591. doi: 10.1089/jpm.2013.0576
18. Lindley LC, Kieim-Malpass J. Quality of paediatric hospice care for children with and without multiple complex chronic conditions. *Int J Palliat Nurs* 2017;23:230-237. doi: 10.12968/ijpn.2017.23.5.230
19. Thienprayoon R, Marks E, Funes M, et al. Perceptions of the pediatric hospice experience among English- and Spanish-speaking families. *J Palliat Med* 2016;19:30-41. doi: 10.1089/jpm.2015.0137
20. Knapp CA, Madden VL, Curtis CM, et al. Partners in Care: Together for Kids: Florida's model of pediatric palliative care. *J Palliat Med* 2008;11:1212-1220. doi: 10.1089/jpm.2008.0080
21. Postier A, Chrastek J, Nugent S, Osenga K, Friedrichsdorf SJ. Exposure to home-based pediatric palliative and hospice care and its impact on hospital and emergency care charges at a single institution. *J Palliat Med* 2014;17:183-188. doi: 10.1089/jpm.2013.0287
22. Field MJ, Behrman RE. *When Children Die: Improving Palliative and End-of-Life care for Children and their Families*. Washington, DC: The National Academies Press, 2003:419-420. Available from: <https://www.nap.edu/catalog/10390/when-children-die-improving-palliative-and-end-of-life-care>. Accessed June 5, 2020.
23. Herd E. Quality assessment in children's hospice care. *Paediatr Nurs* 2005;17:20-23. doi: 10.7748/paed2005.04.17.3.20.c975
24. Carroll JM, Torkildson C, Winsness JS. Issues related to providing quality pediatric palliative care in the community. *Pediatr Clin N Amer* 2007;54:813-27, xiii. doi: 10.1016/j.pcl.2007.06.002
25. Boyden JY, Curley MAQ, Deatrck JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage* 2018;55:117-131. doi: 10.1016/j.jpainsymman.2017.04.017
26. Thienprayoon R, Lee SC, Leonard D, Winick N. Hospice care for children with cancer: Where do these children die? *J Pediatr Hematol Oncol* 2015;37:373-377. doi: 10.1097/MPH.0000000000000331
27. Contro N, Larson J, Scofield S, Sourkes B, Cohen H. Family perspectives on the quality of pediatric palliative care. *Arch Pediatr Adolesc Med* 2002;156:14-19. doi: 10.1001/archpedi.156.1.14
28. Verberne LM, Kars MC, Schouten-van Meeteren AY, et al. Aims and tasks in parental caregiving for children receiving palliative care at home: a qualitative study. *Eur J Pediatr* 2017;176:343-354. doi: 10.1007/s00431-016-2842-3

29. Davies B, Gudmundsdottir M, Worden B, et al. "Living in the dragon's shadow" fathers' experiences of a child's life-limiting illness. *Death Stud* 2004;28:111-135. doi: 10.1080/07481180490254501
30. 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;30:935-949. doi: 10.1177/0269216316649155
31. Conway PH, Mostashari F, Clancy C. The future of quality measurement for improvement and accountability. *JAMA* 2013;309:2215-2216. doi: 10.1001/jama.2013.4929
32. Snyder CF, Aaronson NK, Choucair AK, et al. Implementing patient-reported outcomes assessment in clinical practice: A review of the options and considerations. *Qual Life Res* 2012;21:1305-14. doi: 10.1007/s11136-011-0054-x
33. Hodgson ES, Lannon CM, et al. Principles for the development and use of quality measures. *Pediatrics* 2008;121:411-418. doi: 10.1542/peds.2007-3281
34. Dy SM. Measuring the quality of palliative care and supportive oncology: principles and practice. *J Support Oncol* 2013;11:160-164. doi: 10.12788/j.suonc.0017
35. Kuhlthau KA, Mistry KB, Forrest CB, Dougherty D. Advancing the science of measurement in pediatric quality of care. *Acad Pediatr* 2014;14:S1-3. doi: 10.1016/j.acap.2014.06.016
36. National Quality Forum. The ABCs of Measurement. Washington, DC: National Quality Forum, 2018. Available from: https://www.qualityforum.org/Measuring_Performance/ABCs_of_Measurement.aspx. Accessed June 26, 2020.
37. American Academy of Pediatrics. Pediatric quality measurement. 2018. Available from: <https://www.aap.org/en-us/professional-resources/quality-improvement/Pages/Pediatric-Quality-Measurement.aspx>. Accessed June 26, 2020.
38. Lindley LC, Rotella JD, Ast K, Matzo M, Kamal AH. The Quality Improvement Environment: Results of the 2016 AAHPM/HPNA Membership Needs Assessment Survey. *J Pain Symptom Manage* 2017;54:766-771. doi: 10.1016/j.jpainsymman.2017.07.031
39. Galesic M, Bosnjak M. Effects of questionnaire length on participation and indicators of response quality in a web survey. *Public Opinion Quarterly* 2009;73:349-360. doi: <https://doi.org/10.1093/poq/nfp031>
40. Ronckers C, Land C, Hayes R, Verduijn P, Van Leeuwen F. Factors impacting questionnaire response in a Dutch retrospective cohort study. *Ann Epidemiol* 2004;14:66-72. doi: 10.1016/s1047-2797(03)00123-6
41. Chiu L, Chow E, Cella D, et al. Comparison of three shortened questionnaires for assessment of quality of life in advanced cancer. *J Palliat Med* 2014;17:918-923. doi: 10.1089/jpm.2014.0012
42. Institute for Healthcare Improvement (IHI). Measures. 2018. Available from: <http://www.ihc.org/resources/Pages/Measures/default.aspx>. Accessed March 10, 2020.
43. Friedel M, Aujoulat I, Dubois A-C, Degryse J-M. Instruments to measure outcomes in pediatric palliative care: A systematic review. *Pediatr* 2019;143:e20182379. doi: 10.1542/peds.2018-2379

44. Widger K, Medeiros C, Trenholm M, Zuniga-Villanueva G, Streuli JC. Indicators used to assess the impact of specialized pediatric palliative care: A scoping review. *J Palliat Med* 2019;22:199-219. doi: 10.1089/jpm.2018.0420
45. Andresen EM, Seecharan GA, Toce SS. Provider perceptions of child deaths. *Arch Pediatr Adolesc Med* 2004;158:430-5. doi: 10.1001/archpedi.158.5.430
46. Donnelly JP, Downing K, Cloen J, et al. Development and assessment of a measure of parent and child needs in pediatric palliative care. *J Pain Symptom Manage* 2017;55:1077-1084. doi: 10.1016/j.jpainsymman.2017.12.484
47. 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
48. Meyer EC, Burns JP, Griffith JL, Truog RD. Parental perspectives on end-of-life care in the pediatric intensive care unit. *Crit Care Med* 2002;30:226-231. doi: 10.1097/00003246-200201000-00032
49. Weidner NJ, Cameron M, Lee RC, et al. End-of-life care for the dying child: What matters most to parents. *J Palliat Care* 2011;27:279-286. doi: <https://doi.org/10.1177/082585971102700404>
50. Widger KA, Wilkins K. What are the key components of quality perinatal and pediatric end-of-life care? A literature review. *J Palliat Care* 2004;20:105-112. doi: <https://doi.org/10.1177/082585970402000207>
51. Feudtner C, Friebert S, Jewell J. Pediatric palliative care and hospice care commitments, guidelines, and recommendations. *Pediatr* 2013;132:966-972. doi: 10.1542/peds.2013-2731
52. National Consensus Project for Quality Palliative Care. *Clinical Practice Guidelines for Quality Palliative Care*. 4th ed. Richmond, VA: National Coalition for Hospice and Palliative Care, 2018. Available from: <https://www.nationalcoalitionhpc.org/ncp/>. Accessed June 5, 2020.
53. PAR-19-320: Palliative Care in Home and Community Settings (R21 Clinical Trial Optional). National Institutes of Health. Posted July 31, 2019. Available from: <https://grants.nih.gov/grants/guide/pa-files/par-19-320.html#:~:text=Home%20and%20community%20in%20this,of%20life%20and%20palliative%20care>. Accessed June 27, 2020.
54. National Institute of Nursing Research. *The NINR's Strategic Plan: Advancing Science, Improving Lives*. 2016 September. Available from: https://www.ninr.nih.gov/sites/files/docs/NINR_StratPlan2016_reduced.pdf. Accessed June 26, 2020.
55. Feudtner C, Rosenberg AR, Boss RD, et al. Challenges and priorities for pediatric palliative care research in the U.S. and similar practice settings: Report from a Pediatric Palliative Care Research Network workshop. *J Pain Symptom Manage* 2019. doi: 10.1016/j.jpainsymman.2019.08.011
56. Baker JN, Levine DR, Hinds PS, et al. Research priorities in pediatric palliative care. *J Pediatr* 2015;167:467-470. doi: 10.1016/j.jpeds.2015.05.002
57. Vollenbroich R, Duroux A, Grasser M, et al. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med* 2012;15:294-300. doi: 10.1089/jpm.2011.0196

58. Widger K, Tourangeau AE, Steele R, Streiner DL. Initial development and psychometric testing of an instrument to measure the quality of children's end-of-life care. *BMC Palliat Care* 2015;14:1. doi: 10.1186/1472-684X-14-1
59. Zimmermann K, Cignacco E, Eskola K, et al. Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu): An instrument to assess parental experiences and needs during their child's end-of-life care. *J Adv Nurs* 2015;71:3006-3017. doi: 10.1111/jan.12741
60. Linton JM, Feudtner C. What accounts for differences or disparities in pediatric palliative and end-of-life care? A systematic review focusing on possible multilevel mechanisms. *Pediatr* 2008;122:574-582. doi:
61. Widger K, Brennenstuhl S, Duc J, Tourangeau A, Rapoport A. Factor structure of the Quality of Children's Palliative Care Instrument (QCPCI) when completed by parents of children with cancer. *BMC Palliat Care* 2019;18:23. doi: 10.1186/s12904-019-0406-9
62. Kamal AH, Hanson LC, Casarett DJ, et al. The quality imperative for palliative care. *J Pain Symptom Manage* 2015;49:243-253. doi: 10.1016/j.jpainsymman.2014.06.008
63. Johnston EE, Rosenberg AR, Kamal AH. Pediatric-specific end-of-life care quality measures: An unmet need of a vulnerable population. *J Oncol Pract* 2017;13:e874-e880. doi: 10.1200/JOP.2017.021766
64. Huang IC, Shenkman EA, Madden VL, et al. Measuring quality of life in pediatric palliative care: challenges and potential solutions. *Palliative Medicine* 2010;24:175-182. doi: 10.1177/0269216309352418
65. National Hospice and Palliative Care Organization (NHPCO). Standards of practice for pediatric palliative care. Alexandria, VA: NHPCO, 2019. Available from: https://www.nhpc.org/wp-content/uploads/2019/07/Pediatric_Standards.pdf. Accessed June 12, 2020.
66. Thienprayoon R, Mark MSJ, Grosseohme D. Provider-prioritized domains of quality in pediatric home-based hospice and palliative care: A study of the Ohio Pediatric Palliative Care and End-of-Life Network. *J Palliat Med* 2018;31:290-296. doi: 10.1089/jpm.2017.0333
67. Ryan M. Discrete choice experiments in health care. *BMJ* 2004;328:360. doi: 10.1136/bmj.328.7436.360
68. Sawtooth Software. Bandit MaxDiff: When to use it and why it can be a better choice than standard MaxDiff. Orem, UT; 2018. Available from: <https://www.sawtoothsoftware.com/support/technical-papers/maxdiff-best-worst-scaling/175-support/technical-papers/maxdiff-best-worst-scaling/1943-bandit-maxdiff-when-to-use-it-and-why-it-can-be-a-better-choice-than-standard-maxdiff>. Accessed June 30, 2020.
69. Chrazan K, Orme B. Applied MaxDiff: A practitioner's guide to Best-Worst Scaling Provo, UT: Sawtooth Software, Inc.; 2019.
70. Collins D. Pretesting survey instruments: An overview of cognitive methods. *Qual Life Res* 2003;12:229-238. doi: <https://doi.org/10.1023/A:1023254226592>
71. Knaf K, Deatrick J, Gallo A, et al. The analysis and interpretation of cognitive interviews for instrument development. *Research in Nursing & Health* 2007;30:224-234. doi: 10.1093/jpepsy/jsx095

72. Bate P, Robert G. Experience-based design: From redesigning the system around the patient to co-designing services with the patient. *Qual Saf Health Care* 2006;15:307-10. doi: 10.1136/qshc.2005.016527
73. Boyce MB, Browne JP, Greenhalgh J. The experiences of professionals with using information from patient-reported outcome measures to improve the quality of healthcare: a systematic review of qualitative research. *BMJ Qual Saf* 2014;23:508-18. doi: 10.1136/bmjqs-2013-002524
74. DeWalt DA, Rothrock N, Yount S, Stone AA, Group PC. Evaluation of item candidates: the PROMIS qualitative item review. *Medical Care* 2007;45:S12-21. doi: 10.1097/01.mlr.0000254567.79743.e2
75. Dudgeon D. The impact of measuring patient-reported outcome measures on quality of and access to palliative care. *J Palliative Med* 2017;21:S-76-S-80. doi: 10.1089/jpm.2017.0447
76. Basch E, Torda P, Adams K. Standards for patient-reported performance measures. *JAMA* 2013;310:139-140. doi: 10.1001/jama.2013.6855
77. Boyce MB, Browne JP. Does providing feedback on patient-reported outcomes to healthcare professionals result in better outcomes for patients? A systematic review. *Qual Life Res* 2013;22:2265-78. doi: 10.1007/s11136-013-0390-0
78. Van Der Wees PJ, Nijhuis-Van Der Sanden MWG, Ayanian JZ, et al. Integrating the use of patient-reported outcomes for both clinical practice and performance measurement: Views of experts from 3 countries. *Milbank Q* 2014;92:754-775. doi: 10.1111/1468-0009.12091

TABLES AND FIGURES

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home

Figure 1: EXPERIENCE@Home Measure Development Process

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home

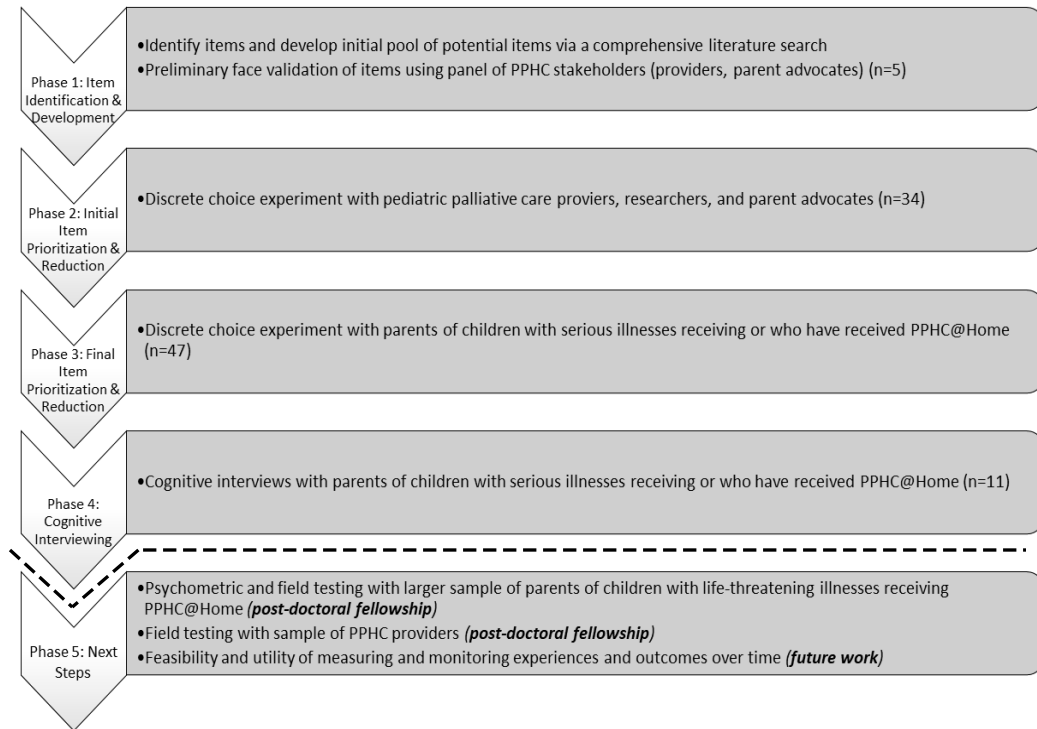
Domains	Description
Communication between family and care team	Care team communicates with the child and family to make sure that the care provided meets the child's and family's values, preferences, goals, and needs
Relationship between family and care team	Relationship between care team and family is built on trust, respect, and advocacy for the child's and family's needs
Knowledge and skills of care team providers	Care team members have the necessary education and training to provide high quality palliative care for children and families
Access to care	Care team provides access to palliative and hospice care to the child and family 24 hours a day, 7 days a week
Physical care: Communication	Care team provides information about treatments for child's pain and other physical symptoms (for example, nausea, fatigue, constipation)
Physical care: Symptom management	Care team assesses and manages pain and other physical symptoms and side effects based on the best available medical evidence
Psychological and emotional aspects of care (<i>Child, Parents, Siblings, and Extended Social Network</i>)*	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the child, family, and family's community based on the best available medical evidence
Practical aspects of care	Care team provides the family with assistance and resources for dealing with financial and insurance related issues
Social aspects of care (<i>Child, Parents</i>)*	Care team helps with social issues to meet child-family needs, promote child-family goals, and maximize child-family strengths and well-being (examples include helping family maintain and strengthen their support network; help family develop strategies to balance caregiving, work, and family needs)
Spiritual and religious aspects of care	Care team assists with religious and spiritual rituals or practices as desired by the child and family
Cultural aspects of care	Care team respects the child's and family's cultural beliefs and language preferences

Communication at the end of life	Care team works with the child and family to develop and implement a care plan to address actual or potential symptoms at the end of life
Caregiver support at the end of life	Care team meets the emotional, spiritual, social, and cultural needs of families at the end of life (for example, preparing parents for the end of life)
Ethical and legal aspects of care	<p>Child's and family's goals, preferences, and choices are respected within the limits of state and federal law, current standards of medical care, and professional standards of practice.</p> <p>These goals/preferences/choices are documented and shared with all professionals involved in the child's care</p>
Coordination of care	Care team works to make sure that when there are transfers between healthcare settings and providers, that there is timely and thorough communication of the child's/family's goals, preferences, values, and clinical information to ensure continuity of care and seamless follow-up (for example, getting needed services, arranging for medical equipment)
Continuity of care	Care team works to make sure that the delivery of palliative and hospice care is seamless across care settings and providers (for example, the same providers work with family)

*Separate sub-domain for each group

Note: These domains are based on the National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) (52), which were further adapted using pediatric palliative care specific guidelines and the literature (22, 51, 65, 66) and informed by a panel of PPHC stakeholders (providers and parent advocates).

Figure 1: EXPERIENCE@Home Measure Development Process



CHAPTER 2: What do parents value regarding pediatric palliative and hospice care in the home setting? (Paper 1)

Jackelyn Y. Boyden, MS, MPH, RN¹

Mary Ersek, PhD, RN, FPCN^{1,2,7}

Janet A. Deatrick, PhD, RN, FAAN¹

Kimberley Widger, RN, PhD, CHPCN(C)^{3,4}

Gwenn LaRagione, RN, BSN, CCM, CHPPN⁵

Blyth Lord, EdM⁶

Chris Feudtner, MD, PhD, MPH^{5,7}

¹ School of Nursing, University of Pennsylvania, Philadelphia PA

² Corporal Michael J. Crescenz VA Medical Center, Philadelphia PA

³ Lawrence S. Bloomberg Faculty of Nursing, University of Toronto, Toronto ON Canada

⁴ Hospital for Sick Children, Toronto ON Canada

⁵ Children's Hospital of Philadelphia, Philadelphia, PA

⁶ Courageous Parents Network

⁷ University of Pennsylvania Perelman School of Medicine, Philadelphia PA

Accepted for publication to the *Journal of Pain and Symptom Management* (July 2020)

ABSTRACT

Context: Children with life-shortening serious illnesses and medically complex care needs are often cared for by their families at home. Little, however, is known about what aspects of pediatric palliative and hospice care in the home setting (PPHC@Home) families value the most.

Objective: To explore how parents rate and prioritize domains of PPHC@Home as the first phase of a larger study that developed a family-reported measure of experiences with PPHC@Home.

Methods: Twenty domains of high-value PPHC@Home, derived from the National Consensus Project's Guidelines for Quality Palliative Care, the literature, and a stakeholder panel, were evaluated. Using a discrete choice experiment, parents provided their ratings of the most and least valued PPHC@Home domains. We also explored potential differences in how subgroups of parents rated the domains.

Results: Forty-seven parents participated. Overall, highest-rated domains included *Physical Aspects of Care: Symptom Management*, *Psychological/Emotional Support for the Child*, and *Care Coordination*. Lowest-rated domains included *Spiritual and Religious Aspects of Care* and *Cultural Aspects of Care*. In exploratory analyses, parents who had other children rated the *Psychological/Emotional Aspects of Care for the Sibling(s)* domain significantly higher than parents who did not have other children ($P=0.02$). Furthermore, bereaved parents rated the *Caregiver Support at the End of Life* domain significantly higher than parents who were currently caring for their child ($P=0.04$). No other significant differences in domain ratings were observed.

Conclusions: Knowing what parents value most about PPHC@Home provides the foundation for further exploration and conversation about priority areas for resource allocation and care improvement efforts.

Keywords: pediatric palliative care, pediatric hospice care, home-based care, discrete choice experiment

Running title: Pediatric palliative and hospice care at home

Key Message

This paper describes parents' priorities for pediatric palliative and hospice care in the home setting (PPHC@Home). Parents provided their ratings and rankings of PPHC@Home domains in a discrete choice experiment. Understanding parents' priorities for PPHC@Home facilitates further exploration and conversation about priority areas for resource allocation and care improvement efforts.

INTRODUCTION

Children with life-shortening serious illnesses and medically complex care needs are increasingly cared for by their families at home, particularly toward the end of life.(1-4) These children and families are supported in the home setting by palliative and hospice care programs that vary in structure, staffing, funding, and patient census.(5-9) As a result, many children and their families in the U.S. have variable and, at times, inadequate access to high-quality pediatric palliative and hospice care in the home setting (PPHC@Home).(7, 10, 11)

Previous research with primarily inpatient-based samples of parents and providers have identified several factors that improve parents' perceptions of the quality of care, including effective pain and symptom management (12-16), child- and family-centered care and decision-making (12, 16), inclusion of siblings in care processes (13, 17), consistent and high-quality communication between family and providers (13-17), family education and preparation for the end of life (14), psychosocial and spiritual care (12, 15-17), a comfortable death (15, 16), care coordination and management (12, 16), and bereavement care.(13) Evidence suggests that these domains of care are as important in the home setting.(7, 10, 13, 18-22) Little is known, however, about which of these care domains parents value the most in supporting their child in their home, and how these domains compare in importance to one another. Previous studies prioritized areas for high-quality PPHC@Home clinical care and research using Delphi methods with PPHC@Home stakeholders, including providers and parents.(18, 20) Among top-rated priorities were services, techniques, and resources for pain and symptom relief and psychological support for children and young adults.(18) Given limitations to Delphi-rating methodologies, including the limited ability to differentiate between similar rating

scores (18) and concerns about stability of rating scores (23), we do not know which of these care domains are most important in supporting children at home. While high-quality PPHC@Home encompasses the spectrum of care domains (20, 22), health care resources, particularly in the home and community setting (7, 8), are finite. Knowing, therefore, what parents value most could help guide allocation of these scarce resources, as well as future clinical and research efforts to measure and improve the quality of PPHC@Home.

The goal of this analysis was to measure parents' priorities for PPHC@Home using a quantitative, choice-based approach. This analysis represents the first phase of a larger study to develop a family-reported measure of experiences with PPHC@Home.(24)

METHODS

Study design

We conducted a cross-sectional assessment of parents' priorities regarding the importance of 20 different PPHC@Home domains of care. This assessment represents the first phase of a larger project to develop a family-reported measure of experiences with PPHC@Home.(24) The Children's Hospital of Philadelphia's Institutional Review Board approved the conduct of this study.

Conceptual framework for the domains

The framework for the domains used in this study came from the National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) (22), which were further adapted using pediatric palliative care specific guidelines and the literature (20, 25-28) and informed by a panel of PPHC stakeholders (providers and parent

advocates). The adapted framework includes 20 PPHC@Home-specific domains and sub-domains (Table 1).

Sample

Parents were eligible if they were English-speaking, over the age of 18 years, and had a child with a serious illness who was younger than 25 years at the time care was received. We included parents whose children were currently receiving PPHC@Home, as well as bereaved parents whose children had previously received PPHC@Home. Parents were recruited from the Children's Hospital of Philadelphia's (CHOP) Pediatric Advanced Care Team (PACT) and from the Courageous Parents Network (CPN), which is a virtual community of parents, families, and providers that supports parents with information, skills, tools, and other resources during their child's illness journey.(29)

For participants recruited from CHOP, the principal investigator (PI; first-author JB) worked closely with PACT's nurse coordinator and social worker to screen for eligible participants. The PI then contacted participants by phone. Interested parents provided their electronic informed consent and completed the web-based survey concurrently (in-person or by phone) with the PI or independently via a web link. For participants recruited from CPN, the PI worked closely with CPN's staff to post recruitment materials to CPN's listserv and social media page. Interested participants contacted the PI via phone or email and were screened for eligibility. Eligible participants provided their electronic informed consent and completed the survey via a web-link either concurrently with the PI or independently. Participants were compensated with a \$30 gift card for their time and effort.

Discrete choice experiment

Participants engaged in a discrete choice experiment (DCE), which is a quantitative, choice-based approach to evaluating individuals' stated preferences regarding choices in health care (30-34) and other settings.(35, 36) We used a DCE with maximum difference (MaxDiff) scaling to: 1) obtain a quantitative estimate of the importance of each domain (i.e., domain importance scores), as rated by parents, and 2) rank the PPHC@Home domains by order of the parent-rated importance scores.

In the DCE, participants first reviewed all 20 PPHC@Home domains and domain definitions (which were also accessible during the DCE exercise). They were then presented with sets displaying 4 of the 20 domains. Within each set, participants were instructed to choose, from the 4 listed domains, the one that they felt was the most important in supporting their child in the home, and the domain they felt was least important. This process was repeated for a total of 15 sets, each set being a different combination of the 20 domains, with the same instructions to choose the most and least important among the 4 domains listed in that set. Across all the sets for each respondent, the DCE's design ensured that each domain was shown exactly 3 times and in a balanced set of combinations and permutations with the other domains. Completing all 15 sets took participants approximately 8 to 10 minutes.

Data analysis

Based on the choices that participants made across the 15 DCE sets, domain importance scores were calculated using a hierarchical Bayesian application of multinomial logistic regression, which estimated the average (mean) probability, with 95% confidence intervals, of each domain being chosen as the most or least important for individual participants and across the entire sample. The raw logit scores were then transformed to a relative importance score on a 0 to 100 probability scale, where scores

sum to 100 across domains.(37, 38) This transformation facilitates a readily interpretable comparison of domains.(39) More specifically, this transformed score indicates the **relative importance** of domains on a common scale; for example, a domain that is given a score of 10 is perceived by respondents (as revealed via their choices) as being twice as important as a domain with a score of 5. Domains were rank ordered according to their importance score, where higher scores indicated higher perceived importance. To describe the variation in how respondents rated each domain, we also calculated the interquartile range (IQR) in each domain, and reported the median, 25th and 75th percentiles, and any outliers.(40)

In additional analyses, we explored how domain ratings differed depending on whether there were siblings in the household, whether the parent was bereaved, and whether the parent was a mother or father. All comparisons were conducted using two-sample independent t-tests. Given the exploratory nature of these sub-group analyses, P values < 0.1 were considered statistically significant.

Sample size considerations

DCEs of the design we used converge on stable estimates of the relative scores for items with as few as 20 participants in subgroups.(41, 42) There are currently no standard guidelines for defining sample sizes for these DCE studies; therefore, simulation studies, where the effect of different sample sizes are simulated to test the effect on the reliability of estimates, are recommended.(43) Using data from a previous DCE with a sample of 200 parents of children with serious illnesses (34), we used a bootstrap approach to draw overall dataset samples of 20, 30, or 50 individual responses to the DCE at a time, doing so with replacement and iterating this process 100 times for each sample size. Across all 3 sample sizes, all items were consistently

ordered from top to bottom. While the confidence intervals indicated that similarly rated items may, for some samples, switch ranking, they demonstrated that with a sample size of 30, high-rated items can be clearly differentiated from low-rated items. Thus, we aimed to recruit a minimum of 30 parent participants from 30 unique families.

Survey and analytic software

We designed the DCE and deployed the survey instruments using Lighthouse Studio (Version 9.6.1, Sawtooth Software, Provo, UT), which includes a cloud-based survey platform. We calculated the domain importance scores based on participants' choices across the DCE sets using Lighthouse Studio and conducted statistical analysis of the importance scores using Stata version 15.1 (StataCorp, College Station, TX).

RESULTS

Forty-seven parents from 45 families participated (Table 2). Most participants were white (89.4%), non-Hispanic (91.5%) mothers (93.6%) who were married or partnered (87.2%) and had completed college or graduate school (68.1%). Parents' mean age was 42.6 years (standard deviation 8.5). Fourteen parents (29.8%) were bereaved, and 33 (70.2%) were currently caring for their child at home.

Participants were parents to 45 children who have received PPHC@Home.

Approximately half of these children were between birth and 9 years, and half were between 10 and 25 years of age. Most prevalent primary diagnoses included neuromuscular, neurologic, or mitochondrial (51.1%), genetic or congenital (48.9%), cardiovascular (22.2%), and metabolic (22.2%) diseases. Over half of these children received home-based support primarily from a palliative care team (53.3%), and a third of these children received PPHC@Home for more than 2 years (33.3%).

Average domain importance scores and ranking

Among the 20 domains of PPHC@Home (Figure 1), parents ranked the *Physical Aspects of Care: Symptom Management* domain the highest (mean score 9.68; 95% CI: 8.71, 10.64), followed by *Psychological/Emotional Aspects of Care for the Child* (mean score 8.97; 95% CI: 7.74, 10.21) and *Care Coordination* (mean score 8.92; 95% CI: 7.92, 9.92). Among the lowest-ranked domains were *Emotional Aspects of Care for the Extended Social Network* (mean score 0.90; 95% CI: 0.36, 1.45), *Spiritual and Religious Aspects of Care* (mean score 0.22; 95% CI: 0.12, 0.32), and *Cultural Aspects of Care* (mean score 0.12; 95% CI: 0.07, 0.17).

In terms of relative importance, parents rated the *Physical Aspects of Care: Symptom Management* domain as being twice as important as the *Practical Aspects of Care* domain (mean score 9.68 vs 4.75) and nearly 10 times as important as the *Emotional Aspects of Care for the Family's Extended Social Network* domain (mean score 9.68 vs. 0.90).

Individual-level variation in domain importance scores and ranking

Domains differed not only regarding their mean importance score, but also in the degree to which the participants, as a group, varied in their scores for given domains. For example, the *Physical Aspects of Care: Symptom Management* domain had a median score of 11.18 and an interquartile (IQR) range of 5.25, whereas the *Psychological/Emotional Aspects of Care for the Sibling(s)* domain had a median score of 6.35, yet a much larger IQR of 9.91, representing greater variation in how respondents rated this domain (Figure 2).

Additional analysis: Sub-group comparisons in domain importance scores

We examined three potential notions of how parents might differ in their importance ratings of domains. First, we assessed how parents with other children rated the sibling-specific domain (*Psychological/Emotional Aspects of Care for the Sibling*) compared to parents who did not have other children. The mean score for parents with at least one other child (n=38) was 7.55 (95% CI: 6.01, 9.10), while for parents without other children (n=9), the mean score was 3.27 (95% CI: -0.48, 7.02) (t= -2.44; P=0.02). Second, we compared parents currently caring for their child (n=33) and bereaved parents (n=14) across all 20 domains; only the *Caregiver Support at the End of Life* domain had significantly different scores at the P < 0.1 level (t = -2.07; P=0.04), where bereaved parents rated this domain 1.5 times as important as parents currently caring for their child. Third, mothers' (n=44) and fathers' (n=3) domain importance scores did not display any statistically significant differences (Table 3).

DISCUSSION

We conducted a DCE to prioritize 20 PPHC@Home domains based on quantitative parent-rated domain importance scores, and identified, through exploratory analysis, potential differences in how sub-groups of parents rated these domains. Several of our findings warrant discussion.

First, parents ranked *Physical Aspects of Care: Symptom Management* and *Psychological/Emotional Support for the Child* as the most important domains. This prioritization is unsurprising, given the proportion of children with serious illness who experience pain and other distressing physical symptoms, such as fatigue, reduced mobility, and constipation (2, 44-47), and psychological symptoms, such as sadness, worry, and anxiety.(2, 45, 48, 49) The relief of symptoms is often described by pediatric and adult patients and their family caregivers as a top concern and priority for quality

palliative and end-of-life care (12, 15, 50-53); yet, the assessment and management of pain and other symptoms has room for improvement for patients across ages, diagnoses, and settings.(13, 15, 49, 53-57) Inadequate pain and symptom management has been associated with child and family outcomes such as lower child health-related quality of life (QOL) (45, 58), long-term parental grief and distress (59, 60), and parents' more negative perceptions of care quality.(12, 13, 61)

Parents also ranked *Care Coordination* among the most important domains. Seriously-ill, medically-complex children often have significant care needs, necessitating care from a complex network of providers across numerous settings and institutions.(62) As a result, parents, providers, and health care leaders have identified care coordination as a significant need (26, 52, 63) and a priority for clinical practice, policy, and research.(25, 26, 64, 65) Notably, care coordination was added as a key theme in each domain in the most recent edition of the NCP Guidelines.(22) Care coordination has also been associated with care quality and child and family outcomes (e.g., QOL, symptom management, death in preferred location) in the home and community setting.(8, 19, 20, 62, 66-68)

The lowest-ranked domains in this study were *Spiritual and Religious Aspects of Care* and *Cultural Aspects of Care*. While we know that these care domains are critical components of effective pediatric palliative and hospice care (69-72), they have been found to be more important for some families than others.(54, 69) While we observed little individual-level variation in how parents ranked these domains in our sample (Figure 2), this observation could be attributed to the lack of sociodemographic heterogeneity of our sample. Another possible explanation is that when parents are “forced” to choose between domains, as in this discrete choice exercise, parents may

prioritize the management of their child's symptoms and the effective coordination of their child's care over spiritual or cultural aspects of care. A previous DCE study identified parents' beliefs about what they need to do to be a "good parent" to their seriously-ill child. This study, conducted with 200 parents of children with serious illness, similarly found that beliefs such as *Making my child feel loved* and *Focusing on my child's comfort* were ranked higher by parents than the belief *Focusing on my child's spiritual well-being*.⁽³⁴⁾ Finally, some children and families may receive support for their spiritual, religious, and cultural needs from community-based organizations or other social support networks and may simply not expect nor require this type of support from their PPHC@Home team.

We did observe potential differences in how subgroups of parents ranked the domains. In particular, we found that parents who had other children ranked the *Psychological and Emotional Aspects of Care for Sibling(s)* domain more than twice as important as parents who did not have other children. This finding is unsurprising in light of the documented needs of siblings of seriously-ill children (52, 56, 73, 74) and the significant stress parents face when balancing the care of their seriously-ill child with care of the child's siblings.^(75, 76) Given the proximity of siblings to their ill sibling's care in the home setting (74), further intervention-based research is needed to understand how this support may best be provided to siblings in the home setting in particular. We also observed differences in how bereaved parents and parents currently caring for their child rated the *Caregiver Support at the End of Life* domain. This is likely because parents who are actively caring for their child who is not yet at the end of life may not see this aspect of care as important when compared with other domains that may represent more immediate needs or concerns.

Finally, we did not observe significant differences in how mothers and fathers ranked the domains, although these rankings represent the views of only 3 fathers and, thus, precludes any conclusions to be drawn. While further exploration with larger samples of fathers is greatly needed in this area, previous research with larger samples of fathers have found that the major areas of problems, hopes, and goals related to a child's care were not significantly different between mothers and fathers (77), and that the type of support parents need to care for their child with serious illness at home may be the same, regardless of gender.(75)

This study has four limitations that warrant mention. First, the gender and sociodemographic characteristics of our sample were relatively homogenous, although our sample included parents of children with various clinical diagnoses who received care from different home-based models of care located in several geographic regions of the country. While the sociodemographic homogeneity is not surprising given issues with gender (78, 79) and minority (80) imbalance in PPC studies, as well as disparities in access to home-based hospice care in general by minority patients (81), additional research with larger and more socio-demographically-diverse samples in the home-based setting is critically needed to more fully understand parental priorities, particularly among underrepresented groups (e.g., fathers, racial and ethnic minorities, single parents, other cultural and religious groups) and within different models of PPHC@Home. Regardless of sociodemographic characteristics, we should keep in mind that the experience of individual families caring for a seriously-ill child in the home may be quite different, and parents may therefore have different priorities based on their unique experiences, as represented by the variation we observed within domain rankings (Figure 2).

Second, this study may not have included all important domains of PPHC@Home. Our PPHC@Home domains were, however, developed based on the NCP Guidelines for Quality Palliative Care (22), and were informed by additional PPC guidelines and literature (20, 25-28) and by a panel of PPC stakeholders. These steps reduced the likelihood that any critical domains were omitted.

Third, parents may have regarded a domain of care as less important if they never received effective care in that domain. We do note that, anecdotally, as part of this overall larger study, several bereaved parents recounted receiving less-than-effective aspects of their PPHC@Home experiences that they felt were very important to improve upon for future children and families. For example, several parents discussed a lack of adequate access to appropriately trained (e.g., pediatric and palliative/hospice care trained) nursing support in the home; yet, all of these parents acknowledged the critical role nursing care plays in good PPHC@Home.

Fourth and finally, because of the cross-sectional design of this study, we do not know how much individual parental ratings of importance may change over time. Future longitudinal studies will be important to examine trends in parents' priorities over the course of their child's illness. Our findings, however, build on and extend beyond findings from previous studies, providing quantitative evidence for the overall importance of excellent pain and symptom management, psychological and emotional support for seriously-ill children, and integrated, coordinated care for children with serious illness and their families in the home setting.(19, 66-68, 75, 82, 83)

CONCLUSION

The priorities for PPHC@Home among parents in this study represent a foundation for further exploration and conversation about how to prioritize finite health care resources, and where to focus care improvement efforts and future research, particularly in the home-based setting, for seriously-ill children and their families.

DISCLOSURE/CONFLICT OF INTEREST

The authors declare that they have no relevant financial relationships to disclose.

ACKNOWLEDGEMENTS

The authors would like to acknowledge the Justin Michael Ingerman Center for Palliative Care at the Children's Hospital of Philadelphia and the Courageous Parents Network, who facilitated the design and data collection phases of this study. In particular, we would like to thank Ms. Karen Crew and Ms. Dana Dombrowski for their guidance and effort on this project, and Dr. Russell Nye for his review of this manuscript. We would also like to acknowledge all of the clinicians, researchers, and parent advocates who shared their time and expertise with us. And finally, we would like to thank the parents who so generously gave their time and energy to participate in this project -- we are truly grateful for their willingness to share their perspectives with us.

FUNDING

This work was supported by the National Institute of Nursing Research of the National Institutes of Health under Award Number F31NR017554. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health. This work was additionally supported by the University of

Pennsylvania School of Nursing Office of Nursing Research's Student Research Grant,
the Independence Blue Cross Foundation's Nurses for Tomorrow Scholars Program,
and the Sawtooth Software Academic Grant.

BIBLIOGRAPHY

1. Feudtner C, Feinstein JA, Satchell M, Zhao H, Kang TI. Shifting place of death among children with complex chronic conditions in the United States, 1989-2003. *JAMA* 2007;297:2725-2732. doi: 10.1001/jama.297.24.2725
2. Feudtner C, Kang TI, Hexem KR, et al. Pediatric palliative care patients: A prospective multicenter cohort study. *Pediatr* 2011;127:1094-1101. doi: 10.1542/peds.2010-3225
3. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980-1998. *Pediatr* 2002;109:656-660. doi: 10.1542/peds.109.4.656
4. Nageswaran S, Radulovic A, Anania A. Transitions to and from the acute inpatient care setting for children with life-threatening illness. *Pediatr Clin North Am* 2014;61:761-783. doi: 10.1016/j.pcl.2014.04.008
5. Feudtner C, Womer J, Augustin R, et al. Pediatric palliative care programs in children's hospitals: A cross-sectional national survey. *Pediatr* 2013;132:1063-1070. doi: 10.1542/peds.2013-1286
6. Friebert S, Williams C. NHPCO'S Facts and Figures: Pediatric Palliative and Hospice Care in America. In: Alexandria, VA: NHPCO, 2015.
7. Kaye E, Rubenstein J, Levine D, et al. Pediatric palliative care in the community. *CA Cancer J Clin* 2015;65:315-333. doi: 10.3322/caac.21280
8. Boyden JY, Curley MAQ, Deatrick JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage* 2018;55:117-131. doi: 10.1016/j.jpainsymman.2017.04.017
9. Lindley L, Mark B, Lee SYD. Providing hospice care to children and young adults: A descriptive study of end-of-life organizations. *J Hosp Palliat Nurs* 2009;11:315-323. doi: 10.1097/NJH.0b013e3181bcfd62
10. Carroll JM, Torkildson C, Winsness JS. Issues related to providing quality pediatric palliative care in the community. *Pediatr Clin N Amer* 2007;54:813-27, xiii. doi: 10.1016/j.pcl.2007.06.002
11. Lindley LC, Kieim-Malpass J. Quality of paediatric hospice care for children with and without multiple complex chronic conditions. *Int J Palliat Nurs* 2017;23:230-237. doi: 10.12968/ijpn.2017.23.5.230
12. Donnelly JP, Huff SM, Lindsey ML, McMahon KA, Schumacher JD. The needs of children with life-limiting conditions: A healthcare-provider-based model. *Amer J Hosp Palliat Med* 2005;22:259-267. doi: 10.1177/104990910502200406
13. Contro N, Larson J, Scofield S, Sourkes B, Cohen H. Family perspectives on the quality of pediatric palliative care. *Arch Pediatr Adolesc Med* 2002;156:14-19. doi: 10.1001/archpedi.156.1.14
14. 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
15. Meyer EC, Ritholz MD, Burns JP, Truog RD. Improving the quality of end-of-life care in the pediatric intensive care unit: Parents' priorities and recommendations. *Pediatr* 2006;117:649-57. doi: 10.1542/peds.2005-0144

16. Weidner NJ, Cameron M, Lee RC, et al. End-of-life care for the dying child: What matters most to parents. *J Palliat Care* 2011;27:279-286. doi: <https://doi.org/10.1177/082585971102700404>
17. Andresen EM, Seecharan GA, Toce SS. Provider perceptions of child deaths. *Arch Pediatr Adolesc Med* 2004;158:430-5. doi: 10.1001/archpedi.158.5.430
18. Malcolm C, Knighting K, Forbat L, Kearney N. Prioritisation of future research topics for children's hospice care by its key stakeholders: A Delphi study. *Palliat Med* 2009;23:398-405. doi: 10.1177/0269216309104061
19. Thienprayoon R, Grosseohme D, Humphrey L, et al. "There's Just No Way to Help, and They Did." Parents name Compassionate Care as a new domain of quality in pediatric home-based hospice and palliative care. *J Palliat Med* 2020;23:767-776. doi: 10.1089/jpm.2019.0418
20. Thienprayoon R, Mark MSJ, Grosseohme D. Provider-prioritized domains of quality in pediatric home-based hospice and palliative care: A study of the Ohio Pediatric Palliative Care and End-of-Life Network. *J Palliat Med* 2018;31:290-296. doi: 10.1089/jpm.2017.0333
21. Widger KA, Wilkins K. What are the key components of quality perinatal and pediatric end-of-life care? A literature review. *J Palliat Care* 2004;20:105-112. doi: <https://doi.org/10.1177/082585970402000207>
22. National Consensus Project for Quality Palliative Care. *Clinical Practice Guidelines for Quality Palliative Care* 4th ed. Richmond, VA: National Coalition for Hospice and Palliative Care, 2018. Available from: <https://www.nationalcoalitionhpc.org/ncpl/>. Accessed June 5, 2020.
23. Trevelyan EG, Robinson PN. Delphi methodology in health research: How to do it? *Euro J Integ Med* 2015;7:423-428. doi: 10.1016/j.eujim.2015.07.002
24. Boyden J, Ersek M, Deatrick J, et al. Developing a parent-reported measure of experiences with home-based pediatric palliative and hospice care: A multi-method, multi-stakeholder approach. In development.
25. Feudtner C, Friebert S, Jewell J. Pediatric palliative care and hospice care commitments, guidelines, and recommendations. *Pediatr* 2013;132:966-972. doi: 10.1542/peds.2013-2731
26. Field MJ, Behrman RE. *When Children Die: Improving Palliative and End-of-Life care for Children and their Families*. Washington, DC: The National Academies Press, 2003:419-420. Available from: <https://www.nap.edu/catalog/10390/when-children-die-improving-palliative-and-end-of-life-care>. Accessed June 5, 2020.
27. National Hospice and Palliative Care Organization (NHPCO). *Standards of practice for pediatric palliative care*. Alexandria, VA: NHPCO, 2019. Available from: https://www.nhpc.org/wp-content/uploads/2019/07/Pediatric_Standards.pdf. Accessed June 12, 2020.
28. Widger K, Brennenstuhl S, Duc J, Tourangeau A, Rapoport A. Factor structure of the Quality of Children's Palliative Care Instrument (QCPCI) when completed by parents of children with cancer. *BMC Palliat Care* 2019;18:23. doi: 10.1186/s12904-019-0406-9
29. Courageous Parents Network. Available from: <https://courageousparentsnetwork.org/>. Accessed April 23, 2020.
30. Gomes B, de Brito M, Sarmiento VP, et al. Valuing attributes of home palliative care with service users: A pilot discrete choice experiment. *J Pain Symptom Manage* 2017;54:973-985. doi: <https://doi.org/10.1016/j.jpainsymman.2017.05.005>
31. Huynh E, Coast J, Rose J, Kinghorn P, Flynn T. Values for the ICECAP-Supportive Care Measure (ICECAP-SCM) for use in economic evaluation at end of life. *Soc Sci Med* 2017;189:114-128. doi: <https://doi.org/10.1016/j.socscimed.2017.07.012>

32. Mangham LJ, Hanson K, McPake B. How to do (or not to do) ... Designing a discrete choice experiment for application in a low-income country. *Health Policy Plan* 2009;24:151-158. doi: 10.1093/heapol/czn047
33. Ryan M, Gerard K, Amaya-Amaya M. *Using Discrete Choice Experiments to Value Health and Health Care*, The Netherlands: Springer, 2008.
34. Feudtner C, Walter JK, Faerber JA, et al. Good-parent beliefs of parents of seriously ill children. *JAMA Pediatr* 2015;169:39-47. doi: 10.1001/jamapediatrics.2014.2341
35. Sawtooth Software. Maximum difference scaling: Improved measures of importance and preference for segmentation. 2003. Available from: <https://www.sawtoothsoftware.com/support/technical-papers/maxdiff-best-worst-scaling/maximum-difference-scaling-improved-measures-of-importance-and-preference-for-segmentation-2003>. Accessed June 5, 2020.
36. Louviere JJ, Flynn TN, Carson RT. Discrete choice experiments are not conjoint analysis. *J Choice Model* 2010;3:57-72. doi: 10.1016/s1755-5345(13)70014-9
37. Chrazan K, Orme B. *Applied MaxDiff: A practitioner's guide to Best-Worst Scaling* Provo, UT: Sawtooth Software, Inc., 2019.
38. Sawtooth Software. Aggregate score estimation via logit analysis. 2018. Available from: https://www.sawtoothsoftware.com/help/lighthouse-studio/manual/maxdiff_aggregate_logit_analysis.html. Accessed June 5, 2020.
39. Sawtooth Software. The MaxDiff System technical paper. 2013. Available from: <https://www.sawtoothsoftware.com/support/technical-papers/maxdiff-best-worst-scaling/maxdiff-technical-paper-2013>. Accessed June 5, 2020.
40. Agresti A, Finlay B. *Statistical Methods for the Social Sciences* 4th ed. Upper Saddle River, NJ: Pearson Prentice Hall, 2009.
41. Bekker-Grob EWd, Donkers B, Jonker MF, Stolk EA. Sample size requirements for discrete-choice experiments in healthcare: A practical guide. *Patient* 2015;8:373-384. doi: 10.1007/s40271-015-0118-z
42. Lancsar E, Louviere J. Conducting discrete choice experiments to inform healthcare decision making. *Pharmacoeconomics* 2008;26:661-677. doi: 10.2165/00019053-200826080-00004
43. Flynn TN, Louviere JJ, Peters TJ, Coast J. Best-worst scaling: What it can do for health care research and how to do it. *J Health Econ* 2007;26:171-189. doi: 10.1016/j.jhealeco.2006.04.002
44. Jalmsell L, Kreicbergs U, Onelöv E, Steineck G, Henter J-I. Symptoms affecting children with malignancies during the last month of life: A nationwide follow-up. *Pediatr* 2006;117:1314-1320. doi: 10.1542/peds.2005-1479
45. Rosenberg AR, Orellana L, Ullrich C, et al. Quality of life in children with advanced cancer: A report from the PediQUEST Study. *J Pain Symptom Manage* 2016;52:243-53. doi: 10.1016/j.jpainsymman.2016.04.002
46. Schmidt P, Otto M, Hechler T, et al. Did increased availability of pediatric palliative care lead to improved palliative care outcomes in children with cancer? *J Palliat Med* 2013;16:1034-1039. doi: 10.1089/jpm.2013.0014
47. Wolfe J, Grier HE, Klar N. Symptoms and suffering at the end of life in children with cancer. *New Eng J Med* 2000;342:326-333. doi: 10.1056/NEJM200002033420506
48. Kersun LS, Shemesh E. Depression and anxiety in children at the end of life. *Pediatr Clin North Am* 2007;54:691-708, xi. doi: 10.1016/j.pcl.2007.06.003

49. Theunissen JM, Hoogerbrugge PM, Achterberg Tv, et al. Symptoms in the palliative phase of children with cancer. *Pediatr Blood Cancer* 2007;49:160-165. doi: 10.1002/pbc.21042
50. Heyland DK, Dodek P, Rocker G, et al. What matters most in end-of-life care: Perceptions of seriously ill patients and their family members. *CMAJ* 2006;174:627-33. doi: 10.1503/cmaj.050626
51. 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;12:15-55. doi: 10.1007/s40271-018-0333-5
52. Stevenson M, Achille M, Lugasi T. Pediatric palliative care in Canada and the United States: A qualitative metasummary of the needs of patients and families. *J Palliat Med* 2013;16:566-577. doi: 10.1089/jpm.2011.0076
53. Vohra JU, Brazil K, Hanna S, Abelson J. Family perceptions of end-of-life care in long-term care facilities. *J Palliat Care* 2004;20:297-302. doi: 10.1177/082585970402000405
54. Meyer EC, Burns JP, Griffith JL, Truog RD. Parental perspectives on end-of-life care in the pediatric intensive care unit. *Crit Care Med* 2002;30:226-231. doi: 10.1097/00003246-200201000-00032
55. Singer P, Martin D, Kelner M. Quality end-of-life care: Patients' perspectives. *JAMA* 1999;281:163-168. doi: 10.1001/jama.281.2.163
56. Thienprayoon R, Marks E, Funes M, et al. Perceptions of the pediatric hospice experience among English- and Spanish-speaking families. *J Palliat Med* 2016;19:30-41. doi: 10.1089/jpm.2015.0137
57. Zheng NT, Li Q, Hanson LC, et al. Nationwide quality of hospice care: Findings from the Centers for Medicare & Medicaid Services Hospice Quality Reporting Program. *J Pain Symptom Manage* 2018;55:427-432. doi: 10.1016/j.jpainsymman.2017.09.016
58. Meeske KA, Patel SK, Palmer SN, Nelson MB, Parow AM. Factors associated with health-related quality of life in pediatric cancer survivors. *Pediatr Blood Cancer* 2007;49:298-305. doi: 10.1002/pbc.20923
59. Kreicbergs U, Valdimarsdottir U, Onelov E, et al. Care-related distress: A nationwide study of parents who lost their child to cancer. *J Clin Oncol* 2005;23:9162-9171. doi: 10.1200/JCO.2005.08.557
60. van der Geest IMM, Darlington A, Streng IC, et al. Parents' experiences of pediatric palliative care and the impact on long-term parental grief. *J Pain Symptom Manage* 2014;47:1043-1053. doi: 10.1016/j.jpainsymman.2013.07.007
61. Homer CJ, Marino B, Cleary PD, et al. Quality of care at a children's hospital: the parent's perspective. *Arch Pediatr Adolesc Med* 1999;153:1123-9. doi: 10.1001/archpedi.153.11.1123
62. Cohen E, Lacombe-Duncan A, Spalding K, et al. Integrated complex care coordination for children with medical complexity: A mixed-methods evaluation of tertiary care-community collaboration. *BMC Health Serv Res* 2012;12:366. doi: 10.1186/1472-6963-12-366
63. Cady RG, Belew JLJC. Parent perspective on care coordination services for their child with medical complexity. *Children (Basel)* 2017;4:45. doi: 10.3390/children4060045
64. Baker JN, Levine DR, Hinds PS, et al. Research priorities in pediatric palliative care. *J Pediatr* 2015;167:467-470. doi: 10.1016/j.jpeds.2015.05.002
65. Lipkin P, Alexander J, Cartwright J. Care coordination in the medical home: Integrating health and related systems of care for children with special health care needs. *Pediatr* 2005;116:1238-1244. doi: 10.1542/peds.2005-2070

66. 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;18:143-150. doi: 10.1089/jpm.2014.0285
67. Bona K, Bates J, Wolfe J. Massachusetts' Pediatric Palliative Care Network: Successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med* 2011;14:1217-1223. doi: 10.1089/jpm.2011.0070
68. Knapp CA, Madden VL, Curtis CM, et al. Partners in Care: Together for Kids: Florida's model of pediatric palliative care. *J Palliat Med* 2008;11:1212-1220. doi: 10.1089/jpm.2008.0080
69. Hexem KR, Mollen CJ, Carroll KW, Lanctot DA, Feudtner C. How parents of children receiving pediatric palliative care use religion, spirituality, or life philosophy in tough times. *J Palliat Med* 2011;14:39-44. doi: 10.1089/jpm.2010.0256
70. Wiener L, McConnell DG, Latella L, Ludi E. Cultural and religious considerations in pediatric palliative care. *Palliat Support Care* 2013;11:47-67. doi: 10.1017/S1478951511001027
71. Davies B, Brenner P, Orloff S, Sumner L, Worden W. Addressing spirituality in pediatric hospice and palliative care. *J Palliat Care* 2002;18:59-67. doi: 10.1177/082585970201800109
72. Davies B, Contro N, Larson J, Widger K. Culturally-sensitive information-sharing in pediatric palliative care. *Pediatr* 2010;125:e859-65. doi: 10.1542/peds.2009-0722
73. Gaab E, Owens G, MacLeod R. Siblings caring for and about pediatric palliative care patients. *J Palliat Med* 2014;17:62-67. doi: 10.1089/jpm.2013.0117
74. Eaton Russell C, Widger K, Beaune L, et al. Siblings' voices: A prospective investigation of experiences with a dying child. *Death Stud* 2018;42:184-194. doi: 10.1080/07481187.2017.1334009
75. Verberne LM, Kars MC, Schouten-van Meeteren AY, et al. Aims and tasks in parental caregiving for children receiving palliative care at home: a qualitative study. *Eur J Pediatr* 2017;176:343-354. doi: 10.1007/s00431-016-2842-3
76. Mooney-Doyle K, Deatrck JA. Parenting in the face of childhood life-threatening conditions: The ordinary in the context of the extraordinary. *Palliat Support Care* 2016;14:187-198. doi: 10.1017/S1478951515000905
77. Hill DL, Miller VA, Hexem KR, et al. Problems and hopes perceived by mothers, fathers and physicians of children receiving palliative care. *Health Expect* 2015;18:1052-1065. doi: 10.1111/hex.12078
78. Hinds PS, Burghen EA, Pritchard M. Conducting end-of-life studies in pediatric oncology. *West J Nurs Res* 2007;29:448-465. doi: 10.1177/0193945906295533
79. Macdonald ME, Chilibeck G, Affleck W, Cadell S. Gender imbalance in pediatric palliative care research samples. *Palliat Med* 2010;24:435-44. doi: <https://dx.doi.org/10.1177/0269216309354396>
80. Knapp CA, Madden VL, Curtis C, Sloyer PJ, Shenkman EA. Assessing non-response bias in pediatric palliative care research. *Palliat Med* 2010;24:340-7. doi: <https://dx.doi.org/10.1177/0269216309351466>
81. Cohen LL. Racial/ethnic disparities in hospice care: A systematic review. *J Palliat Med* 2008;11:763-768. doi: 10.1089/jpm.2007.0216
82. Gans D, Hadler MW, Chen X, et al. Impact of a pediatric palliative care program on the caregiver experience. *J Hosp Palliat Nurs* 2015;17:559-565. doi: 10.1097/NJH.0000000000000203

83. Vollenbroich R, Duroux A, Grasser M, et al. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med* 2012;15:294-300. doi: 10.1089/jpm.2011.0196

TABLES AND FIGURES

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care in the Home

Table 2: Demographic and Clinical Characteristics of Parents and Children

Table 3: Sub-group Domain Score Comparisons: 2-Sample Independent t-tests

Figure 1: Average Importance Scores Across Domains

Figure 2: Individual-level Variation Across Domain Importance Scores

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care in the Home

Domains	Description
Communication between family and care team	Care team communicates with the child and family to make sure that the care provided meets the child's and family's values, preferences, goals, and needs
Relationship between family and care team	Relationship between care team and family is built on trust, respect, and advocacy for the child's and family's needs
Knowledge and skills of care team providers	Care team members have the necessary education and training to provide high quality palliative care for children and families
Access to care	Care team provides access to palliative and hospice care to the child and family 24 hours a day, 7 days a week
Physical care: Communication	Care team provides information about treatments for child's pain and other physical symptoms (for example, nausea, fatigue, constipation)
Physical care: Symptom management	Care team assesses and manages pain and other physical symptoms and side effects based on the best available medical evidence
Psychological and emotional aspects of care (<i>Child, Parents, Siblings, and Extended Social Network</i>) ^A	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the child, family, and family's community based on the best available medical evidence
Practical aspects of care	Care team provides the family with assistance and resources for dealing with financial and insurance related issues
Social aspects of care (<i>Child, Parents</i>) ^A	Care team helps with social issues to meet child-family needs, promote child-family goals, and maximize child-family strengths and well-being (examples include helping family maintain and strengthen their support network; help family develop strategies to balance caregiving, work, and family needs)
Spiritual and religious aspects of care	Care team assists with religious and spiritual rituals or practices as desired by the child and family
Cultural aspects of care	Care team respects the child's and family's cultural beliefs and language preferences

Communication at the end of life	Care team works with the child and family to develop and implement a care plan to address actual or potential symptoms at the end of life
Caregiver support at the end of life	Care team meets the emotional, spiritual, social, and cultural needs of families at the end of life (for example, preparing parents for the end of life)
Ethical and legal aspects of care	<p>Child's and family's goals, preferences, and choices are respected within the limits of state and federal law, current standards of medical care, and professional standards of practice.</p> <p>These goals/preferences/choices are documented and shared with all professionals involved in the child's care</p>
Coordination of care	Care team works to make sure that when there are transfers between healthcare settings and providers, that there is timely and thorough communication of the child's/family's goals, preferences, values, and clinical information to ensure continuity of care and seamless follow-up (for example, getting needed services, arranging for medical equipment)
Continuity of care	Care team works to make sure that the delivery of palliative and hospice care is seamless across care settings and providers (for example, the same providers work with family)

^ASeparate sub-domain for each group

Note: These domains are based on the National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) (22), which were further adapted using pediatric palliative care specific guidelines and the literature (20, 25-28) and informed by a panel of PPC stakeholders (providers and parent advocates).

Table 2: Demographic and Clinical Characteristics of Parents and Children

Parents' Characteristics (n = 47)		No. (%)
Parent type	Mother	44 (93.6%)
	Father	3 (6.4%)
Age	<i>Mean / SD</i>	42.6 (8.5)
Race	White	42 (89.4%)
	Black or African American	1 (2.1%)
	More than one race/Other	3 (6.4%)
	Prefer not to answer	1 (2.1%)
Ethnicity	Non-Hispanic	43 (91.5%)
	Hispanic	3 (6.4%)
	Prefer not to answer	1 (2.1%)
Religious Preference	Christian (Protestant, Catholic, Mormon, etc.)	31 (66.0%)
	Jewish	6 (12.8%)
	Muslim	0
	Buddhist	1 (2.1%)
	Hindu	1 (2.1%)
	Atheist	3 (6.4%)
	Agnostic	2 (4.3%)
	Prefer not to answer	3 (6.4%)
	Highest Education Level Completed	Grade school
High school / general educational development		2 (4.3%)
Trade / technical / vocational		4 (8.5%)
Associates / Professional		8 (17.0%)
College		19 (40.4%)
Graduate school		13 (27.7%)
Relationship Status	Married / partnered	41 (87.2%)
	Separated / divorced / Widowed	6 (12.8%)

Number of Other Children	0	11 (23.4%)
	1-3	35 (74.4%)
	4 or more	1 (2.1%)
Employment Status	Full time	23 (48.9%)
	Part time	5 (10.6%)
	Not employed outside of the home	17 (36.2%)
	Prefer not to answer	2 (4.3%)
Bereavement Status	Bereaved	14 (29.8%)
	Currently caring for child at home	33 (70.2%)
Affiliation	CHOP	16 (34.0%)
	CPN	31 (66.0%)
Children's Characteristics (n = 45)		No. (%)
Age	1 year or less	8 (17.8%)
	2-4 years	9 (20.0%)
	5-9 years	5 (11.1%)
	10-18 years	17 (37.8%)
	19-25 years	6 (13.3%)
Gender	Female	21 (46.7%)
	Male	24 (53.3%)
Race	White	37 (82.2%)
	Black or African American	2 (4.4%)
	More than one race/Other	5 (11.1%)
	Prefer not to answer	1 (2.2%)
Ethnicity	Non-Hispanic	39 (86.7%)
	Hispanic	4 (8.9%)
	Prefer not to answer	2 (4.4%)

<i>(Note: not mutually exclusive; thus, the % does not sum to 100%)</i>	Primary complex chronic condition	Cardiovascular	10 (22.2%)
		Gastrointestinal	4 (8.9%)
		Genetic or congenital	22 (48.9%)
		Hematologic or immunologic	4 (8.9%)
		Malignancy	5 (11.1%)
		Metabolic	10 (22.2%)
		Neuromuscular, neurologic, or mitochondrial	23 (51.1%)
		Respiratory	6 (13.3%)
		Other/Unknown	1 (2.2%)
Primary care team (hospice v. palliative care)	Hospice	19 (42.2%)	
	Palliative Care	24 (53.3%)	
	Unknown/Not sure	2 (4.4%)	
Length of time receiving home-based palliative or hospice care	Less than 1 month	5 (11.1%)	
	1 to 3 months	5 (11.1%)	
	4 to 6 months	7 (15.6%)	
	7 to 12 months	5 (11.1%)	
	1 to 2 years	8 (17.8%)	
	More than 2 years	15 (33.3%)	

Note: Unless otherwise noted, cell entries are percentages. Percentages are within each demographic or clinical characteristic variable.

Table 3: Sub-group Domain Score Comparisons: 2-Sample Independent t-tests

Domain	Sub-group	Mean	[95% CI]	p Value
Single Domain: Parents without other children (n=9) v. Parents with other children (n=38)				
Psychological and Emotional Aspects of Care for the Sibling(s)	No siblings	3.27	[-0.48, 7.02]	0.02 ^A
	One or more siblings	7.55	[6.01, 9.10]	
Across All Domains: Parents currently caring for child (n=33) v. bereaved parents (n=14)				
Communication between family and care team	Currently caring for child	6.08	[4.99, 7.16]	0.98
	Bereaved	6.05	[4.00, 8.10]	
Relationship between family and care team	Currently caring for child	5.53	[4.24, 6.82]	0.45
	Bereaved	4.61	[2.21, 7.00]	
Knowledge and skills of care team providers	Currently caring for child	6.63	[5.12, 8.13]	0.81
	Bereaved	6.94	[4.93, 8.96]	
Access to care	Currently caring for child	6.55	[5.32, 7.79]	0.29
	Bereaved	7.77	[5.56, 9.97]	
Physical care: Communication	Currently caring for child	5.39	[4.46, 6.31]	0.62
	Bereaved	4.96	[3.31, 6.62]	
Physical care: Symptom management	Currently caring for child	9.33	[8.05, 10.60]	0.28
	Bereaved	10.50	[8.92, 12.07]	
Psychological and emotional aspects of care: Parent	Currently caring for child	5.70	[4.42, 6.99]	0.97
	Bereaved	5.75	[3.98, 7.53]	
Psychological and emotional aspects of care: Child	Currently caring for child	8.78	[7.23, 10.32]	0.64
	Bereaved	9.43	[6.92, 11.95]	

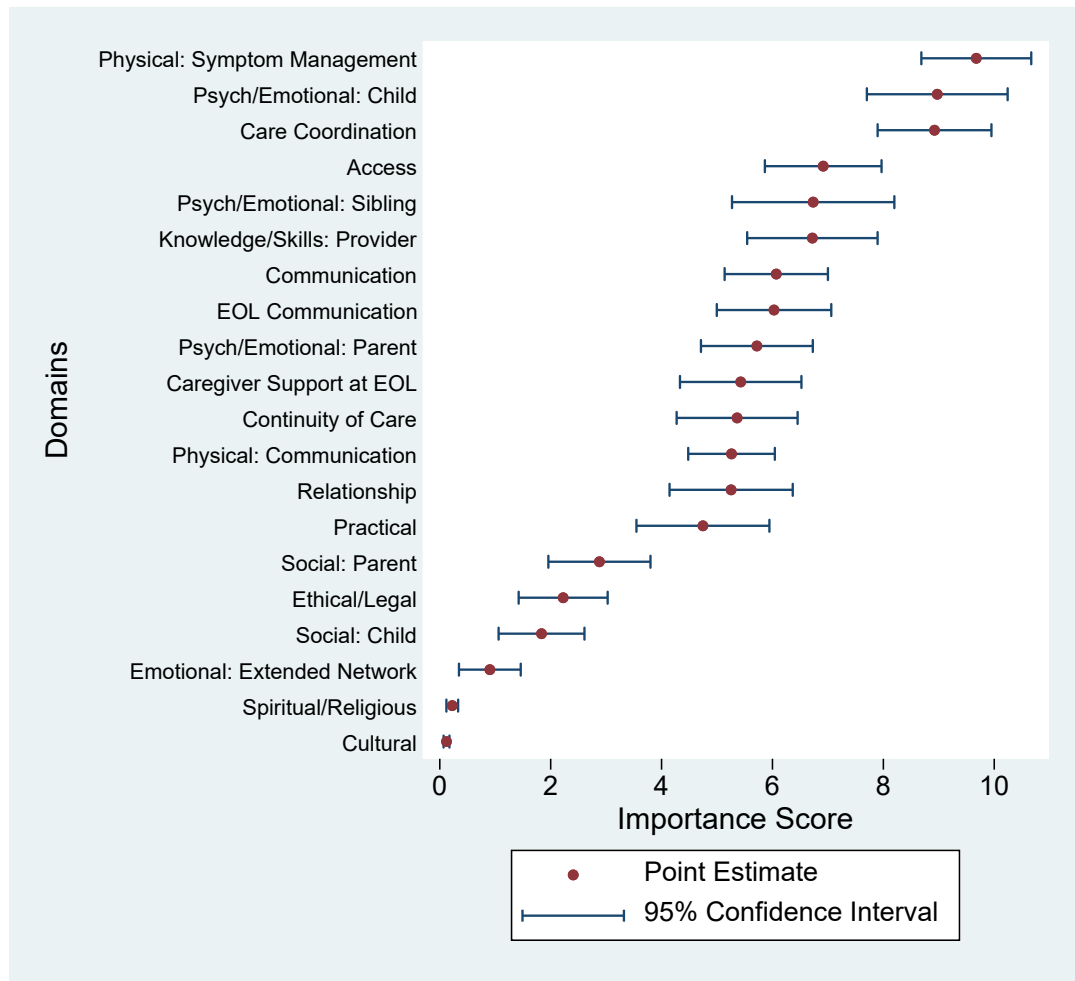
Psychological and emotional aspects of care: Sibling(s)	Currently caring for child	6.84	[4.99, 8.69]	0.83
	Bereaved	6.49	[3.86, 9.13]	
Psychological and emotional aspects of care: Extended Social Network	Currently caring for child	1.04	[0.30, 1.79]	0.45
	Bereaved	0.57	[-0.18, 1.33]	
Practical aspects of care	Currently caring for child	5.21	[3.76, 6.66]	0.24
	Bereaved	3.65	[1.36, 5.95]	
Social aspects of care: Parents	Currently caring for child	3.18	[2.02, 4.35]	0.31
	Bereaved	2.16	[0.60, 3.73]	
Social aspects of care: Child	Currently caring for child	2.02	[0.96, 3.08]	0.47
	Bereaved	1.40	[0.56, 2.24]	
Spiritual and religious aspects of care	Currently caring for child	0.20	[0.09, 0.31]	0.48
	Bereaved	0.28	[0.01, 0.56]	
Cultural aspects of care	Currently caring for child	0.14	[0.07, 0.22]	0.19
	Bereaved	0.07	[0.03, 0.11]	
Communication at the end of life	Currently caring for child	5.80	[4.43, 7.17]	0.51
	Bereaved	6.56	[5.08, 8.04]	
Caregiver support at the end of life	Currently caring for child	4.72	[3.34, 6.10]	0.04 ^A
	Bereaved	7.10	[5.47, 8.72]	
Ethical and legal aspects of care	Currently caring for child	2.04	[1.09, 3.00]	0.49
	Bereaved	2.65	[0.99, 4.31]	
Coordination of care	Currently caring for child	8.99	[7.75, 10.23]	0.84
	Bereaved	8.76	[6.67, 10.85]	

Continuity of care	Currently caring for child	5.82	[4.43, 7.22]	0.20
	Bereaved	4.28	[2.56, 5.99]	
Across All Domains: Mothers (n=44) v. Fathers (n=3)				
Communication between family and care team	Mothers	6.10	[5.13, 7.07]	0.77
	Fathers	5.55	[-3.12, 14.21]	
Relationship between family and care team	Mothers	5.13	[3.97, 6.29]	0.40
	Fathers	7.07	[-1.87, 16.02]	
Knowledge and skills of care team providers	Mothers	6.72	[5.51, 7.93]	0.99
	Fathers	6.73	[-6.81, 20.28]	
Access to care	Mothers	6.99	[5.90, 8.08]	0.60
	Fathers	5.84	[-3.88, 15.56]	
Physical care: Communication	Mothers	5.10	[4.30, 5.91]	0.12
	Fathers	7.57	[1.95, 13.19]	
Physical care: Symptom management	Mothers	9.65	[8.62, 10.68]	0.28
	Fathers	10.08	[-.11, 20.26]	
Psychological and emotional aspects of care: Parent	Mothers	5.74	[4.67, 6.80]	0.89
	Fathers	5.44	[-1.23, 12.11]	
Psychological and emotional aspects of care: Child	Mothers	8.75	[7.42, 10.08]	0.17
	Fathers	12.29	[10.14, 14.44]	
Psychological and emotional aspects of care: Sibling(s)	Mothers	6.80	[5.29, 8.31]	0.72
	Fathers	5.73	[-9.79, 21.25]	
Psychological and emotional aspects of care: Extended Social Network	Mothers	0.91	[0.32, 1.50]	0.97
	Fathers	0.86	[-2.29, 4.01]	
Practical aspects of care	Mothers	4.82	[3.57, 6.06]	0.64
	Fathers	3.67	[-7.66, 15.01]	
Social aspects of care: Parents	Mothers	3.01	[2.04, 3.99]	0.27
	Fathers	0.92	[-0.11, 1.94]	

Social aspects of care: Child	Mothers	1.79	[0.98, 2.59]	0.65
	Fathers	2.52	[-4.44, 9.48]	
Spiritual and religious aspects of care	Mothers	0.24	[0.12, 0.35]	0.43
	Fathers	0.06	[-0.01, 0.14]	
Cultural aspects of care	Mothers	0.13	[0.07, 0.18]	0.40
	Fathers	0.04	[-0.01, 0.08]	
Communication at the end of life	Mothers	6.09	[5.01, 7.18]	0.63
	Fathers	5.06	[-2.87, 13.00]	
Caregiver support at the end of life	Mothers	5.50	[4.36, 6.64]	0.60
	Fathers	4.33	[-5.20, 13.87]	
Ethical and legal aspects of care	Mothers	2.34	[1.50, 3.19]	0.26
	Fathers	0.49	[-0.88, 1.85]	
Coordination of care	Mothers	8.89	[7.81, 9.96]	0.78
	Fathers	9.48	[1.41, 17.55]	
Continuity of care	Mothers	5.30	[4.20, 6.41]	0.67
	Fathers	6.27	[-7.75, 20.29]	

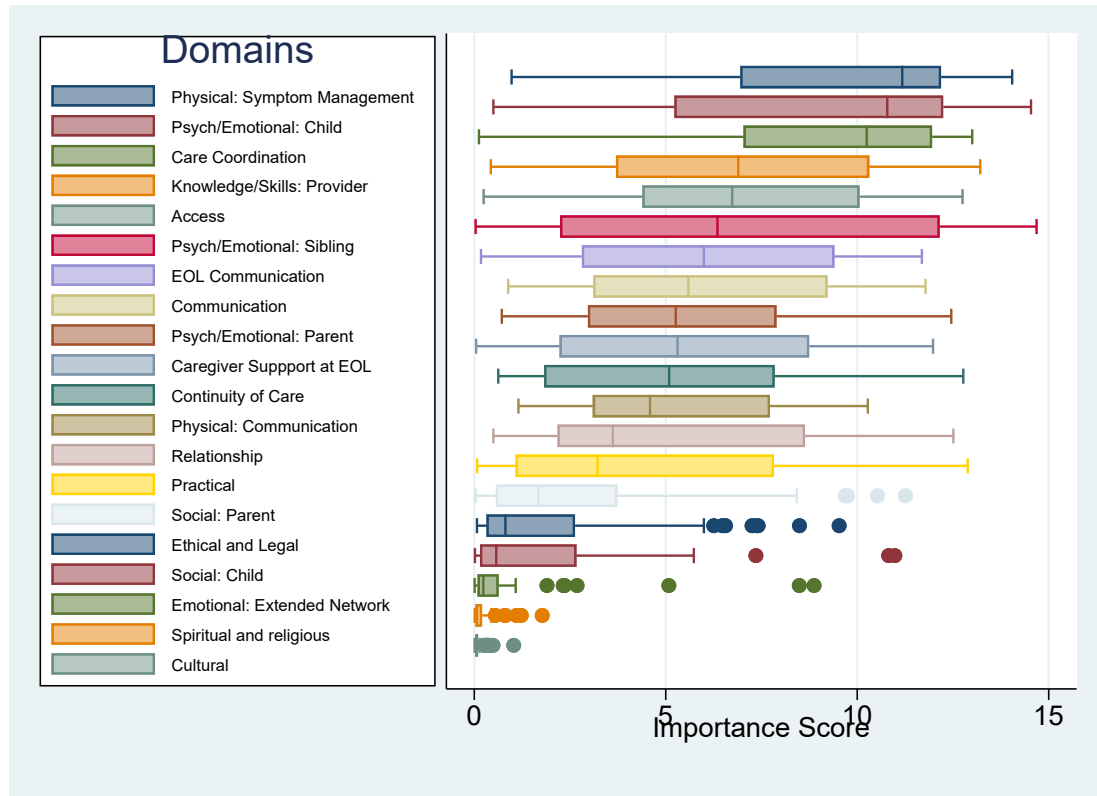
^ASignificant at $P < 0.1$ level

Figure 1: Average Importance Scores Across Domains



Note: This figure contains the point estimates of the mean importance score for each domain. The 95% confidence interval around each point estimate represent our level of confidence that the interval captures the true point estimate.

Figure 2: Individual-Level Variation Across Domain Importance Scores



Note: This graph presents the interquartile range (IQR) of domains, which represents the spread of the middle half of the scores in each domain. The line within each box represents the median, the left edge of the box represents the 25th percentile, and the right edge represents the 75th percentile of scores. The whiskers extending out of the box represents minimum and maximum scores, except for outliers (defined as more than 1.5 IQR beyond the 25th and 75th quartiles), which are represented by the dots.(40)

CHAPTER 3: Developing a family-reported measure of experiences with home-based pediatric palliative and hospice care: A multi-method, multi-stakeholder approach (Paper 2)

Jackelyn Y. Boyden^{1^}

Chris Feudtner^{2,3}

Janet A. Deatrick¹

Kimberley Widger^{4,5}

Gwenn LaRagione²

Blyth Lord⁶

Mary Ersek^{1,3,7}

¹ School of Nursing, University of Pennsylvania, Philadelphia PA

²Children's Hospital of Philadelphia, Philadelphia, PA

³University of Pennsylvania Perelman School of Medicine, Philadelphia PA

⁴Lawrence S. Bloomberg Faculty of Nursing, University of Toronto, Toronto ON Canada

⁵Hospital for Sick Children, Toronto ON Canada

⁶Courageous Parents Network

⁷Corporal Michael J. Crescenz VA Medical Center, Philadelphia PA

Submitted to *BMC Palliative Care* (July 2020)

ABSTRACT

Background: Many children with serious illnesses are receiving palliative and end-of-life care from pediatric palliative and hospice care teams at home (PPHC@Home). Despite the growth in PPHC@Home, no standardized measures exist to evaluate whether PPHC@Home provided in the U.S. meets the needs and priorities of children and their families.

Methods: We developed and conducted a preliminary evaluation of a family-reported measure of PPHC@Home experiences using a multi-method, multi-stakeholder approach. Our instrument development process consisted of four phases. Item identification and development (Phase 1) involved a comprehensive literature search of existing instruments, guidelines, standards of practice, and PPHC@Home outcome studies, as well as guidance from a PPHC stakeholder panel. Phase 2 involved the initial item prioritization and reduction using a discrete choice experiment (DCE) with PPHC professionals and parent advocates. Phase 3 involved a second DCE with bereaved parents and parents currently receiving care for their child to further prioritize and winnow the items to a set of the most highly-valued items. Finally, we conducted cognitive interviews with parents to provide information about the content validity and clarity of the newly-developed instrument (Phase 4).

Results: Items were compiled predominantly from three existing instruments. Phase 2 participants included 34 PPHC providers, researchers, and parent advocates; Phase 3 participants included 47 parents; and Phase 4 participants included 11 parents. At the completion of Phase 4, the Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home) Measure contains 22 of the most highly-valued items for evaluating PPHC@Home. These items include “*The care team treats my child’s physical symptoms so that my child has as good a quality of life as possible*”, “*I have regular access to on-call services from our care team*”, and “*The nurses have the knowledge, skills, and experience to support my child’s palliative or hospice care at home.*”

Conclusions: The EXPERIENCE@Home Measure is the first known to specifically measure family-reported experiences with PPHC@Home in the U.S. Future work will include formal psychometric evaluation with a larger sample of parents, as well as evaluation of the clinical utility of the instrument with PPHC@Home teams.

Keywords: pediatric palliative care, pediatric hospice care, home-based care, family experience, instrument development

BACKGROUND

Children with serious illnesses are generally living longer (1-3) and are increasingly being cared for by their families at home (1, 4, 5) with the support of pediatric palliative and hospice care teams.(1, 6, 7) Pediatric palliative and hospice care at home, hereafter referred to as PPHC@Home, is palliative and hospice care provided primarily outside of the hospital, often integrating the care provided by different services in the home, outpatient, hospital, and hospice settings.(8, 9) PPHC@Home supports children and families by providing a wide spectrum of services, including pain and symptom management, psychosocial and emotional support for the child and family, on-call services, expressive and other therapies, and care coordination across medical and social service providers and institutions.(8-13)

In the U.S., no standard model for PPHC@Home exists, but services are primarily provided by home hospice, home health care, or hospital-based pediatric palliative care (PPC) programs that conduct home visits. The composition of and services provided by these PPHC@Home programs are significantly influenced by state and local regulations and resources; therefore, PPHC@Home varies considerably across programs and geographic areas.(8, 9, 14, 15)

In order to improve care for all children with serious illnesses and their families at home, the development of appropriate and feasible measures is critical.(16) More specifically, a family-reported measure of PPHC@Home is needed. A measure of patient and family perceptions of and experiences with the care they receive would provide invaluable information regarding the care provided, including if services meet patient and family priorities and expectations, as well as areas of unmet need and potential improvement for individual patients and families. Ultimately, providers, researchers, policymakers, and

other stakeholders could use this information to improve the quality of care within and across programs.(17, 18)

While several family-reported measures have been developed to evaluate PPC provided in hospital and community settings for children, these instruments were developed for populations outside the U.S. (19-21), where aspects of care differ due to varying health care system structures, funding mechanisms, cultural norms, social policies, and provider practices.(22) The application of these instruments to U.S.-based care may therefore not be appropriate. The one known existing instrument developed to measure PPHC@Home outcomes in a U.S-based program evaluates only one specific domain (namely, health-related quality of life).(23) The development of a comprehensive family-reported experience measure of PPHC@Home provided in the U.S. is necessary to ensure that care teams are meeting children's and families' most important needs and priorities.

METHODS AND RESULTS

The purpose of this project was to develop and conduct a preliminary evaluation of a family-reported measure of experiences with PPHC@Home using a multi-method, multi-stakeholder approach. Starting with a consensus-based conceptual framework (described below), the project was conducted in four phases: Phase 1 - Item identification and development; Phase 2 – Initial prioritization and reduction of items by PPHC professionals; Phase 3 – Final prioritization and reduction of items by parents; and Phase 4 - Cognitive interviewing with parents. Since each phase built on results from the previous phase, the methods and results for each phase are presented together. The Children's Hospital of Philadelphia's (CHOP) Institutional Review Board approved this study.

CONCEPTUAL FRAMEWORK

We used the National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) (24) as a framework for this project. We adapted these general guidelines for the PPHC context using published PPC-specific practice guidelines (16, 25), standards of practice (26), and peer-reviewed literature (27), along with critical feedback from a panel of PPHC stakeholders (providers and parent advocates), resulting in a total of 20 PPHC@Home domains and sub-domains (Table 1).

Phase 1: Item Identification and Development

We identified items and developed the initial pool of items based on a comprehensive review of the literature on existing measures of PPHC@Home quality and outcomes. We conducted the initial biomedical librarian-assisted literature search in Medline/Pubmed, CINAHL, Scopus, and PsycINFO in February 2017 and updated this search in March 2018. Search terms included pediatric palliative, pediatric hospice, quality of care, quality measures, outcome measures, clinical assessment, tools, and instruments.

Out of nearly 200 papers, we identified three comprehensive instruments for evaluating PPC in the home and hospital settings in Canada, Germany, and Switzerland.(19-21) We supplemented the items from these instruments with additional items from four sources: first, the literature describing outcomes from PPHC@Home programs in the U.S. (10-12, 23, 27-30); second, adult hospice quality measures (31, 32); third, PPC-specific quality guidelines and standards of practice (16, 25, 26); and fourth, general palliative care quality guidelines.(24) We compiled over 100 items from these sources. After removing duplicate and irrelevant items, we ended up with a pool of 70 items

(Figure 1). The research team aligned each item with one of the 20 domains of PPHC@Home (Table 1).

We then reviewed the 70 items with a panel of five PPHC stakeholders (physician, nurse practitioner, social worker, and two bereaved parents/parent advocates) from across the U.S. Based on the panel's feedback, we revised items and added five new items for a total of 75 items for evaluation in Phase 2 (Figure 1).

Phase 2: Initial Prioritization and Reduction of Items by PPHC Professionals

To prioritize and reduce the number of candidate items for the measure, we conducted two discrete choice experiments (DCE). First, we conducted a DCE with PPHC professionals (providers, researchers, and parent advocates) to reduce the initial item pool (Phase 2). Second, we conducted a DCE with parent participants to further prioritize and winnow the items (Phase 3, described in the following section).

Overview of DCE Method – DCE is a quantitative, choice-based approach to understanding individuals' stated preferences regarding choices related to healthcare (33-38) and consumer decision-making.(39, 40) We used this approach to obtain quantitative estimates of the relative importance of each item and domain, and to rank order and winnow items. We used a DCE with Bandit MaxDiff Scaling, which is an approach that oversamples top-rated items to increase the precision of estimates of these items.(41, 42) This approach also minimizes sample size requirements and decreases the cognitive and time burden placed on participants by allowing each participant to rate a sub-set of the overall item pool, using Thompson Sampling to select the items for each new respondent based on estimates of each item's mean and variance from previous respondents.(41, 42)

In both DCEs, participants were presented with sets of four items. Within each set, participants were instructed to choose which of the four items was the most important for supporting families caring for a child with serious illness at home, and which item was least important (Figure 2). This process was repeated across different sets, where participants chose the most and least important items among each set that contained a different four-item combination.

Design – Phase 2 participants were professionals who were recruited from hospital, community, and academic settings from the U.S. and Canada using the following inclusion criteria:

- For health care providers: member of an interdisciplinary PPC or hospice team (nurses, physicians, advanced practice nurses, social workers, child life therapists, art therapists, bereavement counselors, chaplains); palliative care or hospice board certification (physicians, nurses, advanced practice nurses) or ≥ 3 years' clinical experience in PPC or hospice (≥ 0.5 FTE); or established PPC researcher.
- For parent advocates: employed by a health system or parent advocacy organization, and has or has had a child who received PPHC@Home services.
- Parent advocates were chosen for involvement in this early phase of the research because of their unique perspective that is based not only on their personal experiences with PPHC@Home, but also by their professional experiences working alongside families, providers, and other stakeholders. Our goal was to recruit 32 interdisciplinary professionals and at least four parent advocates.

Participants were recruited by the principal investigator (PI; co-author JB) via email. Interested participants contacted the PI and were emailed a web link for the discrete

choice survey. They provided electronic informed consent to participate. If surveys were not completed after one week, the PI sent an electronic reminder. Each participant evaluated a subset of 30 items out of the overall 75-item pool from Phase 1 that were displayed in 38 total sets, as well as the 20 PPHC@Home domains that were displayed in 15 sets, for 53 total sets. The entire survey took 30 to 35 minutes to complete.

Analysis – We determined the average ratings and rankings of items using Lighthouse Studio Version 9.6.1 (Sawtooth Software, Inc., Provo, UT), which used an aggregate logit application to multinomial logistic regression in order to estimate the average (mean) probability of choosing each item as most or least important across all participants (represented as a raw logit score). Raw logit scores were transformed to a 0 to 100 probability scale, where the lowest-rated item has a score of zero and scores for all items summed to 100.(42, 43) This transformation facilitates a readily interpretable comparison of items, as this transformed score indicates the relative importance of items on a common scale; for example, an item that is given a score of four is perceived by respondents as being twice as important as an item with a score of two. We then applied the same analytic process to calculate importance scores for each of the 20 PPHC@Home domains (Table 1).

To winnow the number of items, we first rank-ordered the domains by domain importance scores and calculated a maximum number of items “allowed” in each domain based on these importance scores. We then rank-ordered items by item-importance score, and retained the top-ranked items from each domain based on each domain’s item allowance.

Results – Thirty-four PPC professionals participated in this phase, representing all interdisciplinary roles and 3 parent advocates (Table 2). Fifty percent of these

professionals practiced in a hospital-based setting (inpatient and outpatient), 26.5% worked primarily in the home setting, 5.9% worked in both home and hospital settings, and 8.8% were primarily in an academic setting. Over 60% of our sample had eight or more years of PPC or hospice experience.

Supplemental Table 1 presents the top-ranked items, which included *I trust the care team* (Mean score 3.50; SE: 1.27), *Access to on-call services from palliative care or hospice team* (Mean score 3.04; SE: 1.26), and *Care team helps me do the best for my child* (Mean score 3.01; SE: 1.276). Lowest-ranked items included *Care team helps prepare my child for school* (Mean score 0.09; SE: 1.44), *Care team keeps me informed about their arrival time* (Mean score 0.07; SE: 1.50), and *Care team helps with arranging transportation* (Mean score 0.05; SE: 1.54).

In terms of relative importance, professionals rated the item *I trust the care team* (Mean score 3.50; SE: 1.27) as approximately twice as important as *Care team helps me to advocate for my child's needs* (Mean score 1.80; SE: 1.30), which is twice as important as *Care team provides support for my spiritual needs* (Mean score 0.92; SE: 1.35) (Supplemental Table 1).

The top three domains included *Physical Aspects of Care: Symptom Management* (Mean score 11.20; SE: 5.04), *Psychological/Emotional Aspects of Care: Child* (Mean score 10.55; SE: 5.04), and *Psychological/Emotional Aspects of Care: Parent(s)* (Mean score 8.21; SE: 5.01). Lowest-ranked domains included *Social Aspects of Care: Child* (Mean score 1.61; SE: 5.01), *Practical Aspects of Care* (Mean score 1.60; SE: 5.00), and *Emotional Aspects of Care: Extended Social Network* (Mean score 0.36; SE: 5.17) (domain scores not depicted).

Top-ranked items in each domain were retained based on each domain's item allowance (Supplemental Table 1; bolded items). Two new items were added to two domains, *Physical Aspects of Care: Symptom Management* and *Psychological/Emotional Aspects of Care: Child*, which had high importance scores, but not enough items in the item pool to fill these domains. In total, 42 items were retained for further evaluation (Figure 1).

Phase 3: Final Prioritization and Reduction of Items by Parents

Design – Parents were recruited from the CHOP Pediatric Advanced Care Team's (PACT) service area and from the online Courageous Parents Network (CPN), which is a virtual community of parents, families, and providers that provides information, skills, tools, and other resources to support parents and families during their child's illness journey. Participants were English-speaking parents over the age of 18 who had a child with a serious illness and who was younger than 25 years at the time care was received. Parents whose child was currently receiving PPHC@Home, as well as bereaved parents whose child had previously received PPHC@Home, were included in this study.

The PI identified CHOP-based participants with the assistance of PACT's nurse coordinator and social worker and then contacted eligible parents by phone. Interested parents provided electronic informed consent and completed the web-based discrete choice survey concurrently by phone or in-person with the PI, or independently via a web link. The PI recruited participants from CPN with the assistance of CPN's staff, who posted recruitment materials and promoted the study through CPN's email database and social media page. Interested participants reached out to the PI via phone or email and completed the survey via an emailed web link. Parents were contacted a maximum of three times. All parents were compensated for their participation with a \$30 gift card.

In the DCE survey, participants provided their most-least important ratings on a subset of 20 items out of the 42-item pool and on the 20 domains. Items were displayed in different combinations of four items per set over 20 sets, and domains were displayed in different combinations of four domains per set over 15 sets. Participants, therefore, rated 35 sets that took 20 to 30 minutes to complete.

Analysis – We had three goals at this phase, namely to winnow the item pool to include only the highest-priority items, but also to ensure that in the final set of items, each domain was represented in a manner proportional to the rated importance of each domain, and that no single domain was over-represented. We therefore calculated weighted item scores, multiplying each item’s individual score by the importance score for the item’s associated domain, and capped each domain at a maximum of two items to avoid overrepresentation of any domain in the instrument. We then retained the top-rated items by weighted importance score, in keeping with the domain cap.

Results – Forty-seven parents from 45 families participated in this phase. Participants had a mean age of 42.6 years (SD 8.5), and most were white (89.4%), college-educated (68.1% college graduates) mothers (93.6%). Most parents (87.2%) were married or partnered and 48.9% were employed full-time (Table 3).

Approximately 70% of parents were currently caring for their child at home, and over one-third of children received care at home for more than two years. Over 50% of children were between 10 and 25 years of age. While children had a range of diagnoses, 51.1% had a neuromuscular, neurologic, or mitochondrial disease and 48.9% had a genetic or congenital disease (note: disease groups not mutually exclusive) (Table 3).

We calculated weighted item scores and retained the top 20 items. While we capped each domain at 2 items, some domains, as expected, did not have any top-ranked items and were thus removed from the instrument.(44) Top-ranked items by weighted score included *Care team treats my child's physical symptoms so that my child has as good a quality of life as possible*, *I feel prepared to treat my child's symptoms at home*, and *My child can easily get necessary care* (Table 4). In total, 20 items were retained for further evaluation in Phase 4 (Table 4; bolded items). Parent participants' prioritization of the domains are described elsewhere.(44)

Phase 4: Cognitive Interviews with Parents

Design – To examine the clarity of items and the measure's content validity, we conducted cognitive interviews (45) with 11 parents. Parent participants met the same inclusion criteria as Phase 3, and a sub-group of participants were re-contacted from Phase 3.

After agreeing to participate, parents engaged in an in-person, phone-based, or video-based interview with the PI. With consent, interviews were audio-recorded and professionally transcribed. The structured interviews included specific probes about each item and about the overall instrument. Participants provided their interpretation of each item, their feedback on the relevance of items and comprehensiveness of the overall instrument, and their perceptions of issues regarding the clarity of the items and the overall instrument. Participants were compensated for their participation with a \$30 gift card.

Analysis – We followed Knafl et al.'s (2007) protocol for analysis.(45) We first summarized data item-by-item across participants to reflect parents' understanding and

interpretation of each item and to identify potential problems (e.g., limited applicability, unclear reference or perspective, problems with wording/tone) with each item. We then summarized item interpretations and problems in a summary matrix. The PI and co-author JD reviewed all items and decided whether to retain, revise, or omit each item and whether to add new items. These decisions were discussed with the research team.

Results – Eleven parents participated, two of whom also participated in Phase 2.

Participants' sociodemographic and children's clinical characteristics resembled that of Phase 2 participants, although a larger proportion of families in this phase had received PPHC@Home for a longer period of time (Supplemental Table 2).

Interviews lasted, on average, 81 minutes (range: 53 to 106 minutes). We revised several items because parents interpreted the meaning in a way that was not our intent, felt that the item did not apply to families in their situation, or felt the wording to be insensitive or inappropriate. For example, three parents felt that the item *The care team has prepared me for what my child's last weeks of life may be like* was not applicable to their child's rare disease since clinicians were not able to provide an accurate idea of what to expect at the end of life, and, thus, were not able to provide this type of preparation or guidance (code: limited applicability). Additionally, three parents did not like the word "prepare" because, as one parent noted, "I'm not sure that any [parent] would ever say that...I feel ready for that" (code: problems with wording). Based on these parents' suggestions, we revised this item to *The care team has talked with me about my child's last weeks of life and what they may be like*. Overall, we retained the original wording for five items, made minor revisions to 10 items (i.e., changes in one or two words), and made more substantial revisions to five items (i.e., changes in three or more words or otherwise substantial re-organization of the item).

While parents thought that the instrument was comprehensive, five parents suggested that we add an item assessing emotional support for siblings. As a result, we added the item *The care team provides support for my other children's feelings and emotions*. Additionally, while only one parent suggested an item about support for the parent caregiver, we agreed that this was a critical gap in the instrument and developed a new item, *The care team has provided or directed me to resources that support my needs as my child's caregiver*. Ultimately, 22 items were retained as the final version of the **Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home)** Measure (Table 5).

DISCUSSION

Employing a multi-method, multi-stakeholder approach for instrument development, we have developed the 22-item EXPERIENCE@Home Measure, which measures families' experiences with PPHC@Home in the U.S. We began with broad palliative care guidelines and the peer-reviewed PPC and PPHC@Home literature and incorporated the perspectives of different stakeholders. To our knowledge, this is the first published study to have used a DCE approach for health care instrument development.

A strength of the EXPERIENCE@Home instrument is the multiple perspectives used to develop the items, including existing guidelines, peer-reviewed literature, and instruments; interdisciplinary PPHC professionals, researchers, and parent advocates; and bereaved parents and parents who are currently caring for their child at home. For example, we developed the item, *The care team treats my child's physical symptoms so that my child has as good a quality of life as possible*, based on NCP Guidelines' recommendation for assessing "physical symptoms and their impact on well-being, quality of life, and functional status."⁽²⁴⁾ Our item not only evaluates whether or not a

child's physical symptoms were treated, as in existing instruments (20, 21, 23, 46, 47), but additionally, if treatment was perceived as effective – that is, if symptoms were treated in a way that the child could enjoy as good a quality of life as possible. This emphasis on quality of life was a priority for parents in our study, who rated this item as the most important (nearly twice as important as the next most important item) (Table 4). In cognitive interviews, parents emphasized the importance of treating symptoms in a way that their child could remain a part of family life and participate in social activities at school and in the community. Parents in other studies have also reported that PPHC@Home services were crucial for managing their children's symptoms and supporting their children's health-related quality of life in the home setting.(10, 12, 23, 30)

Another strength is that our instrument also assesses home-specific aspects of care that were not represented in existing instruments. For example, the item, *The care team helps adapt our home to better support my child's current care needs*, is based on recommendations from the Institute of Medicine (16) and the National Hospice and Palliative Care Organization.(26) Interestingly, several parents in our study spoke not only of the importance of medical equipment and other home adaptations, such as a hospital bed or adaptive chairs, but also the importance of equipment that does not look too “medicalized” and that does not drastically alter the home environment. This is similar to a finding in a previous qualitative study of 12 families with children requiring mechanical ventilation in the home, where parents spoke of the importance of their homes looking “normal,” and that medical devices, equipment, and other adaptations (such as elevators or ceiling rails for facilitating mobilization) were camouflaged, hidden, or discrete so that they did not “dominate” the home environment.(48) More work is needed to better understand families' needs in this area.

Another item unique to the home setting is *The care team helps me to feel confident in managing my child's symptoms at home without needing to go to the hospital*. We adapted this item from two existing studies on community-based PPC.(10, 23) Parents in our study spoke of the importance of having knowledge, supplies, medications, equipment, phone-based or in- person support from providers, and a plan in place when crisis situations arose at home. Parents' confidence and perceived ability to manage their child's illness and care needs have been observed to be an important facilitator of PPHC@Home.(9) Comprehensive programs that provide families access to 24/7 on-call services, care coordination, and home visits by nursing and medical providers support the family's ability to manage symptoms, particularly at the end of life.(10, 29, 49) These programs also help reduce unwanted hospital utilization and facilitate death in the family's preferred location.(10, 12, 20, 23, 50)

Finally, the item *The nurses have the knowledge, skills, and experience to support my child's palliative or hospice care at home* was adapted from one existing instrument (20) and informed by NCP Guidelines.(24) The availability of appropriately-trained nurses was a significant issue for many parents in our study. Several parents spoke of challenges in finding nurses who had both end-of-life and pediatric expertise. While home-based palliative and hospice nursing support is especially critical for helping caregivers troubleshoot technical problems and make decisions, and for providing respite care and overall emotional support to the family (49, 51), finding adequate palliative and hospice nursing support is often challenging.(51-53) Additionally, home care nurses play a significant role in the home-based care of many children with serious illnesses. Yet, finding adequate skilled and/or private duty nurses is often a significant challenge for families (54, 55), which could lead to outcomes such as unintended re-hospitalizations (56, 57), increased hospital use (56, 58), and poor parental health and

wellbeing.(55, 59) One parent in our study expressed frustration at being eligible for a certain number of skilled nursing hours for her child, but not being able to find nurses to fill those hours: "It's just a sad scenario when you have the hours in place, and then you can't find qualified people to cover them. That's why you end up back in the hospital, right...sometimes you do settle because you need the extra hands because you're just exhausted. Other times, you have to consider the situation and say, oh, we need to be back in the hospital because we're just not going to be able to do it here."

The cognitive interviews, of note, informed the addition of the item *The care team provides support for my other children's feelings and emotions*. Several parents spoke of the importance of the team's support for their other children, particularly as siblings may often be very involved in the day-to-day care of the ill sibling in the home. This proximity of siblings to the ill child's care may be particularly important to PPHC provided in the home setting: one qualitative study found that siblings may fill many different roles at home, including playmate, companion, and helper, which included providing direct care such as feeding, toileting, and carrying their sibling from room to room.(60) Future work will need to evaluate if our new item adequately addresses siblings' most pressing needs in the home.

This instrument development project has some limitations. First, while we followed a rigorous process of item selection, prioritization, reduction, and cognitive interviewing, we may have missed or excluded important items or domains in our instrument. We will continue to evaluate the content validity of the instrument in future work. Second, our PPC professional sample was relatively socio-demographically homogenous, although it does reflect the overall demographic profile of hospice and palliative physicians (61) and the nursing workforce more generally (62) in the U.S. Furthermore, professional

participants represented several different professions, including parent advocates, nurses, physicians, social workers, chaplains, and expressive therapists. The sociodemographic profile of parent participants was also relatively homogenous, although not surprising given issues with gender (63, 64) and racial and ethnic minority (65) imbalance in PPC studies, as well as racial and ethnic disparities in access to home-based hospice care.(66) We did, however, include parents of children with diverse diseases, and we also recruited a diverse sample with regard to illness trajectories by including parents whose children were currently receiving care, as well as bereaved parents. Additionally, both the professional and the parent samples came from a wide range of geographic regions, institutions, and care models. Nonetheless, it will be important to recruit a greater representation of fathers, racial and ethnic minorities, and persons from varying socioeconomic backgrounds in future work. Third, because we recruited a portion of our parent participants from an online network, we were unable to assess our nonresponse rate and potential differences in how responders compared to non-responders. In our CHOP-based parents, however, reasons for non-participation typically related to being too busy or their child being too sick at the time of the study.

The next steps in the assessment and refinement of the EXPERIENCE@Home Measure will include psychometric testing with a larger sample of parents of children currently receiving care at home, as well as an evaluation of the clinical utility of the instrument with PPHC providers for providing real-time, family-reported feedback to palliative care and hospice teams. While this further evaluation work is ongoing, we have developed a new instrument using rigorous methods that promises to be clinically useful for children with serious illness and their families. Our parent participants reiterated the importance of having a way to provide feedback about their care experiences at home to their providers. As one parent told us, "I think it would be a really effective tool... it just gives

families like a, I don't know, some agency over like what's happening, and I think it's a good thing."

LIST OF ABBREVIATIONS

PPC: pediatric palliative care; PPHC: pediatric palliative and hospice care;
PPHC@Home: pediatric palliative and hospice care at home; NCP: National Consensus Project; DCE: discrete choice experiment; CHOP: Children's Hospital of Philadelphia; PACT: Pediatric Advanced Care Team; CPN: Courageous Parents Network; SE: standard error; EXPERIENCE@Home: **Ex**periences of **P**alliative and **H**ospice Care for **C**hildren and **C**aregivers at Home Measure.

DECLARATIONS

- **Ethics approval and consent to participate.** The Children's Hospital of Philadelphia's (CHOP) Institutional Review Board approved this study. All participants provided their electronic informed consent to participate.
- **Consent for publication.** Not applicable.
- **Availability of data and materials.** The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.
- **Competing interests.** The authors declare that they have no competing interests.
- **Funding.** Research reported in this publication was supported by the National Institute of Nursing Research of the National Institutes of Health under Award Number F31NR017554. The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Institutes of Health. Additional research funding was provided by the University of

Pennsylvania School of Nursing Office of Nursing Research Student Research Grant, the Independence Blue Cross Foundation Nurses for Tomorrow Scholars Program, and the Sawtooth Software Academic Grant. Funders were not involved in the design of the study; in data collection, analysis, or interpretation; or in writing the manuscript.

- **Authors' contributions.** JB was the principal investigator and was primarily responsible for the design, conduction (including data collection, analysis, and interpretation), and write-up of this manuscript. Only JB had full access to the data collected. ME was the dissertation chair and oversaw all aspects of the study design, conduction, and manuscript write-up. ME, CF, JD, and KW were dissertation committee members and were involved in the study conceptualization and design, had access to de-identified data, and were directly involved with data analysis and interpretation. GL and BL were involved in the study design and interpretation of results. All authors revised drafts critically for important intellectual content, and read and approved the final manuscript. Each author agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.
- **Acknowledgements.** The authors would like to acknowledge the Children's Hospital of Philadelphia's Pediatric Advanced Care Team and the Courageous Parents Network, who supported the design and data collection phases of this study. In particular, we would like to thank Ms. Karen Crew and Ms. Dana Dombrowski for their guidance and effort on this project. We would also like to thank Ms. Solim Lee, Ms. Heather Carlino, and Dr. Patricia Pawlow from the

University of Pennsylvania School of Nursing for their assistance during the development of the instrument. We would like to acknowledge all of the clinicians, parent advocates, and researchers who shared their time and expertise with us. And finally, we would like to acknowledge the parents who so generously gave their time and energy to participate in this project. We are deeply grateful for their willingness to share their perspectives with us and to help us work toward advancing the quality of care provided in the home setting for future children and families.

BIBLIOGRAPHY

1. Feudtner C, Feinstein JA, Satchell M, Zhao H, Kang TI. Shifting place of death among children with complex chronic conditions in the United States, 1989-2003. *JAMA*. 2007;297(24):2725-32.
2. Feudtner C, Kang TI, Hexem KR, Friedrichsdorf SJ, Osenga K, Siden H, et al. Pediatric palliative care patients: A prospective multicenter cohort study. *Pediatr*. 2011;127(6):1094-101.
3. Burke RT, Alverson B. Impact of children with medically complex conditions. *Pediatr*. 2010;126(4):789-90.
4. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980-1998. *Pediatr*. 2002;109(4):656-60.
5. Friebert S, Williams C. NHPCO'S Facts and Figures: Pediatric Palliative and Hospice Care in America. Alexandria, VA: NHPCO; 2015.
6. Feudtner C, Hays RM, Haynes G, Geyer JR, Neff JM, Koepsell TD. Deaths attributed to pediatric complex chronic conditions: National trends and implications for supportive care services. *Pediatr*. 2001;107(6):E99.
7. Nageswaran S, Radulovic A, Anania A. Transitions to and from the acute inpatient care setting for children with life-threatening illness. *Pediatr Clin North Am*. 2014;61(4):761-83.
8. Kaye E, Rubenstein J, Levine D, Baker JN, Dabbs D, Friebert S. Pediatric palliative care in the community. *CA Cancer J Clin*. 2015;65:315-33.
9. Boyden JY, Curley MAQ, Deatrick JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage*. 2018;55(1):117-31.
10. Bona K, Bates J, Wolfe J. Massachusetts' Pediatric Palliative Care Network: Successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med*. 2011;14(11):1217-23.
11. Knapp CA, Madden VL, Curtis CM, Sloyer PJ, Huang IC, Thompson LA, et al. Partners in Care: Together for Kids: Florida's model of pediatric palliative care. *J Palliat Med*. 2008;11(9):1212-20.
12. 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.
13. Thienprayoon R, Grossoehme D, Humphrey L, Pestian T, Frimpong-Manso M, Malcolm H, et al. "There's Just No Way to Help, and They Did." Parents name Compassionate Care as a new domain of quality in pediatric home-based hospice and palliative care. *J Palliat Med*. 2020;23(6):767-76.
14. Lindley LC, Lyon ME. A profile of children with complex chronic conditions at end of life among Medicaid beneficiaries: Implications for health care reform. *J Palliat Med*. 2013;16(11):1388-93.
15. Carroll JM, Santucci G, Kang TI, Feudtner C. Partners in Pediatric Palliative Care: A program to enhance collaboration between hospital and community palliative care services. *Amer J Hosp Palliat Med*. 2007;24(3):191-5.
16. Field MJ, Behrman RE. *When Children Die: Improving Palliative and End-of-Life care for Children and their Families*. Washington, DC: The National Academies Press; 2003.

17. Conway PH, Mostashari F, Clancy C. The future of quality measurement for improvement and accountability. *JAMA*. 2013;309(21):2215-6.
18. Snyder CF, Aaronson NK, Choucair AK, Elliott TE, Greenhalgh J, Halyard MY, et al. Implementing patient-reported outcomes assessment in clinical practice: A review of the options and considerations. *Qual Life Res*. 2012;21(8):1305-14.
19. Widger K, Brennenstuhl S, Duc J, Tourangeau A, Rapoport A. Factor structure of the Quality of Children's Palliative Care Instrument (QCPCI) when completed by parents of children with cancer. *BMC Palliat Care*. 2019;18(1):23.
20. Vollenbroich R, Duroux A, Grasser M, Brandstatter M, Borasio GD, Fuhrer M. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med*. 2012;15(3):294-300.
21. Zimmermann K, Cignacco E, Eskola K, Engberg S, Ramelet A-S, Von der Weid N, et al. Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu): An instrument to assess parental experiences and needs during their child's end-of-life care. *J Adv Nurs*. 2015;71(12):3006-17.
22. Linton JM, Feudtner C. What accounts for differences or disparities in pediatric palliative and end-of-life care? A systematic review focusing on possible multilevel mechanisms. *Pediatr*. 2008;122:574-82.
23. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based pediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. *BMC Palliat Care*. 2016;15:73.
24. National Consensus Project for Quality Palliative Care. *Clinical Practice Guidelines for Quality Palliative Care*. Richmond, VA: National Coalition for Hospice and Palliative Care; 2018. Available from: <https://www.nationalcoalitionhpc.org/ncp/>. Accessed June 5, 2020.
25. Feudtner C, Friebert S, Jewell J. Pediatric palliative care and hospice care commitments, guidelines, and recommendations. *Pediatr*. 2013;132(5):966-72.
26. National Hospice and Palliative Care Organization (NHPCO). *Standards of practice for pediatric palliative care*. Alexandria, VA: NHPCO; 2019 2009. Available from: https://www.nhpc.org/wp-content/uploads/2019/07/Pediatric_Standards.pdf. Accessed June 12, 2020.
27. Thienprayoon R, Mark MSJ, Grosseohme D. Provider-prioritized domains of quality in pediatric home-based hospice and palliative care: A study of the Ohio Pediatric Palliative Care and End-of-Life Network. *J Palliat Med*. 2018;31(3):290-6.
28. Gans D, Kominski GF, Roby DH, Diamant AL, Chen X, Lin W, et al. Better outcomes, lower costs: Palliative care program reduces stress, costs of care for children with life-threatening conditions. *UCLA Center for Health Policy Research*. 2012.
29. Thienprayoon R, Marks E, Funes M, Martinez-Puente LM, Winick N, Lee SC. Perceptions of the pediatric hospice experience among English- and Spanish-speaking families. *J Palliat Med* 2016;19(1):30-41.
30. Hays RM, Valentine J, Haynes G, Geyer JR, Villareale N, McKinstry B, et al. The Seattle Pediatric Palliative Care Project: Effects on family satisfaction and health-related quality of life. *J Palliat Med* 2006;9(3):716-28.
31. Anhang Price R, Stucky B, Parast L, Elliott MN, Haas A, Bradley M, et al. Development of valid and reliable measures of patient and family experiences of hospice care for public reporting. *J Palliat Med*. 2018;21(7):924-32.
32. Kutney-Lee A, Carpenter J, Smith D, Thorpe J, Tudose A, Ersek M. Case-mix adjustment of the Bereaved Family Survey. *Amer J Hosp Palliat Med*. 2018;35(7):1015-22.

33. Gomes B, de Brito M, Sarmiento VP, Yi D, Soares D, Fernandes J, et al. Valuing attributes of home palliative care with service users: A pilot discrete choice experiment. *J Pain Symptom Manage.* 2017;54(6):973-85.
34. Huynh E, Coast J, Rose J, Kinghorn P, Flynn T. Values for the ICECAP-Supportive Care Measure (ICECAP-SCM) for use in economic evaluation at end of life. *Soc Sci Med.* 2017;189:114-28.
35. Mangham LJ, Hanson K, McPake B. How to do (or not to do) ... Designing a discrete choice experiment for application in a low-income country. *Health Policy Plan.* 2009;24(2):151-8.
36. Ryan M, Gerard K, Amaya-Amaya M. Using Discrete Choice Experiments to Value Health and Health Care. Bateman IJ, editor. The Netherlands: Springer; 2008.
37. Feudtner C, Walter JK, Faerber JA, Hill DL, Carroll KW, Mollen CJ, et al. Good-parent beliefs of parents of seriously ill children. *JAMA Pediatr.* 2015;169(1):39-47.
38. October TW, Fisher K, Feudtner C, Hinds PS. The parent perspective: "being a good parent" when making critical decisions in the PICU. *Pediatr Crit Care Med.* 2014;15(4):291-8.
39. Cohen SH. Maximum difference scaling: Improved measures of importance and preference for segmentation. Sequim, WA: Sawtooth Software; 2003.
40. Louviere JJ, Flynn TN, Carson RT. Discrete choice experiments are not conjoint analysis. *J Choice Model.* 2010;3(3):57-72.
41. Sawtooth Software. Bandit MaxDiff: When to use it and why it can be a better choice than standard MaxDiff. Orem, UT; 2018. Available from: <https://www.sawtoothsoftware.com/support/technical-papers/maxdiff-best-worst-scaling/175-support/technical-papers/maxdiff-best-worst-scaling/1943-bandit-maxdiff-when-to-use-it-and-why-it-can-be-a-better-choice-than-standard-maxdiff>. Accessed June 30, 2020.
42. Chrazan K, Orme B. Applied MaxDiff: A practitioner's guide to Best-Worst Scaling Provo, UT: Sawtooth Software, Inc.; 2019.
43. Sawtooth Software. Aggregate score estimation via logit analysis Orem, UT: Sawtooth Software, Inc.; 2018. Available from https://www.sawtoothsoftware.com/help/lighthouse-studio/manual/maxdiff_aggregate_logit_analysis.html. Accessed June 30, 2020.
44. Boyden JY, Ersek M, Deatrick JA, Widger K, LaRagione G, Lord B, Feudtner C. What do parents value regarding home-based pediatric palliative and hospice care? *J Pain Symptom Manage.* *Accepted for publication (July 2020).*
45. Knafelz K, Deatrick J, Gallo A, Holcombe G, Bakitas M, Dixon J, et al. The analysis and interpretation of cognitive interviews for instrument development. *Research in Nursing & Health.* 2007;30(2):224-34.
46. Donnelly JP, Downing K, Cloen J, Fragen P, Gupton AW, Misasi J, et al. Development and assessment of a measure of parent and child needs in pediatric palliative care. *J Pain Symptom Manage.* 2017;55(4):1077-84.
47. Widger K, Tourangeau AE, Steele R, Streiner DL. Initial development and psychometric testing of an instrument to measure the quality of children's end-of-life care. *BMC Palliat Care.* 2015;14(1):1.
48. Carnevale FA, Alexander E, Davis M, Rennick J, Troini R. Daily living with distress and enrichment: The moral experience of families with ventilator-assisted children at home. *Pediatrics.* 2006;117(1):e48.
49. Davies B, Gudmundsdottir M, Worden B, Orloff S, Sumner L, Brenner P. "Living in the dragon's shadow" fathers' experiences of a child's life-limiting illness. *Death Stud.* 2004;28(2):111-35.

50. Postier A, Chrastek J, Nugent S, Osenga K, Friedrichsdorf SJ. Exposure to home-based pediatric 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.
51. Bee PE, Barnes P, Luker KA. A systematic review of informal caregivers' needs in providing home-based end-of-life care to people with cancer. *J Clin Nurs.* 2009;18(10):1379-93.
52. Rehm RS, Bradley JF. Normalization in families raising a child who is medically fragile/technology dependent and developmentally delayed. *Qual Health Res.* 2005;15(6):807-20.
53. Varela AMS, Deal AM, Hanson LC, Blatt J, Gold S, Dellon EP. Barriers to hospice for children as perceived by hospice organizations in North Carolina. *Amer J Hospice Palliat Care.* 2012;29(3):171-6.
54. Weaver MS, Wichman B, Bace S, Schroeder D, Vail C, Wichman C, et al. Measuring the impact of the home health nursing shortage on family caregivers of children receiving palliative care. *J Hosp Palliat Nurs.* 2018;20(3):260-5.
55. Meltzer LJ, Boroughs DS, Downes JJ. The relationship between home nursing coverage, sleep, and daytime functioning in parents of ventilator-assisted children. *J Pediatr Nurs.* 2010;25(4):250-7.
56. Gay JC, Thurm CW, Hall M, Fassino MJ, Fowler L, Palusci JV, et al. Home health nursing care and hospital use for medically complex children. *Pediatr.* 2016;138(5):e20160530.
57. Sobotka SA, Lynch E, Peek ME, Graham RJ. Readmission drivers for children with medical complexity: Home nursing shortages cause health crises. *Pediatr Pulmonol.* 2020;55(6):1474-80.
58. Boss RD, Williams EP, Henderson CM, Seltzer RR, Shapiro MC, Hahn E, et al. Pediatric chronic critical illness: Reducing excess hospitalizations. *Hosp Pediatr.* 2017;7(8):460-70.
59. Caicedo C. Health and functioning of families of children with special health care needs cared for in home care, long-term care, and medical day care settings. *J Dev Behav Pediatr.* 2015;36:352-61.
60. Eaton Russell C, Widger K, Beaune L, Neville A, Cadell S, Steele R, et al. Siblings' voices: A prospective investigation of experiences with a dying child. *Death Stud.* 2018;42(3):184-94.
61. Salsberg E, Mehfoud N, Quigley L, Lupu D. A profile of active hospice and palliative medicine physicians, 2016. American Academy of Hospice and Palliative Medicine & Health Workforce Institute, The George Washington University 2017 March. Available from: http://aahpm.org/uploads/Profile_of_Active_HPM_Physicians_September_2017.pdf. Accessed June 30, 2020.
62. Nursing workforce projections by ethnicity and race 2014-2030. HRSA Health Workforce; 2017 December. Available from <https://bhw.hrsa.gov/sites/default/files/bhw/health-workforce-analysis/research/projections/hrsa-bhw-rn-lpn-factsheet-12-17.pdf>. Accessed June 30, 2020.
63. Hinds PS, Burghen EA, Pritchard M. Conducting end-of-life studies in pediatric oncology. *West J Nurs Res.* 2007;29(4):448-65.
64. Macdonald ME, Chilibeck G, Affleck W, Cadell S. Gender imbalance in pediatric palliative care research samples. *Palliat Med.* 2010;24(4):435-44.
65. Knapp CA, Madden VL, Curtis C, Sloyer PJ, Shenkman EA. Assessing non-response bias in pediatric palliative care research. *Palliat Med.* 2010;24(3):340-7.
66. Cohen LL. Racial/ethnic disparities in hospice care: A systematic review. *J Palliat Med.* 2008;11(5):763-8.

67. CAHPS Hospice Survey: Frequently asked questions. Centers for Medicare & Medicaid Services, Baltimore, MD 2019 Feb 28. Available from: <https://hospicecahpsurvey.org/en/hospice-specific-faqs2/#Q8>. Accessed June 30, 2020.
68. Charlebois J, Cyr C. Quality indicators for paediatric palliative care. *Paediatric Child Health*. 2015;20(3):145-7.
69. Widger KA, Wilkins K. What are the key components of quality perinatal and pediatric end-of-life care? A literature review. *J Palliat Care*. 2004;20(2):105-12.
70. Hinds PS, Menard JC, Jacobs SS. The child's voice in pediatric palliative and end-of-life care. *Prog Palliat Care*. 2012;20(6):337-42.

TABLES AND FIGURES

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home

Table 2: Characteristics of Professional Participants (Phase 2)

Table 3: Demographic and Clinical Characteristics of Parent Participants and Their
Children (Phase 3)

Table 4: Items Prioritized by Parents (Phase 3)

Table 5: Final EXPERIENCE@Home Measure

Figure 1: Item Selection and Reduction Process

Figure 2: Sample DCE Choice Set

Supplemental Table 1: Items Prioritized by PPHC Professionals (Phase 2)

Supplemental Table 2: Demographic and Clinical Characteristics of Parent Participants
and Their Children (Phase 4)

Table 1: Domains of High-Quality Pediatric Palliative and Hospice Care at Home

Domains	Domain Definitions
1. Access to care	Care team provides access to palliative and hospice care to the child and family 24 hours a day, 7 days a week
2. Caregiver support at the end of life	Care team meets the emotional, spiritual, social, and cultural needs of families at the end of life (for example, preparing parents for the end of life)
3. Communication at the end of life	Care team works with the child and family to develop and implement a care plan to address actual or potential symptoms at the end of life
4. Communication between family and care team	Care team communicates with the child and family to make sure that the care provided meets the child's and family's values, preferences, goals, and needs
5. Coordination of care	Care team works to make sure that when there are transfers between healthcare settings and providers, that there is timely and thorough communication of the child's/family's goals, preferences, values, and clinical information to ensure continuity of care and seamless follow-up (for example, getting needed services, arranging for medical equipment)
6. Continuity of care	Care team works to make sure that the delivery of palliative and hospice care is seamless across care settings and providers (for example, the same providers work with family)
7. Cultural aspects of care	Care team respects the child's and family's cultural beliefs and language preferences
8. Ethical and legal aspects of care	Child's and family's goals, preferences, and choices are respected within the limits of state and federal law, current standards of medical care, and professional standards of practice. These goals/preferences/choices are documented and shared with all professionals involved in the child's care
9. Knowledge and skills of care team providers	Care team members have the necessary education and training to provide high quality palliative care for children and families
10. Physical aspects of care: Communication	Care team provides information about treatments for child's pain and other physical symptoms (for example, nausea, fatigue, constipation)
11. Physical aspects of care: Symptom management	Care team assesses and manages pain and other physical symptoms and side effects based on the best available medical evidence

12. Practical aspects of care	Care team provides the family with assistance and resources for dealing with financial and insurance related issues
13. Psychological and emotional aspects of care: Child	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the child based on the best available medical evidence
14. Psychological and emotional aspects of care: Parent(s)	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the parent(s)
15. Psychological and emotional aspects of care: Sibling(s)	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the child's sibling(s)
16. Psychological and emotional aspects of care: Extended social network	Care team assesses and manages psychological and emotional issues (such as anxiety, distress, coping, grief) of the family's familial and social community
17. Relationship between family and care team	Relationship between care team and family is built on trust, respect, and advocacy for the child's and family's needs
18. Social aspects of care: Child	Care team helps with social issues to meet child-family needs, promote child-family goals, and maximize child-family strengths and well-being (examples include helping family maintain and strengthen their support network; help family develop strategies to balance caregiving, work, and family needs)
19. Social aspects of care: Parent(s)	Care team helps with social issues to meet child-family needs, promote child-family goals, and maximize child-family strengths and well-being (examples include helping family maintain and strengthen their support network; help family develop strategies to balance caregiving, work, and family needs)
20. Spiritual and religious aspects of care	Care team assists with religious and spiritual rituals or practices as desired by the child and family

Note: The above domains are based on the National Consensus Project's (NCP) Clinical Guidelines for Quality Palliative Care (4th edition) (24), which we further adapted using pediatric palliative care specific guidelines and the literature (16, 19, 25-27) and using critical feedback from a panel of PPHC stakeholders (providers and parent advocates).

Table 2: Characteristics of Professional Participants (Phase 2)

Participant Characteristics (n = 34)		No. (%)
Age (years)	<i>Mean (SD)</i>	48.4 (9.7)
Gender	Female	31 (91.2%)
	Male	3 (8.8%)
Race	Asian	2 (5.9%)
	Black or African American	1 (2.9%)
	White	31 (91.2%)
Ethnicity	Non-Hispanic	34 (100%)
Professional Role	Chaplain	6 (17.6%)
	Expressive Therapist	1 (2.9%)
	Nurse	2 (5.9%)
	Nurse practitioner	5 (14.7%)
	Parent Advocate	3 (8.8%)
	Physician	9 (26.5%)
	Researcher	3 (8.8%)
	Social Worker	5 (14.8%)
Primary practice setting	Academic	3 (8.8%)
	Hospital (inpatient, outpatient)	17 (50.0%)
	Home	9 (26.5%)
	Home and Hospital	2 (5.9%)
	Not applicable	2 (5.9%)
	Other	1 (2.9%)
Years of PPC experience	3 to 7 years	13 (38.2%)
	8 to 10 years	7 (20.6%)
	More than 10 years	14 (41.2%)

Table 3: Demographic and Clinical Characteristics of Parent Participants and Their Children (Phase 3)

Parents' Characteristics (n = 47)		No. (%)
Parent type	Mother	44 (93.6%)
	Father	3 (6.4%)
Age	<i>Mean / SD</i>	42.6 (8.5)
Race	White	42 (89.4%)
	Black or African American	1 (2.1%)
	More than one race/Other	3 (6.4%)
	Prefer not to answer	1 (2.1%)
Ethnicity	Non-Hispanic	43 (91.5%)
	Hispanic	3 (6.4%)
	Prefer not to answer	1 (2.1%)
Highest Education Level Completed	Grade school	1 (2.1%)
	High school / general educational development	2 (4.3%)
		4 (8.5%)
	Trade / technical / vocational	8 (17.0%)
	Associates / Professional	19 (40.4%)
	College	13 (27.7%)
	Graduate school	
Relationship Status	Married / partnered	41 (87.2%)
	Separated / divorced / Widowed	6 (12.8%)
Number of Other Children	0	11 (23.4%)
	1-3	35 (74.5%)
	4 or more	1 (2.1%)
Employment Status	Full time	23 (48.9%)
	Part time	5 (10.6%)
	Not employed outside of the home	17 (36.2%)
	Prefer not to answer	2 (4.3%)

Bereavement Status	Bereaved	14 (29.8%)
	Currently caring for child at home	33 (70.2%)
Affiliation	CHOP	16 (34.0%)
	CPN	31 (66.0%)
Children's Characteristics (n = 45)		No. (%)
Age	1 year or less	8 (17.8%)
	2-4 years	9 (20.0%)
	5-9 years	5 (11.1%)
	10-18 years	17 (37.8%)
	19-25 years	6 (13.3%)
Gender	Female	21 (46.7%)
	Male	24 (53.3%)
Race	White	37 (82.2%)
	Black or African American	2 (4.4%)
	More than one race/Other	5 (11.1%)
	Prefer not to answer	1 (2.2%)
Ethnicity	Non-Hispanic	39 (86.7%)
	Hispanic	4 (8.9%)
	Prefer not to answer	2 (4.4%)
Primary complex chronic condition (Note: not mutually exclusive; thus, the % does not sum to 100%)	Cardiovascular	10 (22.2%)
	Gastrointestinal	4 (8.9%)
	Genetic or congenital	22 (48.9%)
	Hematologic or immunologic	4 (8.9%)
	Malignancy	5 (11.1%)
	Metabolic	10 (22.2%)
	Neuromuscular, neurologic, or mitochondrial	23 (51.1%)
	Respiratory	6 (13.3%)
	Other/Unknown	1 (2.2%)

Primary care team (hospice v. palliative care)	Hospice	19 (42.2%)
	Palliative Care	24 (53.3%)
	Unknown/Not sure	2 (4.4%)
Length of time receiving home-based palliative or hospice care	Less than 1 month	5 (11.1%)
	1 to 3 months	5 (11.1%)
	4 to 6 months	7 (15.6%)
	7 to 12 months	5 (11.1%)
	1 to 2 years	8 (17.8%)
	More than 2 years	15 (33.3%)

Table 4: Items Prioritized by Parents (Phase 3)

****Bolded** items were retained for evaluation in next phase*

Item	Domain	Weighted Item Score	Original Instrument
1. Care team treats my child's physical symptoms so that my child has as good a quality of life as possible.	Physical aspects of care: Symptom management	61.08	New item after Phase 2 analysis (NCP 4th Edition Domains) (24)
2. I feel prepared to treat my child's symptoms at home	Physical aspects of care: Symptom management	35.47	Massachusetts PPCN Evaluation (Bona, 2011) (10)
3. My child can easily get necessary care	Access to care team	30.30	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
4. Care team uses medicines to ease my child's pain and other symptoms.	Physical aspects of care: Symptom management	27.50	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
5. I trust the care team	Relationship between family and care team	26.94	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
6. Care team works together with me and my child to make medical decisions	Relationship between family and care team	26.19	Parental PELICAN Questionnaire (Zimmerman, 2015) (21); Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
7. Care team helps me do the best for my child	Relationship between family and care team	24.43	New item (PPHC expert panel, Sept 2018)
8. Care teams asks for my opinions and concerns about my child	Communication between family and care team	24.20	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)

9. Care teams are all working towards the same goals for my child's care	Continuity of care	24.10	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
10. Care team gives me enough information to make good health care decisions	Communication between family and care team	23.84	Parental PELICAN Questionnaire (Zimmerman, 2015) (21); Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
11. Care team helps me to advocate for my child's needs	Relationship between family and care team	23.20	New item (PPHC expert panel, Sept 2018)
12. Access to on-call service from palliative care or hospice team	Access to care team	23.20	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
13. I have access to care provider who can coach or guide me to care for my child	Care coordination	22.67	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
14. Care team looks at all of my child's needs	Continuity of care	20.38	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
15. It is easy to contact the care team	Access to care team	20.23	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
16. Knowledge/skills of nurse(s)	Knowledge and skills of care team providers	18.72	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
17. Information shared between <u>me</u> and the <u>care team</u> is clear	Communication between family and care team	18.63	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)

18. Knowledge/skills of physician(s)	Knowledge and skills of care team providers	18.34	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
19. Care team provides information about treatments for my child's pain and other symptoms	Physical aspects of care: Communication	17.19	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
20. Care team takes time to listen carefully	Communication between family and care team	16.83	Consumer Assessment of Healthcare Providers and Systems (CAHPS) Hospice Survey (67)
21. Care team helps me to use non-drug measures to ease my child's pain and other symptoms	Physical aspects of care: Symptom management	15.62	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
22. Care team is kind, caring, and respectful	Relationship between family and care team	13.71	Bereaved Family Survey (32)
23. Care team helps me adapt my home to support my child's care needs	Care coordination	13.38	IOM 2003 report (16); NHPCO 2019 standards (26)
24. Care team provides emotional support for my child	Psychological/emotional aspects of care: <u>Child</u>	12.38	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
25. Care team provides opportunities for my child to talk about his/her worries and fears	Psychological/emotional aspects of care: <u>Child</u>	12.31	New item (developed based on recommendation from dissertation committee; Jan 2019)
26. Care team helps me hope for best outcome while also helping me prepare in case that outcome does not happen	Psychological/emotional aspects of care: <u>Parent(s)</u>	11.11	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
27. I can talk about my child's end of life with care team	Communication at end of life	11.11	Parental Questionnaire 1 (Vollenbroich, 2012) (20)

28. Care team has prepared me for what my child's last weeks of life may be like	Caregiver support at the end of life	9.01	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
29. Care team talks with me about my fears and worries	Psychological/emotional aspects of care: <u>Parent(s)</u>	8.88	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
30. My child receives complementary and alternative medicine	Physical aspects of care: Symptom management	7.53	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
31. Care team helps me talk about my child's preferred place of death	Communication at end of life	6.42	Quality indicators for paediatric palliative care (Charlebois) (68)
32. Care team provides emotional support for my other children	Psychological/emotional aspects of care: <u>Sibling(s)</u>	6.20	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
33. Care team helps me cope with the stress of caregiving	Social aspects of care: <u>Parent(s)</u>	5.19	NCP 4th Edition Domains (24)
34. Care team provides emotional support for me	Psychological/emotional aspects of care: <u>Parent(s)</u>	4.53	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
35. Care team helps me find resources to cope with financial strain	Practical aspects of care	4.41	NCP 4th Edition Domains (24); Massachusetts PPCN Evaluation (Bona, 2011) (10)
36. Care team helps me talk about whether to stop life-sustaining measures	Ethical and legal aspects of care	3.76	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
37. Knowledge/skills of social worker(s)	Knowledge and skills of care team providers	3.18	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
38. Care team helps me talk with my child about death and dying	Communication at end of life	2.58	Parental Questionnaire 1 (Vollenbroich, 2012) (20)

39. Care team helps prepare my child for school	Social aspects of care: Child	0.47	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
40. Care team provides emotional support for my child's extended social network (e.g., classmates, neighbors, extended family)	Emotional aspects of care: Extended social network	0.22	New item (PPHC expert panel, Sept 2018)
41. Care team is respectful of my spiritual/religious beliefs	Spiritual, religious, and existential aspects of care	0.20	Bereaved Family Survey (32)
42. Care team is respectful of my cultural beliefs/practices	Cultural aspects of care	0.18	Bereaved Family Survey (32)

Table 5: Final EXPERIENCE@Home Measure

Please choose the answer that best fits your experience with your child’s home-based palliative or hospice care team over the past week.

Your answers will help the care team to better understand if your child’s and family’s most important needs are being met, and to guide the care team to better address those needs.

Note: For all questions, the care team is your child’s home-based palliative care or hospice team (that is, the nurses, physicians, social workers, chaplains, child life therapists, art therapists who care for your child and family in your home).

Use this scale for all questions:		Strongly Disagree	Disagree	Neutral	Agree	Strongly Agree	Does not apply
1	The care team treats my child's physical symptoms so that my child has as good a quality of life as possible.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2	The care team helps me to feel confident in managing my child's symptoms at home without needing to go to the hospital.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3	My care team provides the care or guidance we need to keep my child at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4	I trust the care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5	The care team works together with me and my child to make decisions about my child's medical care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6	The care team asks for my opinions and	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

	concerns about my child.						
7	The palliative care or hospice team works closely with my child's other providers (for ex, neurology, cardiology, oncology, primary care) to support our family's goals for my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8	The care team gives me enough information to make informed health care decisions about my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9	I have regular access to on-call services from our care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10	I have access to a provider who can help me coordinate my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11	The care team considers all of my child's needs.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12	The nurses have the knowledge, skills, and experience to support my child's palliative or hospice care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13	The doctors and nurse practitioners have the knowledge, skills, and experience to support my child's palliative or hospice care at	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

	home.						
14	The care team provides the information and strategies to effectively manage my child's pain and other physical symptoms at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15	The care team helps adapt our home to better support my child's current care needs.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
16	The care team provides opportunities for my child to express his/her feelings, such as fears, worries, and hopes.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
17	The care team provides support for <u>my other children's</u> feelings and emotions.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
18	The care team provides support for <u>my</u> feelings and emotions, including fear, worry, sadness, and hope.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
19	The care team helps me hope for the best possible outcome for my child's health, while also helping me prepare if that outcome doesn't happen.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
20	The care team has provided or	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

	directed me to resources that support my needs as my child's caregiver.						
21	I can talk about my child's end of life with the care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
22	The care team has talked with me about my child's last weeks of life and what they may be like.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

***Bolded items** are additional items based on Phase 4 analysis

Figure 1: Item Selection and Reduction Process

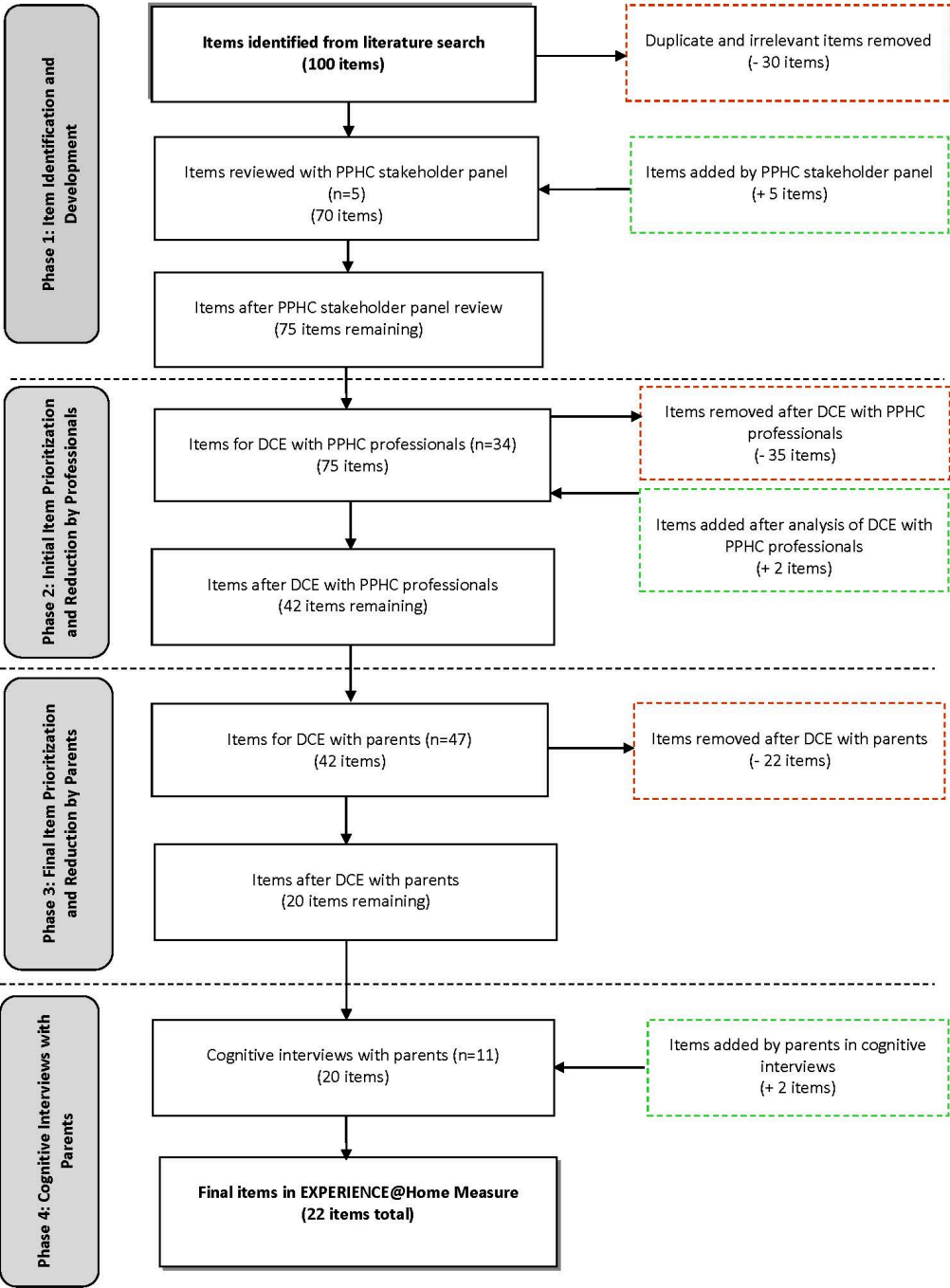


Figure 2: Sample DCE Choice Set


Considering only the following 4 items, please choose the one you feel is the **Most Important, and the one you feel is the **Least Important** aspect of care provided by a home-based pediatric palliative and hospice care team:**

(1 of 20)

Most Important		Least Important
<input type="radio"/>	Care teams asks for my opinions and concerns about my child	<input type="radio"/>
<input type="radio"/>	Care team has prepared me for what my child's last weeks of life may be like	<input type="radio"/>
<input type="radio"/>	Access to on-call service from palliative care or hospice team	<input type="radio"/>
<input type="radio"/>	My child receives complementary and alternative medicine	<input type="radio"/>

** Care team refers to the members of a home-based palliative care or hospice team (for example, nurses, physicians, social workers, chaplains, child life therapists, art therapists, etc.)

Click the 'Next' button to continue...

0%  100%

Supplemental Table 1: Items Prioritized by PPHC Professionals (Phase 2)

****Bolded** items were retained for evaluation in next phase*

Item	Score	Domain [^]	Original Instrument
1. I trust the care team	3.50	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
2. Access to on-call service from palliative care or hospice team	3.04	Access to care team	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
3. Care team helps me do the best for my child	3.01	Relationship between family and care team	New item (PPHC expert panel, Sept 2018)
4. Care team helps me hope for the best outcome while also helping me prepare in case that outcome does not happen	2.94	Psychological/e motional aspects of care: <u>Parent(s)</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
5. Care team works together with me and my child to make medical decisions	2.88	Relationship between family and care team	Parental PELICAN Questionnaire (Zimmerman, 2015) (21); Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
6. Care teams are all working towards same goals for my child's care	2.84	Continuity of care	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
7. I feel prepared to treat my child's symptoms at home	2.79	Physical aspects of care: Symptom management	Massachusetts PPCN Evaluation (Bona, 2011) (10)
8. Care teams asks for my opinions and concerns about my child	2.69	Communication between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
9. Care team looks at all of my child's needs	2.66	Continuity of care	Quality of Children's Palliative Care Instrument (Widger,

			2015) (47)
10. Information shared between <u>me and the care team</u> is clear	2.63	Communication between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
11. Care team takes time to listen carefully	2.63	Communication between family and care team	Consumer Assessment of Healthcare Providers and Systems (CAHPS) Hospice Survey (67)
12. My child can easily get necessary care	2.50	Access to care team	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
13. Care team is kind, caring, and respectful	2.47	Relationship between family and care team	Bereaved Family Survey (32)
14. Care team talks with me about my fears and worries	2.42	Psychological/e motional aspects of care: <u>Parent(s)</u>	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
15. Care team provides information about treatments for my child's pain and other symptoms	2.42	Physical aspects of care: Communication	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
16. Care team uses medicines to ease my child's pain and other symptoms.	2.40	Physical aspects of care: Symptom management	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
17. Care team gives me enough information to make good health care decisions	2.34	Communication between family and care team	Parental PELICAN Questionnaire (Zimmerman, 2015) (21); Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
18. Care team is sensitive to my and my family's feelings	2.26	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
19. I can talk about my child's end of life with care team	2.23	Communication at end of life	Parental Questionnaire 1 (Vollenbroich, 2012)

			(20)
20. Knowledge/skills of nurse(s)	2.16	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
21. I have access to care provider who can coach or guide me to care for my child	2.11	Care coordination	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
22. Care team has prepared me for what my child's last weeks of life may be like	1.97	Caregiver support at the end of life	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
23. Care team is respectful of my cultural beliefs/practices	1.91	Cultural aspects of care	Bereaved Family Survey (32)
24. Care team helps me cope with the stress of caregiving	1.88	Social aspects of care: <u>Parent(s)</u>	NCP 4th Edition Domains (24)
25. Information shared <u>across care teams</u> is clear	1.81	Continuity of care	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
26. Care team helps me to advocate for my child's needs	1.80	Relationship between family and care team	New item (PPHC expert panel, Sept 2018)
27. Care team helps me talk with my child about death and dying	1.60	Communication at end of life	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
28. Care team helps me stay involved in my child's care	1.57	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
29. Care team treats my child as a unique person	1.38	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
30. Care team provides emotional support for me	1.36	Psychological/e motional aspects of care: <u>Parent(s)</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)

31. It is easy to contact the care team	1.30	Access to care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
32. Care team helps me talk about whether to stop life-sustaining measures	1.30	Ethical and legal aspects of care	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
33. I receive the same information about my child across providers	1.12	Continuity of care	Quality of Children's Palliative Care Instrument (Widger) (47)
34. Care team helps me adapt my home to support my child's care needs	1.12	Care coordination	IOM 2003 report (16); NHPCO 2009 standards (26)
35. Information shared between care team and my child is clear	1.09	Communication between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
36. Care team accepts me and my family	1.09	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
37. Care team is respectful of my spiritual/religious beliefs	1.09	Spiritual, religious, and existential aspects of care	Bereaved Family Survey (32)
38. Care team knows medical details about my child's condition	1.01	Communication between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
39. Care team guides me on how to support my other children	0.99	Social aspects of care: <u>Sibling(s)</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
40. Care team provides emotional support for my child	0.98	Psychological/emotional aspects of care: <u>Child</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
41. Care team helps me find resources to balance work, family, and caregiving demands	0.97	Social aspects of care: <u>Parent(s)</u>	NCP 4th Edition Domains(24); Massachusetts PPCN Evaluation (Bona, 2011) (10)

42. Care team provides support for my spiritual needs	0.92	Spiritual, religious, and existential aspects of care	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
43. I can communicate with the care team in my preferred language	0.91	Cultural aspects of care	New item (PPHC expert panel, Sept 2018)
44. Care team provides emotional support for my other children	0.87	Psychological/emotional aspects of care: <u>Sibling(s)</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
45. Same care providers consistently work with my family	0.84	Continuity of care	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
46. Care team helps me talk about my child's preferred place of death	0.84	Communication at end of life	Quality indicators for paediatric palliative care (Charlebois, 2015) (68)
47. Care team coordinates my child's care	0.84	Care coordination	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
48. I feel a close connection with the care team	0.82	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
49. Care team helps me to use non-drug measures to ease my child's pain and other symptoms	0.79	Physical aspects of care: Symptom management	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
50. Care team spends enough time with me	0.77	Relationship between family and care team	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
51. Care team helps with getting needed community-based services	0.75	Care coordination	Quality of Children's Palliative Care Instrument (Widger, 2015) (47); Massachusetts PPCN Evaluation (Bona, 2011) (10)
52. Care team helps me talk about whether to stop non-helpful	0.73	Ethical and legal aspects of care	Parental PELICAN Questionnaire (Zimmerman, 2015)

treatments			(21)
53. I have clearly documented my child's preferences for medical care	0.66	Ethical and legal aspects of care	Widger, 2004 (69); Hinds, 2012 (70)
54. Care team helps with getting my child's medical equipment	0.65	Care coordination	Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
55. Knowledge/skills of physician(s)	0.64	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
56. Knowledge/skills of social worker(s)	0.62	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
57. Care team provides the right amount of support for my social needs (e.g., maintaining relationships with family/friends, identifying social support network)	0.61	Social aspects of care: <u>Parent(s)</u>	Quality of Children's Palliative Care Instrument (Widger, 2015) (47)
58. Care team helps with getting my child's medications	0.61	Care coordination	Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
59. Care team provides opportunities to ask questions	0.59	Communication between family and care team	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
60. Care team helps me find resources to cope with financial strain	0.53	Practical aspects of care	NCP 4th Edition Domains (24); Massachusetts PPCN Evaluation (Bona, 2011) (10)
61. Care team helped make transfer from hospital to home as smooth as possible	0.52	Continuity of care	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
62. Knowledge/skills of spiritual provider(s)	0.43	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)

63. Knowledge/skills of expressive therapist (e.g., art therapist; child life therapist)	0.42	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
64. I have access to respite care to allow me to take a break	0.42	Continuity of care	Parental PELICAN Questionnaire (Zimmerman, 2015) (21); Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
65. I have documented and shared advance care planning decisions with the care team	0.33	Ethical and legal aspects of care	New item (PPHC expert panel, Sept 2018)
66. My child receives complementary and alternative medicine	0.32	Physical aspects of care: Symptom management	Parental PELICAN Questionnaire (Zimmerman, 2015) (21)
67. Care team helps me work with my child's health (insurance) plan	0.23	Practical aspects of care	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
68. Care team deals with administrative problems that impact my child's care	0.22	Care coordination	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
69. Care team helps with issues with housing and utilities	0.22	Practical aspects of care	Community PedsCare HRQoL instrument (Goldhagen, 2016) (23)
70. Care team helps me fit care of my child's illness into our usual family routine	0.21	Social aspects of care: <u>Parent(s)</u>	New item (developed based on recommendation from dissertation committee; Oct 2019)
71. Knowledge/skills of physical, occupational, speech therapist	0.11	Knowledge and skills of care team providers	Parental Questionnaire 1 (Vollenbroich, 2012) (20)
72. Care team provides emotional support for my child's extended social network (e.g., classmates, neighbors, extended	0.10	Emotional aspects of care: <u>Extended social network</u>	New item (PPHC expert panel, Sept 2018)

family)			
73. Care team helps prepare my child for school	0.09	Social aspects of care: <u>Child</u>	Seattle Pediatric Palliative Care Project evaluation (Hays, 2006) (30)
74. Care team keeps me informed about their arrival time	0.07	Communication between family and care team	Consumer Assessment of Healthcare Providers and Systems (CAHPS) Hospice Survey (67)
75. Care team helps with arranging transportation	0.05	Care coordination	NCP 4 th Edition Domains (24)

[^]See Table 1 for domain descriptions

Supplemental Table 2: Demographic and Clinical Characteristics of Parent Participants and Their Children (Phase 4)

Parents' Characteristics (n = 11)		No. (%)
Parent type	Mother	10 (90.9%)
	Father	1 (9.1%)
Age	<i>Mean (SD)</i>	43.8 (6.5)
Race	White	11 (100%)
Ethnicity	Non-Hispanic	8 (72.7%)
	Hispanic	2 (18.2%)
	Prefer not to answer	1 (2.1%)
Highest Education Level Completed	Some college	3 (2.1%)
	Associates / Professional	1 (9.1%)
	Bachelor's	3 (27.3%)
	Master's	4 (36.4%)
Relationship Status	Married / partnered	9 (81.8%)
	Separated / divorced / Widowed	1 (9.1%)
	Prefer not to answer	1 (9.1%)
Number of Other Children	0	1 (9.1%)
	1	6 (54.5%)
	2	4 (36.4%)
Bereavement Status	Bereaved	7 (63.6%)
	Currently caring for child at home	4 (36.4%)
Affiliation	CHOP	2 (18.2%)
	CPN	9 (81.8%)
Children's Characteristics (n = 11)		No. (%)
Age	<i>Mean (SD)</i>	9.0 (6.4)
Gender	Female	3 (27.3%)
	Male	8 (72.7%)

Race	White	8 (72.7%)
	More than 1 race	2 (18.2%)
	Prefer not to answer	1 (9.1%)
Ethnicity	Non-Hispanic	8 (72.7%)
	Hispanic	2 (18.2%)
	Prefer not to answer	1 (9.1%)
Primary complex chronic condition <i>(Note: not mutually exclusive; thus, the % does not sum to 100%)</i>	Cancer	1 (9.1%)
	Genetic or congenital	4 (36.4%)
	Metabolic	1 (9.1%)
	Neurologic, neuromuscular, or mitochondrial	7 (63.6%)
	Other/Unknown	1 (9.1%)
Primary care team (hospice v. palliative care)	Hospice	2 (18.2%)
	Palliative Care	8 (72.7%)
	Unknown/Not sure	1 (9.1%)
Length of time receiving home-based palliative or hospice care	6 months or less	2 (18.2%)
	7 to 12 months	2 (18.2%)
	1 to 2 years	0 (0%)
	More than 2 years	7 (63.6%)

CHAPTER 4: Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home) Project (Paper 3)

An Application for the National Institute of Nursing Research Ruth L. Kirschstein National Research Service Award (NRSA) Individual Postdoctoral Fellowship (Parent F32)

Sponsor: Chris Feudtner, MD, MPH, PhD

Co-Sponsor: Mary Ersek, PhD, RN, FPCN

Submitted: December 2019

Impact Score: 27

Funded: July 2020

SPECIFIC AIMS

Children with serious life-threatening illnesses (SLTIs), or illnesses that may or may not have potential curative or life-prolonging treatments but all-too-often result in death (1), are generally living longer with significant and complex care needs.(2-7) Every day, an estimated 5,000 U.S. children are living within the last six months of their lives (8), and approximately 15,000 children a year die of conditions that could benefit from specialized pediatric supportive care.(9) Pediatric palliative care (PPC) is child and family-centered care that “optimizes quality of life by anticipating, preventing, and treating suffering.”(2) National organizations such as the American Academy of Pediatrics (10) and the Institute of Medicine (7) have given particular attention to PPC provided in the home.(3, 6, 8, 11-15) **Pediatric palliative and hospice care provided at home** (PPHC@Home) supports children and families by providing interdisciplinary care across health care and social service providers and settings.(7, 10, 16, 17)

Many children with SLTIs and their families are not receiving consistently high-quality care in the home setting.(7, 18-20) Sub-par PPHC@Home has far-reaching implications for child and family quality of life, satisfaction with care, family bereavement outcomes, cost of care, and health care utilization.(21-27) With growing numbers of children cared for primarily at home, **we need to standardize the assessment of families’ experiences with PPHC@Home to evaluate whether the care provided is aligned with what they value and need the most.** Pediatric palliative and hospice care (PPHC) providers can use these assessments to improve outcomes and quality of care for each child and family.

In our current study, we developed the Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home) Measure, which is the first known standardized instrument to assess families’ experiences with PPHC@Home in the U.S. We conducted a preliminary examination of the content validity of this measure, but the feasibility, acceptability, clinical utility, and psychometric properties of this instrument for use in a broader sample of families receiving PPHC@Home is currently unknown. Additionally, while previous research on PPC outcomes evaluation have primarily been completed with bereaved parents (27-31), the **assessment of real-time patient and family experiences** may allow for more timely and effective management of children’s and family’s needs in the home setting.(7, 32-37) We therefore propose to evaluate the EXPERIENCE@Home Measure from the perspective of parents whose children are currently receiving PPHC@Home, as well as from the perspective of providers who will ultimately be implementing this tool in clinical practice through three specific aims:

Aim 1: Evaluate evidence for score validity and reliability of the EXPERIENCE@Home Measure from parents of children with serious life-threatening illnesses currently receiving PPHC@Home.

Method: We will administer study measures at two time points: baseline (n=100) and re-test (n=30).

Aim 2: Evaluate the feasibility and acceptability of using the EXPERIENCE@Home Measure in real-time in parents of children with serious life-threatening illnesses receiving PPHC@Home.

Method: We will conduct brief interviews with participants to evaluate acceptability, and analyze descriptive statistics (measure completion, missing items, time for completion) to evaluate feasibility.

Aim 3: Evaluate the perceived clinical utility of the EXPERIENCE@Home Measure with an interdisciplinary group of pediatric palliative and hospice care stakeholders.

Method: We will conduct weekly meetings with interdisciplinary PPC providers, as well as quarterly co-design sessions with an interdisciplinary group of PPHC stakeholders.

Impact: The proposed project is the first to examine real-time use of an instrument developed to specifically evaluate family-reported PPHC@Home experiences in the U.S. The aims of this project closely align with the NINR's strategic research plan and funding priorities to advance high-quality, evidence-based palliative care for children and families through the development of effective ways to screen, assess, and monitor the met and unmet needs of patients and families (38), particularly in the community setting.(39) Ultimately, the EXPERIENCE@Home Measure will facilitate the timely assessment of the needs of children and families by PPHC@Home teams. We therefore have the unique opportunity to address gaps in care at the time care is provided, thereby improving the care we provide to children with SLTIs and their families living at home.

RESEARCH STRATEGY

Significance

Pediatric palliative and hospice care at home (PPHC@Home) strives to meet the health care needs of children with serious life-threatening illnesses (SLTIs) within the broader contextual influences of children’s familial and social environments.

These children and families are supported by complex networks of hospital-based, outpatient, and home-based providers and services (6, 40), and team composition and services are influenced by state and local regulations and resources. Unsurprisingly, considerable variation exists in the provision and quality of PPHC@Home in the U.S. (18-20, 41)

While children and families need consistent, high-quality care, experts are concerned that they may not receive consistently high-quality palliative and hospice care at home.(7, 19, 20, 42) Suboptimal PPC is associated with negative child and family outcomes, including an increase in potentially avoidable hospitalizations (27), “profound and lasting emotional distress” (43), and persistent parental guilt in the year following their child’s death.(44) On the other hand, perceived good quality of care is associated with improved child and family quality of life, higher satisfaction with care, decreased cost of care, and decreased health care resource utilization.(21, 23, 24, 26, 27, 45) Thus, the quality of palliative care we provide to children and families matters, irrespective of the care setting.

The first step to ensuring high-quality PPHC@Home for all children is measuring families’ experiences with PPHC@Home to assess whether their experiences are aligned with what they value and need the most. In our current study, we developed the Experiences of Palliative and Hospice Care for Children and Caregivers at Home (EXPERIENCE@Home) Measure, which is the first known standardized instrument to assess families’ experiences with PPHC@Home in the U.S. After conducting a preliminary examination of the content validity of this measure (paper under review), we aim to complete further psychometric and usability testing in this proposed project.

Innovation

The EXPERIENCE@Home Measure is the first known multi-dimensional family-reported measure developed to assess experiences with U.S.-based PPHC@Home programs. Additionally, it is among the first measures designed to survey parents whose child is currently receiving PPHC@Home services.(46) Existing measures of care experiences in adult and pediatric palliative care typically survey families after the patient has died.(29, 47-49) While this approach is valuable for improving care for future children and families, it cannot be responsive to the care needs of children and families currently receiving care. **Evaluating care in real-time affords pediatric palliative and hospice care (PPHC) providers the unique opportunity to monitor and respond to child and family needs, and to perceived gaps in care, at the time care is provided.**(32, 33, 50, 51) The proposed training plan and the opportunity to use innovative methods, such as real-time collection of data (that is, collecting data directly from families who are currently receiving care (32, 51)) and use of experience-based co-design (an approach that seeks to improve care experiences through the incorporation of patients, family members, and staff perspectives on their experiences (52-54)), will help the applicant build a foundation for future intervention-based work in improving PPHC@Home for children and their families.

Approach

We will evaluate the psychometric properties, feasibility, and acceptability of the measure from the perspective of parents of children currently receiving PPHC@Home (Aims 1 and 2), as well as the clinical utility from the perspective of interdisciplinary PPHC stakeholders (providers, parent advocates) (Aim 3).

CONCEPTUAL FRAMEWORK: We will use an adapted version of domains from the National Consensus Project (NCP) Clinical Practice Guidelines for Quality Palliative Care (4th edition) (55) and key domains of quality PPC (10, 40, 56, 57) to guide our work. We have further adapted these guidelines based on our current study for a total of 4 overall domains and 12 sub-domains (Fig 1).

PRELIMINARY DATA: In my current dissertation project, we used a discrete choice experiment (DCE) with “Bandit” maximum difference (MaxDiff) scaling to identify domain-specific aspects of PPHC@Home that parents value the most. We began with a pool of 75 items related to PPHC@Home, drawn from previously developed outcome measures and a literature review. We asked a sample of 34 interdisciplinary experts in PPHC (nurses, advanced practice nurses, physicians, chaplains, social workers, expressive therapists, researchers, and parent advocates) to perform the preliminary importance weighting and ranking, using a DCE to winnow the initial item pool to a smaller set of items to reduce the time demand placed on parent participants later in the study. In brief, each provider participant was taken through 38 sets of items, and for each set, participants chose the most and least important item among each set of 4 items. Through the Bandit MaxDiff approach, items were selected for each new respondent using prior estimates of each item’s mean and variance via Thompson sampling, which increases the precision of item-weight estimates.(58) Provider participants were also asked to rate and rank the key domains of PPHC@Home (Fig 1). We determined the average provider ratings and rankings of the items using Lighthouse Studio Version 9.6.1 (Sawtooth Software, Inc., Sequim, WA). Leveraging both domain- and item-level importance scores, we winnowed the 75 initial items to a reduced set of 42 of the most highly-rated items by domain importance. We then asked 47 parents of children who were either currently receiving PPHC@Home for their child (n = 33), or who had previously received PPHC@Home for their child (n = 14) across the U.S. to complete the same process of rating and ranking the domains and 42 items. To winnow these items to the most highly-rated items, we first accounted for items that were highly ranked but the corresponding domain was ranked low (or vice versa, where the domain was ranked high but the item(s) ranked low) by calculating weighted item scores (multiplying item scores by corresponding domain scores). We then retained the top 20 items by weighted importance score (capping domains at a maximum of 2 items to avoid redundancy and ensure domain coverage). Finally, we asked a sample of 11 parents, including bereaved parents and parents currently receiving care for their child, to participate in cognitive interviewing, where, through structured interviews, parents provided preliminary data on the instrument’s content validity. Based on parents’ feedback, we revised the wording of most items and added 2 items for 22 final items. Selected items and associated domains are highlighted in Table 1.

Parent participants in Aim 3 were also asked to complete the instrument in its current form. The mean score for participants (n=11) was 78.3 (SD 29.5) out of 100 possible points (with 100 indicating the best possible experience). Instrument scoring is discussed below (*Aim 1 approach*).

OVERALL DESIGN FOR AIMS 1 AND 2:

Design. Employing a prospective design, parents of children currently receiving PPHC@Home will complete 3 data collection points: A) a baseline assessment of all study measures (Table 2) to evaluate evidence for validity based on hypothesized relationships to external variables and reliability based on internal consistency (*Aim 1 approach*), B) a re-test of the EXPERIENCE@Home Measure within 48-72 hours to evaluate evidence for reliability based on temporal stability (*Aim 1 approach*), and C) an interview following re-test completion to evaluate acceptability (*Aim 2 approach*). We will evaluate feasibility based on survey completion rates, missing item rates, and time for survey completion (*Aim 2 approach*).

Sample. Parent participants will be recruited from the Children's Hospital of Philadelphia (CHOP) and the Courageous Parents Network (CPN). Parents from CHOP will be recruited in collaboration with the Pediatric Advanced Care Team (PACT) which serves, on average, 300 new patients per year, with more than 50 patients enrolling in either home hospice or dedicated community-based palliative care services each year. CPN is a virtual network of parents, families, and providers, with nearly 4,000 unique users every month, 6,780 followers on Facebook, and 750 official parent members. CPN offers families the information, skills, tools, and virtual support they need during their child's illness journey through education, community, and advocacy.

Recruitment. Eligible participants from CHOP will be identified and recruited by the PI, with assistance from PACT. Staff from CPN will assist with posting recruitment materials to their blog and social media page. We will use purposeful sampling strategies to ensure minority representation in our sample.

Inclusion and Exclusion Criteria. Inclusion criteria: Child has a serious life-threatening illness (1) and is aged 25 years or younger; parent/caregiver is over the age of 18 years; Child is currently receiving PPHC@Home services; Parent is able to read, write, and speak English. Exclusion criteria: The child and parent/family caregiver do not reside in the same household; Parent/caregiver is not the legal guardian and/or primary decision-maker for the child.

APPROACH FOR AIM 1: Evaluate evidence for score validity and reliability on the EXPERIENCE@Home Measure from parents of children with serious life-threatening illnesses currently receiving palliative or hospice care at home.

Study Procedures. The PI will contact all potential CHOP participants by phone and provide information about the study. If the parent agrees to participate, the PI will obtain an email address to send study instruments electronically via REDCap. If the parent prefers to participate in person or by phone/video, the PI or trained research assistant (RA) will determine with the parent the preferred meeting date and location and complete all study instruments. Interested CPN parents will contact the PI directly by phone or email. After screening for eligibility and confirming interest, the PI or RA will send study instruments electronically via REDCap, or schedule a time to meet and complete study instruments by phone or video. For all participants: A reminder will be automatically sent after 1 week for incomplete measures. Additionally, on the last page of the electronic survey, participants will be given the option to opt-out from being re-contacted for re-test. If a participant has not opted-out of the re-test, an invitation for a re-test of the EXPERIENCE@Home Measure and the Child's Health Status will be sent 48-72 hours after the baseline completion. While there is no standard timeframe for

administration of a re-test in the health measurement literature, we anticipate that this timeframe will be long enough to minimize carry-over effect, yet short enough to minimize the probability of significant changes in the child's health from baseline to re-test.(59) All participants will receive a \$10 gift card for the initial completion of all study instruments and a \$10 gift card for completion of the re-test.

Study Measures. The 22-item EXPERIENCE@Home Measure (see Appendix) will be administered at baseline and at re-test. Each item is measured on a 5-point scale (1 = Strongly Disagree, 2 = Disagree, 3 = Neutral, 4 = Agree, 5 = Strongly Agree). All items have an ascending positive valence (care is perceived as better) as the scores increase. To ease interpretation, scores will be linearly transformed on a 0 to 100 scale (Strongly Disagree = 0, Disagree = 25, Neutral = 50, Agree = 75, Strongly Agree = 100).(59, 60) Two scores will be generated: the worst-item score and the average total score (mean of all completed items; any items scored "Does Not Apply" or missing will not be included). Taken together, these two scores indicate not only a family's overall average perceptions about all measured domains of care (average total score), but also the maximum extent of a family's negative perceptions about any aspect of care (worst-item score). This knowledge could allow a care team to identify which aspects of care are potentially the most problematic for the child and family, and where intervention could make the greatest impact.(61) We will also include an unscored write-in option for "Additional concerns not elsewhere addressed on this instrument." In addition to the EXPERIENCE@Home Measure, participants will also complete the instruments described in Table 2.

Data Collection. The baseline data collection will include the completion of the EXPERIENCE@Home Measure, other relevant measures (Table 2) and a demographic questionnaire. The second data collection point will be the re-test of the EXPERIENCE@Home Measure, in addition to the Child Health Status question (Table 2) and a single question asking participants to list any home-based visits since the first assessment.

Sample Size. Using the commonly accepted sample size guidelines of 5 to 10 participants per item (62, 63), we will aim to recruit at least 100 parents (50 from CHOP, 50 from CPN). To evaluate stability of scores (test-retest), we will aim for at least 30 parents to re-take the instrument for an ICC = 0.90 (SE 0.05).(59)

Analysis Plan. We will describe item-level statistics for each scale item, including means, variances, minimum/maximum, missingness, and inter-item correlations. We will evaluate evidence for reliability based on internal consistency (i.e., do items measure the same construct? (71, 72)) for the total score and within the 4 domains (Fig 1) (Cronbach's α). We anticipate that the total score reliability will be lower than the domain reliabilities, given that the total score encompasses very different domains; therefore, we predict that the α for the total score to be between 0.50 and 0.60, and the α for the domain scores to be ≥ 0.70 .(31, 67, 73-75) We will evaluate evidence for reliability based on temporal stability (i.e., does measure produce similar results from test to retest? (71)) for parents who complete both administrations via intraclass correlation coefficient (ICC) estimates with 95% confidence intervals. We will account for changes, baseline to re-test, in the child's health status item, and intervening home-based visits, to assure that test-retest is not confounded by these factors.

We evaluated content validity (72) through cognitive interviews with parents in our current study. We will also evaluate evidence for validity based on hypothesized relationships to external variables (i.e., do scores correlate with scores from another outcome for which a relationship is expected?).(71, 72) We will first compare scores from our instrument against a criterion measure of satisfaction. Similar to previous PPC instrument development studies (29, 31), we will ask parents to answer a question: “Overall, how satisfied are you with your child’s home-based care over the last week?” To ensure that each item in the EXPERIENCE@Home Measure is appropriately correlated with this criterion, after adjustment for other items, we will compute the correlation (Pearson correlation coefficient) between the score on each item on our instrument and the criterion measure of satisfaction to evaluate consistency of scores from our measure against the criterion measure. Items with correlation <0.30 will be considered for deletion.(31, 59) Additionally, each item should add to our ability to estimate the criterion response by adding information about a different domain of PPHC@Home (Fig 1), telling us about the completeness of scores across instrument domains against our criterion measure of satisfaction. Using a model-fitting approach, we will use the importance weights for each domain (estimated in our current study), and evaluate the extent to which each of the items in the measure will have explanatory value proportional to their importance weights. Our hypothesis is that in no instance should reporting better care in one domain be associated with lower scores on this criterion, after adjustment. We will also evaluate evidence for validity based on hypothesized relationships to external variables by calculating the disattenuated correlations (Pearson correlation coefficient) of the EXPERIENCE@Home score with the Desire for Care Improvement, Kessler-6, and Family Life Difficulty Item (Table 2). Based on existing literature and clinical expertise, we will test the following hypotheses (Table 2): a) a significant positive relationship between items on the EXPERIENCE@Home Measure and satisfaction with care (29, 31); b) significant negative relationships between the EXPERIENCE@Home Measure and desire for care improvement (64), parental distress (66), and family life difficulty.(68, 69) All statistical analyses will be performed using the statistical software package Stata/IC 15.1.

APPROACH FOR AIM 2: Evaluate the feasibility and acceptability of using the EXPERIENCE@Home Measure in real-time in parents of children with serious life-threatening illnesses receiving palliative or hospice care at home.

Sample Size / Sampling Plan. We will conduct interviews with at least 15 participants from Aim 1 (or until we reach data redundancy and thematic saturation), recruited via purposive criterion-based sampling strategies.

Data Collection. Upon completion of all study measures or if a participant decides to end participation (Aim 1), the PI or RA will contact each participant to request a brief (15-30 minute) interview (in-person or phone/video). If a parent agrees to participate, the PI or RA will schedule a meeting. To evaluate acceptability, participants will be asked specific questions during semi-structured interviews about his/her experience with completing the instrument, the overall perceived acceptability and utility of the survey, and any perceived barriers/facilitators to future implementation of the instrument. Interview questions are guided by Proctor’s Implementation Outcomes (76) and the Consolidated Framework for Implementation Research (77) (*draft interview guide in Appendix*). Interviews will be audio-recorded and professionally transcribed. Participants will receive a \$15 gift card for completion of the interview. We will assess feasibility using

data collected from the REDCap platform (Aim 1), including time for survey completion, percent of surveys completed, and rates of missingness of items.

Analysis Plan: We will analyze interview data inductively using a content analytic approach to identify codes, categories, and themes.(78) The PI will read through each interview, consider the topics addressed in the interview guide, and code each interview with preliminary codes. To ensure rigor of the analytic process, the PI and RA will independently code randomly selected interviews, and the PI, Dr. Deatruck (Consultant), and RA will meet to discuss the coding, reconcile any discrepancies, and revise the codebook. Interviews will be re-coded, and across interviews, the PI and Dr. Deatruck will construct informational matrices to organize the data, group codes into larger categories, and then develop the categories into themes. Themes will be discussed with the larger study team, and conclusions about the acceptability of the EXPERIENCE@Home Measure will be drawn. We will conduct all qualitative analyses within Atlas.ti. We will run descriptive statistics on time for survey completion, missing surveys, and missing items within REDCap.

APPROACH FOR AIM 3: Evaluate the perceived clinical utility of the EXPERIENCE@Home Measure with an interdisciplinary group of pediatric palliative and hospice care stakeholders.

Design. The ultimate goal for the EXPERIENCE@Home Measure is to give PPHC providers the ability to monitor and respond to child and family needs and perceived gaps in care during the time care is provided. To ensure that the measure is clinically useful, we will obtain PPHC stakeholders' perceptions on the perceived clinical utility through A) weekly meetings with interdisciplinary providers from CHOP's PACT team who support the home-based/outpatient families, and B) quarterly experience-based co-design meetings. Experience-based co-design is a design process that incorporates users' experience – leading service design alongside patient, family, or staff “users” – to improve the user's experience.(54) We will use a modified version of this process used in a previous study with clinicians to design interventions for introducing PPC.(52)

Provider Sample and Sampling Plan. CHOP's home-based and outpatient PPHC patients are supported by the following members from the PACT team: 4 attending physicians, 2 nurse practitioners, 2 nurse coordinators, 1 art therapist, 1 child life therapist, and 2 social workers. For the weekly meetings, we aim to include at least 2 interdisciplinary PACT members. For the co-design group, we will aim to recruit at least 3 physicians, 2 nurse practitioners, 1 nurse coordinator, 1 social worker, and 1 expressive therapist, as well as 2 parent advocates (hospital employees or volunteers who has, or has had, a child who received PPHC@Home services) and 2 community-based PPHC providers, who will be identified with assistance from PACT.

Procedures and Data Collection. Each family enrolled from CHOP will have a report generated of their EXPERIENCE@Home Measure scores. The PI will attend PACT team rounds weekly. Together with at least 2 members of the PACT team who support home-based patients, we will review the reports from newly enrolled families over the previous week, and discuss the following questions: 1) what is already known versus new information; 2) what information is clinically actionable over the short-term; 3) what information may be clinically actionable over the long-term; and 4) what information is not clinically actionable and why (*meeting guide in Appendix*). Each meeting will be audio-recorded for reference, and the PI will write-up main discussion points after each meeting. Main discussion points will be compiled. On a quarterly basis, the co-design

group will convene in-person or by video. At each of 4 co-design sessions, the group will review the PI's de-identified summary notes from the previous quarter, and discuss the clinical utility of the instrument, as well as facilitators and barriers to implementation of the instrument in clinical practice (*co-design interview guide in Appendix*). Each co-design session will be led by the PI and audio-recorded, and a RA will observe and take descriptive observation notes on the conversation, noting any salient issues or recommendations introduced by the group. Audio recordings will be professionally transcribed. A meal will be provided at each session.

Data Analysis. We will employ a similar analytic approach as Aim 2 above to analyze the transcriptions and field notes. Following each co-design session, the PI will discuss major issues or recommendations with the larger study team. The PI will write-up final issues and recommendations at the conclusion of Aim 3.

Strengths and Challenges

Sampling Plan for Parents. Recruiting parents may be challenging, given the stress of caring for a seriously-ill child at home.(26, 27, 79-81) CHOP's PACT has, however, extensive experience in working with this population in clinical and research capacities. The PI also has experience working with this population in her F31-funded dissertation project. Recruiting from CPN's online community will expand recruitment to parents outside CHOP's system and increase the sample's geographic diversity. This is important, given different issues that may affect PPHC@Home experiences, including local/state resources, access to and organization of care, and regulations and funding.(82) While it is not within the scope of this project to investigate differences across geographical areas, recruiting geographically diverse families helps to ensure that the EXPERIENCE@Home Measure is tested in a diverse sample, which sets the stage for future multi-site work.

Sampling Plan for Providers. This proposed study will require a substantial commitment of time and effort from PACT team members. The PI, however, has worked and established professional relationships with many members of the team over the course of her F31 project. Nevertheless, the PI will work to ensure that study meetings fit within existing clinical schedules (e.g., keeping meetings brief and on target, attending team rounds on days that are clinically less busy).

Real-Time Assessment. Only one previous study, conducted in Canada by Dr. Widger (Consultant), evaluated an instrument developed to measure experiences and satisfaction with PPC with families who are currently receiving care.(83) Our proposed study will be the first known to evaluate real-time responses in parents receiving PPHC@Home in the U.S. This project will also be the first step in implementing this measure for use in real-time monitoring of care.

Co-design Approach. The experience-based co-design approach, while used in other areas of health care (54, 84-86), has not been extensively used in the PPC context. One previous study utilizing this approach for improving a PPC intervention within pediatric oncology observed that the approach allowed participants to learn from each other's experiences, perspectives, and perceived barriers; increased buy-in from participants; and resulted in the design of a tailored, clinically-actionable intervention (PI: Feudtner).(52) We will employ a co-design approach with PPHC clinicians and parent advocates in this study to help ensure that the EXPERIENCE@Home Measure will be acceptable and clinically-useful for families and providers in the future.

Implications. The EXPERIENCE@Home Measure will fill an important gap in outcomes measurement by comprehensively evaluating, in real time, what families caring for their children with SLTIs experience in the home setting. Developing measures to “accurately assess and track palliative care delivery and outcomes in home and community settings,” particularly in pediatrics, is well in-line with NINR’s Strategic Research Plan (38) and funding priorities.(39) This study serves as a timely and necessary stepping stone to future intervention-based research in measuring, and improving, PPHC@Home care quality for children and families.

Future Directions. This project and training plan will serve as a foundation for a future independent research program in PPHC@Home, including a K-23 career development award that will build the bridge into clinical intervention-based research in PPC, particularly in the home and community setting. Because measuring perceptions of care is only assessing one aspect of quality (87), future work will evaluate evidence for the validity of the instrument against other measures, such as symptom assessment outcome measures and clinical/administrative reviews (88), as well as test the feasibility of collecting data over time in repeated measurement. Additionally, we anticipate further work in refining the instrument’s scoring and interpretability of scores (i.e., developing a “cutpoint” for clinical action). Finally, the PI will develop a self-report version of the EXPERIENCE@Home Measure to better understand perceptions of children who are able to self-report.(89)

PROTECTION OF HUMAN SUBJECTS

1. Risks to Human Subjects

A. Human Subjects Involvement, Characteristics, and Design

The proposed study seeks to evaluate the psychometric properties, feasibility, and acceptability of the EXPERIENCE@Home Measure in parents of children receiving pediatric palliative or hospice care in the home setting (PPHC@Home), as well as the clinical utility in a sample of interdisciplinary pediatric palliative and hospice care (PPHC) stakeholders. This project will be conducted using web-based surveys and phone/video-enabled interviews for parent participants, and in-person/video-enabled interviews and meetings with stakeholder participants.

There will be four possible types of study participants drawn from two sources:

Children’s Hospital of Philadelphia (CHOP)	Courageous Parents Network (CPN)	Anticipated Numbers
1. Parents or other non-parental primary caregivers (over 18 years of age) of children (birth to 25 years of age) with serious, life-threatening illness who are currently receiving PPHC@Home (<u>Aims 1 and 2</u>)	1. Parents or other non-parental primary caregivers (over 18 years of age) of children (birth to 25 years of age) with serious, life-threatening illness who are currently receiving PPHC@Home (<u>Aims 1 and 2</u>)	Approximately <u>100 total parents</u> (50 from CHOP, 50 from CPN)
2. Interdisciplinary PPC professionals (including physicians, nurse practitioners, nurse coordinators, social workers, expressive therapists) who support the home-based patients and families (<u>Aim 3</u>)		At least 8 (at least 3 physicians, 2 nurse practitioners, 1 nurse coordinator, 1 social worker, and 1 expressive therapist)
3. Parent advocates, who are hospital employees or volunteers, who has or has had a child who received PPHC@Home services (<u>Aim 3</u>)		At least 2
4. Community-based PPC or hospice providers (including		At least 2

physicians, nurses, social workers, child life therapists, chaplains who work primarily in the home setting) (<u>Aim 3</u>)		
----------------------------------------------------------------------------------------------------------------------------------------------	--	--

CHOP-based parents and providers will be recruited through CHOP's Pediatric Advanced Care Team (PACT). CPN-based parents will be recruited through CPN's blog and Facebook page.

Of note, while outcomes of children with serious illnesses are the focus of this study, these children will not directly participate during the proposed study.

Prior to conducting the research, Institutional Review Board approval will be obtained from CHOP.

B. Study Procedures, Materials, and Potential Risks

Study Procedures.

Aims 1 and 2. For CHOP participants: After identifying potential participants, the PI will first check with the clinical team (PACT) to ensure that there are no restrictions on contacting the family about research participation (e.g., the child or family member experienced an acute medical event, the family had expressed a desire to not participate in research, unsafe or particularly challenging family dynamics or circumstances). The PI will then contact all potential CHOP participants by phone and provide information about the study and answer any questions. If the parent agrees to participate, the PI will obtain an email address to send all study instruments electronically. If the parent prefers to participate in person or by phone/video to complete the study instruments, the PI will determine with the parent the preferred meeting date and location; the PI or a trained Research Assistant (RA) will complete the survey instruments during this meeting. For CPN participants, the PI will work with CPN leadership to post all recruitment and eligibility information on the CPN blog and Facebook page. Interested participants will contact the PI directly by phone or email. After confirming interest, the PI or RA will email study materials, or schedule a time to meet and complete all instruments by phone or video. For all participants: on the last page of the electronic survey instrument, participants will be given the option to opt-out from being re-contacted for re-test. If a participant has not opted-out, they will automatically be sent an invitation, within 48-72 hours of the initial completion, to re-take the EXPERIENCE@Home Measure. Participants will receive a \$10 gift card for the initial completion of all study instruments and a \$10 gift card for completion of the re-test.

Upon completion of the study or if a participant decides to end participation at any point, the PI or RA will contact each participant to request a 15- to 30-minute interview (in-person, by phone, or by video). If a parent agrees to participate, the PI or RA will set up a date/time to complete the interview. With each participant's consent, the interview will be audiotaped using digital voice recorders so that the

research team can refer back to specific points of the interview during data analysis. Participants will receive a \$15 gift card for participating in this interview.

Aim 3. We will generate a report of the EXPERIENCE@Home Measure scores for each family enrolled at CHOP. The PI will attend PACT team rounds weekly. Together with at least 2 interdisciplinary members of the PACT team who support home-based patients, the PI will review the reports from newly enrolled families over the previous week, and ask providers for their feedback on the novelty and clinical action-ability of the information. Each meeting will be audio-recorded for reference, and the PI will write up main discussion points after each meeting.

Main discussion points from the weekly meetings will be compiled. On a quarterly basis, the co-design group of providers from PACT (n=8), parent advocates (n=2), and community-based PPC and hospice providers (n=2) will convene, either in-person or by video. At each of the 4 co-design sessions, the group will review the PI's de-identified summary notes from the previous quarter, and discuss questions about the clinical utility of the instrument, as well as questions about the facilitators and barriers to implementation of the instrument in clinical practice. Each co-design session will be led by the PI and audio-recorded, and a trained RA will observe and take descriptive field notes on the conversation, recording any salient issues or recommendations introduced by the group. Audio recordings will be professionally transcribed. A meal will be provided at each co-design session.

Materials. The main source of data for Aim 1 will include the web-based surveys (EXPERIENCE@Home Measure, as well as the Kessler-6, and single items about satisfaction with care, desire for care improvement, family life difficulty, and child's health status) in REDCap. Additionally, a demographic questionnaire will collect information including the parent's age, sex, race/ethnicity, educational attainment, number of children, and marital status, as well as the child's age, sex, race/ethnicity, complex chronic condition diagnostic group (90), and length of time receiving PPHC@Home. The main source of data for Aim 2 will be professional transcriptions of the audiotaped interviews with parent participants and field notes.

The main source of data for Aim 3 will be audiotaped weekly meetings with providers, audiotaped and transcribed co-design sessions with PPHC stakeholders, as well as field notes and summary reports of both the meetings and co-design sessions.

Potential Risks. Study risks for the PPHC professionals are minimal, as their participation is voluntary and involves them providing their expert opinions on family's responses collected using the EXPERIENCE@Home Measure.

Study risks for parents/caregivers include: a) potential psychological distress caused by items on the survey or questions posed in the interview, and b) potential breach of confidentiality. We believe that these potential risks are minimal because previous studies have found that parents caring for a child with a serious illness are willing to participate in research (91), participation in research helped parents discover their "voice" and a sense of empowerment (92), and parents often appreciate the opportunity to help other families through participation in research studies.(93) Our current F31-funded study involving both parents currently caring for

their child and bereaved parents supports these findings. Additionally, any potential breach of confidentiality poses minimal risk, as we are not collecting any information that will reasonably place participants at risk of criminal or civil liability, or could be damaging to participants' financial standing, employability, educational advancement, or reputation.(94) Nonetheless, strategies to mitigate the potential risks are addressed in detail in the section "Protections Against Risks" below.

2. Adequacy of Protection Against Risks

a. Informed Consent and Assent

Parent/Caregiver Participants (Aim 1 – Web-based survey). After confirming interest from participants, the PI will send the participant the link to complete study measures in the research-secure REDCap platform. The first page of the electronic questionnaire will detail the study content and potential risks and benefits of participation. Respondents will then either provide informed consent by clicking "Next Page" and proceeding with the rest of the questionnaire (indicating, by proceeding, their consent to participate), or they will not proceed and not participate in the study.

Parent/Caregiver Participants (Aim 2 – In-person, video, or phone-based interviews). Parent/caregiver participants will view a disclosure statement that will detail the study content and potential risks and benefits of participation. Participants will be informed that their participation is voluntary and that their child's health care will not be affected, whether or not they participate. Participants will also be reminded that they can stop the interview at any time or decline to answer any question. The interviews will also require consent to audio-record the interview. Should the parent/caregiver choose to participate, he/she will 1) sign the informed consent via a paper informed consent form if the parent is participating in the interview in person, or 2) provide verbal consent if he/she is participating by video or by phone.

Provider Participants and Parent Advocates (Aim 3 – In-person or video meetings/interviews). Providers and parent advocates will view a disclosure statement that will detail the study content and potential risks and benefits of participation. Participants will be informed that their participation is voluntary, and parent participants that their child's health care will not be affected, whether or not they participate. The participant will also be reminded that they can stop their participation in the interview at any time or decline to answer any question. The interviews will also require consent to audio-record the interview. Should the participant choose to participate, he/she will 1) sign the informed consent via a paper informed consent form if the participant is participating in person, or 2) provide verbal consent if he/she is participating by video.

b. Protection Against Risk

Potential Risk	Protections/Safeguards
Aim 1: Parent Participants – Web-based questionnaire	

Breach of Confidentiality	<p>Web-based questionnaires will be completed on a secure, online survey software platform (REDCap). Each participant will receive a unique study alpha-numeric code identifier. The master list encoding study code identifiers to personal identifiers will be encrypted; only the PI will have access to this file.</p> <p>Only aggregate data, presented in group format, will be used in presentations and publications. All email correspondences with subjects (for the purposes of recruitment or other communication about the study) will be deleted at the conclusion of the study. All data files will be downloaded to a designated, password-protected, research drive of the secure server at CHOP for data management and analysis. The research drive will only be accessible to the research team.</p>
Emotional Distress	<p>The first page of the survey will inform participants that they could experience emotional distress during the survey, and that if they feel distressed, they should reach out to the PI. The PI's email address and phone number will be provided. The PI has clinical experience working with children with serious illnesses and their families in both the hospital and community settings, and is well-supported by a mentorship team that includes experienced clinicians and researchers who have worked extensively with seriously-ill individuals and their families. If any participants reach out with emotional distress, the PI will immediately seek assistance from the PACT team providers and Dr. Feudtner (Sponsor), who has many years of experience working with parents of seriously ill children.</p>
Aim 2: Parent Participants – Interviews	
Emotional Distress	<p>If the PI or trained RA detects that the participant is experiencing emotional distress, she/he will halt the interview and provide reassurance that the participant can stop the process at any time or refuse to answer specific questions. The PI/RA will also encourage the participant to contact their clinical team or other health care provider for support, or seek permission from the participant to contact their provider on their behalf. The PI has clinical experience working with persons with serious illnesses and their families in both the hospital and community settings, and is well-supported by a mentorship team that includes experienced clinicians and researchers who have worked extensively with seriously-ill individuals and their families. If any emotional distress is detected, the PI will immediately seek assistance from the PACT team providers and from Dr. Feudtner (Sponsor), who has many years of experience working with parents of seriously ill children.</p>
Breach of Confidentiality	<p>All interview data will be de-identified. No names or other identifying information will be recorded in the field notes. During data analysis, the PI will assign all interview respondents in the study a de-identified code. Only the PI will have access to the raw data generated from each interview, and she will immediately upload and store all audio recordings to a designated, password-protected, research drive of the secure server at CHOP for data</p>

	<p>management and analysis. She will then transmit the uploaded recordings securely to a local transcription service that meets all requirements of CHOP's IRB regarding confidentiality.</p> <p>Interview transcripts will be stored on a research drive of the secure server at CHOP, and the PI will review the transcribed data, remove participants' personal identifiers, and assign a pseudonym to each participant. If transcribed data include participants' or other family members' or providers' names or private information, the PI will change them to pseudonyms or remove the private information. The de-identified, redacted, transcribed data will be analyzed and stored on the same electronic research drive. Only de-identified data will be used in presentations and publications.</p> <p>The PI will shred the hand-written field notes once notes have been transcribed and verified for accuracy. She will upload field notes and interview transcripts to Atlas.ti qualitative data analysis software on the designated, password-protected, research drive of the secure server at CHOP. The PI will set Atlas.ti to prompt each authorized user for a password before being granted access to the data for analysis. All electronic and hard copy research records and data will be destroyed at the completion of the study, according to NIH and CHOP IRB policies.</p>
<p>Aim 3: PPHC Providers and Parent Advocates</p>	
<p>Emotional Distress</p>	<p>If the PI or trained RA detects that the participant is experiencing emotional distress, she/he will halt the exercise and provide reassurance that the participant can stop the process at any time or refuse to answer specific questions. If any emotional distress is detected from the PPHC providers from CHOP, the PI will immediately seek assistance from Dr. Feudtner (Sponsor), who has long-standing professional relationships with many of the PPHC providers at CHOP. If any emotional distress is detected from the community-based providers or parent advocates, the PI will immediately seek assistance from PACT team members.</p>
<p>Breach of Confidentiality</p>	<p>All interview data will be de-identified. No names or other identifying information will be recorded in the field notes. During data analysis, the PI will assign all interview respondents in the study a de-identified code. Only the PI will have access to the raw data generated from each interview, and she will immediately upload and store all audio recordings to a designated, password-protected, research drive of the secure server at CHOP for data management and analysis. She will then transmit the uploaded recordings securely to a local transcription service that meets all requirements of CHOP's IRB regarding confidentiality.</p> <p>Interview transcripts will be stored on a research drive of the secure server at CHOP, and the PI will review the transcribed data, remove participants' personal identifiers, and assign a pseudonym to each participant. If transcribed data include participants' or other family members' or providers' names or private information, the PI will change them to pseudonyms or remove the private information. The de-identified, redacted, transcribed data will be analyzed and stored on the same</p>

	<p>electronic research drive. Only de-identified data will be used in presentations and publications.</p> <p>The PI will shred the hand-written field notes once notes have been transcribed and verified for accuracy. She will upload field notes and interview transcripts to Atlas.ti qualitative data analysis software on the designated, password-protected, research drive of the secure server at CHOP. The PI will set Atlas.ti to prompt each authorized user for a password before being granted access to the data for analysis. All electronic and hard copy research records and data will be destroyed at the completion of the study, according to NIH and CHOP IRB policies.</p>
--	----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------

c. Vulnerable Subjects (Not applicable)

3. Potential Benefits of the Proposed Research to Research Participants and Others

The risks to participation in this study are minimal, as described above. Potential benefits for parent participants include the opportunity to share their experiences and perspectives in order to contribute to the goal of measuring, and eventually improving, the quality of PPHC@Home services for future children and families. The professionals who participate in this study could potentially benefit by contributing to the advancement of knowledge in understanding families' experiences with PPHC@Home services for the children and families they care for currently and in the future.

4. Importance of Knowledge to be Gained

The conduct of this study will produce knowledge that will ultimately lead to more effective and higher quality of care for children with serious illnesses and their families who receive palliative and hospice care in the home setting. The risks to participation in this study are minimal, as described above, and producing this knowledge is not possible without the involvement of the parents and providers of these children.

Groups that are traditionally excluded from clinical research “have suboptimal clinical care because of a weak evidence base.”(95) This study population of children with serious illness and their families have historically been excluded from research. As such, “we have little information about the epidemiology and characteristics of children and adolescents; the use, organization, and financing of services for them; the best practices; methods of assessing and improving care...and basic issues of long-term clinical management.”(96) This is especially urgent for children with serious illnesses at the end of life: The Institute of Medicine (2003) cautions that clinicians and parents must often make difficult decisions about the care of children with “little guidance from clinical or health services research that documents the potential burdens as well as the potential benefits of medical interventions.”(7) Following the Institute of Medicine’s recommendations, it is critically important to study this population of children with serious illnesses and their families to better

understand how best to care for these children and their families who spend the last phase of life at home.

BIBLIOGRAPHY

1. Fraser LK, Miller M, Aldridge J, McKinney PA, Parslow RC. Life-limiting and life-threatening conditions in children and young people in the United Kingdom; national and regional prevalence in relation to socioeconomic status and ethnicity. In: University of Leeds, 2011.
2. Friebert S, Williams C. NHPCO'S Facts and Figures: Pediatric Palliative and Hospice Care in America. In: Alexandria, VA: NHPCO, 2015.
3. Feudtner C, Feinstein JA, Satchell M, Zhao H, Kang TI. Shifting place of death among children with complex chronic conditions in the United States, 1989-2003. *JAMA* 2007;297:2725-2732. doi: 10.1001/jama.297.24.2725
4. Feudtner C, Kang TI, Hexem KR, et al. Pediatric palliative care patients: A prospective multicenter cohort study. *Pediatr* 2011;127:1094-1101. doi: 10.1542/peds.2010-3225
5. Himmelstein BP, Hilden JM, Boldt AM, Weissman D. Pediatric palliative care. *N Engl J Med* 2004;350:1752-1762. doi:
6. Nageswaran S, Radulovic A, Anania A. Transitions to and from the acute inpatient care setting for children with life-threatening illness. *Pediatr Clin North Am* 2014;61:761-783. doi: 10.1016/j.pcl.2014.04.008
7. Field MJ, Behrman RE. When Children Die: Improving Palliative and End-of-Life care for Children and their Families. Washington, DC: The National Academies Press, 2003:419-420. Available from: <https://www.nap.edu/catalog/10390/when-children-die-improving-palliative-and-end-of-life-care>. Accessed June 5, 2020.
8. Feudtner C, Christakis DA, Connell FA. Pediatric deaths attributable to complex chronic conditions: A population-based study of Washington State, 1980-1997. *Pediatr* 2000;106:205-209.
9. Feudtner C, Christakis DA, Zimmerman FJ, et al. Characteristics of deaths occurring in children's hospitals: Implications for supportive care services. *Pediatr* 2002;109:887-893. doi: 10.1542/peds.109.5.887
10. Feudtner C, Friebert S, Jewell J. Pediatric palliative care and hospice care commitments, guidelines, and recommendations. *Pediatr* 2013;132:966-972. doi: 10.1542/peds.2013-2731
11. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980-1998. *Pediatr* 2002;109:656-660. doi: 10.1542/peds.109.4.656
12. Burke RT, Alverson B. Impact of children with medically complex conditions. *Pediatr* 2010;126:789-790. doi: 10.1542/peds.2010-1885
13. Ramer-Chrastek J. Hospice care for a terminally-ill child in the school setting. *J Sch Nurs* 2000;16:52-56. doi: 10.1177/105984050001600210
14. Jamorabo DS, Belani CP, Martin EW. Complex chronic conditions in Rhode Island's pediatric populace: Implications for palliative and hospice services, 2000-2012. *J Palliat Med* 2015;18:350-357. doi: 10.1089/jpm.2014.022
15. Ryan JG, Haas K, Kramer N, et al. When technology fails: Community-based pediatric hospice. *J Pediatr Nurs* 2016;31:353-355. doi: 10.1016/j.pedn.2016.02.008
16. Rowse V. Palliative care for children: A public health initiative. *Paed Nurs* 2006;18:41-45. doi:
17. Jones BW. The need for increased access to pediatric hospice and palliative care. *Dimens Crit Care Nurs* 2011;30:231-235. doi: 10.1097/DCC.0b013e3182276ded

18. Lindley LC, Lyon ME. A profile of children with complex chronic conditions at end of life among Medicaid beneficiaries: Implications for health care reform. *J Palliat Med* 2013;16:1388-1393. doi: 10.1089/jpm.2013.0099
19. Kaye E, Rubenstein J, Levine D, et al. Pediatric palliative care in the community. *CA Cancer J Clin* 2015;65:315-333. doi: 10.3322/caac.21280
20. Carroll JM, Torkildson C, Winsness JS. Issues related to providing quality pediatric palliative care in the community. *Pediatr Clin N Amer* 2007;54:813-27, xiii. doi: 10.1016/j.pcl.2007.06.002
21. Feudtner C. Perspectives on quality at the end of life. *Arch Pediatr Adolesc Med* 2004;158:415-418. doi: 10.1001/archpedi.158.5.415
22. Hays RM, Valentine J, Haynes G, et al. The Seattle Pediatric Palliative Care Project: Effects on family satisfaction and health-related quality of life. *Journal of Palliative Medicine* 2006;9:716-28. doi: 10.1089/jpm.2006.9.716
23. Bona K, Bates J, Wolfe J. Massachusetts' Pediatric Palliative Care Network: Successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med* 2011;14:1217-1223. doi: 10.1089/jpm.2011.0070
24. Postier A, Chrastek J, Nugent S, Osenga K, Friedrichsdorf SJ. Exposure to home-based pediatric palliative and hospice care and its impact on hospital and emergency care charges at a single institution. *J Palliat Med* 2014;17:183-188. doi: 10.1089/jpm.2013.0287
25. Tan JS, Docherty SL, Barfield R, Brandon DH. Addressing parental bereavement support needs at the end of life for infants with complex chronic conditions. *J Palliat Med* 2012;15:579-584. doi: 10.1089/jpm.2011.0357
26. Davies B, Gudmundsdottir M, Worden B, et al. "Living in the dragon's shadow" fathers' experiences of a child's life-limiting illness. *Death Stud* 2004;28:111-135. doi: 10.1080/07481180490254501
27. Thienprayoon R, Marks E, Funes M, et al. Perceptions of the pediatric hospice experience among English- and Spanish-speaking families. *J Palliat Med* 2016;19:30-41. doi: 10.1089/jpm.2015.0137
28. Vollenbroich R, Borasio GD, Duroux A, et al. Listening to parents: The role of symptom perception in pediatric palliative home care. *Palliat Support Care* 2016;14:13-9. doi: <https://dx.doi.org/10.1017/S1478951515000462>
29. Zimmermann K, Cignacco E, Eskola K, et al. Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu): An instrument to assess parental experiences and needs during their child's end-of-life care. *J Adv Nurs* 2015;71:3006-3017. doi: 10.1111/jan.12741
30. 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;18:143-150. doi: 10.1089/jpm.2014.0285
31. Widger K, Tourangeau AE, Steele R, Streiner DL. Initial development and psychometric testing of an instrument to measure the quality of children's end-of-life care. *BMC Palliat Care* 2015;14:1. doi: 10.1186/1472-684X-14-1
32. Stone AA, Shiffman S, Atienza A, N'ebeling L. *The Science of Real-Time Data Capture: Self-Reports in Health Research*, New York, NY: Oxford University Press, 2007.
33. Conway PH, Mostashari F, Clancy C. The future of quality measurement for improvement and accountability. *JAMA* 2013;309:2215-2216. doi: 10.1001/jama.2013.4929

34. Boyce MB, Browne JP. Does providing feedback on patient-reported outcomes to healthcare professionals result in better outcomes for patients? A systematic review. *Quality of Life Research* 2013;22:2265-78. doi: 10.1007/s11136-013-0390-0
35. Van Der Wees PJ, Nijhuis-Van Der Sanden MWG, Ayanian JZ, et al. Integrating the use of patient-reported outcomes for both clinical practice and performance measurement: Views of experts from 3 countries. *Milbank Q* 2014;92:754-775. doi: 10.1111/1468-0009.12091
36. Bevans KB, Moon J, Carle AC, et al. Patient reported outcomes as indicators of pediatric health care quality. *Acad Pediatr* 2014;14:S90-S96. doi: 10.1016/j.acap.2014.06.002
37. Kamal AH, Hanson LC, Casarett DJ, et al. The quality imperative for palliative care. *J Pain Symptom Manage* 2015;49:243-253. doi: 10.1016/j.jpainsymman.2014.06.008
38. National Institute of Nursing Research. The NINR's Strategic Plan: Advancing Science, Improving Lives. 2016 September. Available from: https://www.ninr.nih.gov/sites/files/docs/NINR_StratPlan2016_reduced.pdf. Accessed June 26, 2020.
39. PAR-19-320: Palliative Care in Home and Community Settings (R21 Clinical Trial Optional). National Institutes of Health. Posted July 31, 2019. Available from: <https://grants.nih.gov/grants/guide/pa-files/par-19-320.html#:~:text=Home%20and%20community%20in%20this,of%20life%20and%20palliative%20care>. Accessed June 27, 2020.
40. National Hospice and Palliative Care Organization (NHPCO). Standards of practice for pediatric palliative care. Alexandria, VA: NHPCO, 2019. Available from: https://www.nhpco.org/wp-content/uploads/2019/07/Pediatric_Standards.pdf. Accessed June 12, 2020.
41. Boyden JY, Curley MAQ, Deatrick JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage* 2018;55:117-131. doi: 10.1016/j.jpainsymman.2017.04.017
42. Lindley LC, Kieim-Malpass J. Quality of paediatric hospice care for children with and without multiple complex chronic conditions. *Int J Palliat Nurs* 2017;23:230-237. doi: 10.12968/ijpn.2017.23.5.230
43. Contro NA, Larson J, Scofield S, Sourkes B, Cohen H. Hospital staff and family perspectives regarding quality of pediatric palliative care. *Pediatr* 2004;114:1248-1252. doi: 10.1542/peds.2003-0857-L
44. Surkan PJ, Kreicbergs U, Valdimarsdottir U, et al. Perceptions of inadequate health care and feelings of guilt in parents after the death of a child to a malignancy: A population-based long-term follow-up. *J Palliat Med* 2006;9:317-331. doi: 10.1089/jpm.2006.9.317
45. Hays RM, Valentine J, Haynes G, et al. The Seattle Pediatric Palliative Care Project: Effects on family satisfaction and health-related quality of life. *J Palliat Med* 2006;9:716-28. doi: 10.1089/jpm.2006.9.716
46. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based pediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. *BMC Palliat Care* 2016;15:73. doi: 10.1186/s12904-016-0138-z
47. Roza KA, Lee EJ, Meier DE, Goldstein NE. A survey of bereaved family members to assess quality of care on a palliative care unit. *J Palliat Med* 2015;18:358-365. doi: 10.1089/jpm.2014.0172
48. Anhang Price R, Stucky B, Parast L, et al. Development of valid and reliable measures of patient and family experiences of hospice care for public reporting. *J Palliat Med* 2018;21:924-932. doi: 10.1089/jpm.2017.0594

49. Vollenbroich R, Duroux A, Grasser M, et al. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med* 2012;15:294-300. doi: 10.1089/jpm.2011.0196
50. Basch E, Torda P, Adams K. Standards for patient-reported performance measures. *JAMA* 2013;310:139-140. doi: 10.1001/jama.2013.6855
51. Boyce MB, Browne JP. Does providing feedback on patient-reported outcomes to healthcare professionals result in better outcomes for patients? A systematic review. *Qual Life Res* 2013;22:2265-78. doi: 10.1007/s11136-013-0390-0
52. Hill DL, Walter JK, Casas JA, et al. The codesign of an interdisciplinary team-based intervention regarding initiating palliative care in pediatric oncology. *Support Care Cancer* 2018;26:3249-3256. doi: 10.1007/s00520-018-4190-5
53. Institute for Healthcare Improvement (IHI). Experience-based co-design of health care services. 2019. Available from: <http://www.ihl.org/resources/Pages/Publications/Experience-Based-Co-Design-Health-Care-Services-Innovation-Case-Study.aspx>. Accessed Nov 29, 2019.
54. Bate P, Robert G. Experience-based design: From redesigning the system around the patient to co-designing services with the patient. *Qual Saf Health Care* 2006;15:307-10. doi: 10.1136/qshc.2005.016527
55. National Consensus Project for Quality Palliative Care. Clinical Practice Guidelines for Quality Palliative Care. 4th ed. Richmond, VA: National Coalition for Hospice and Palliative Care, 2018. Available from: <https://www.nationalcoalitionhpc.org/ncp/>. Accessed June 5, 2020.
56. Widger KA, Wilkins K. What are the key components of quality perinatal and pediatric end-of-life care? A literature review. *J Palliat Care* 2004;20:105-112. doi: <https://doi.org/10.1177/082585970402000207>
57. Thienprayoon R, Mark MSJ, Grosseohme D. Provider-prioritized domains of quality in pediatric home-based hospice and palliative care: A study of the Ohio Pediatric Palliative Care and End-of-Life Network. *J Palliat Med* 2018;31:290-296. doi: 10.1089/jpm.2017.0333
58. Sawtooth Software. Bandit MaxDiff: When to use it and why it can be a better choice than standard MaxDiff. In: Orem, UT: 2018. Available from: <https://www.sawtoothsoftware.com/support/technical-papers/175-support/technical-papers/maxdiff-best-worst-scaling/1943-bandit-maxdiff-when-to-use-it-and-why-it-can-be-a-better-choice-than-standard-maxdiff>. Accessed June 27, 2020.
59. Streiner DL, Norman GR, Cairney J. *Health Measurement Scales : A Practical Guide to Their Development and Use*, Oxford: OUP Oxford, 2015.
60. Varni J, Seid M, Kurtin P. PedsQL 4.0: Reliability and validity of the Pediatric Quality of Life Inventory Version 4.0 Generic Core Scales in healthy and patient populations. *Med Care* 2001;39:800-812. doi: 10.1097/00005650-200108000-00006
61. McMillan S, Mahon M. Measuring quality of life in hospice patients using a newly developed quality of life index. *Qual Life Res* 1994;3:437-447. doi: 10.1007/BF00435396
62. Floyd F, Widaman K. Factor analysis in the development and refinement of clinical assessment instruments. *Psychol Assess* 1995;7:286-299. doi: <https://doi.org/10.1037/1040-3590.7.3.286>
63. Pearson R, Mundfrom D. Recommended sample size for conducting exploratory factor analysis on dichotomous data. *J Mod Appl Stat Methods* 2010;9:359-368. doi: 10.22237/jmasm/1288584240
64. Bredart A, Robertson C, Razavi D, et al. Patients' satisfaction ratings and their desire for care improvement across oncology settings from France, Italy, Poland and Sweden. *Psychooncol* 2003;12:68-77. doi: 10.1002/pon.620

65. Kessler RC, Andrews G, Colpe LJ, et al. Short screening scales to monitor population prevalences and trends in non-specific psychological distress. *Psychol Med* 2002;32:959-976. doi: 10.1017/s0033291702006074
66. Rosenberg AR, Wolfe J, Bradford MC, et al. Resilience and psychosocial outcomes in parents of children with cancer. *Pediatr Blood Cancer* 2014;61:552-7. doi: 10.1002/psc.24854
67. Knafk K, Deatrck JA, Gallo A, et al. Assessment of the psychometric properties of the Family Management Measure. *J Pediatr Psychol* 2011;36:494-505. doi: 10.1093/jpepsy/jsp034
68. Hobbie WL, Ogle SK, Reilly M, et al. Identifying the educational needs of parents at the completion of their child's cancer therapy. *J Ped Oncol Nurs* 2010;27:190-195. doi: 10.1177/1043454209360778
69. Deatrck JA, Mullaney EK, Mooney-Doyle K. Exploring family management of childhood brain tumor survivors. *J Pediatr Oncol Nurs* 2009;26:303-11. doi: 10.1177/1043454209343210
70. Boyden JY, Hill DL, Carroll KW, et al. The association of perceived social support with anxiety over time in parents of children with serious illnesses. *J Palliat Med* 2020;23:529-534. doi: 10.1089/jpm.2019.0387
71. Cook DA, Beckman TJ. Current concepts in validity and reliability for psychometric instruments: Theory and application. *Am J Med* 2006;119:166 e7-16. doi: 10.1016/j.amjmed.2005.10.036
72. Messick S. Validity of psychological assessment: Validation of inferences from persons' responses and performances as scientific inquiry into score meaning. *Amer Psychol* 1995;50:741-744.
73. Fernstrom KM, Shippee ND, Jones AL, Britt HR. Development and validation of a new patient experience tool in patients with serious illness. *BMC Palliat Care* 2016;15:99. doi: 10.1186/s12904-016-0172-x
74. Hays RD, Berman LJ, Kanter MH, et al. Evaluating the psychometric properties of the CAHPS Patient-centered Medical Home survey. *Clin Ther* 2014;36:689-696 e1. doi: 10.1016/j.clinthera.2014.04.004
75. Streiner DL. Starting at the beginning: An introduction to coefficient alpha and internal consistency. *J Pers Assess* 2003;80:99-103. doi: 10.1207/S15327752JPA8001_18
76. Proctor E, Silmere H, Raghavan R, et al. Outcomes for implementation research: Conceptual distinctions, measurement challenges, and research agenda. *Adm Policy Ment Health* 2011;38:65-76. doi: 10.1007/s10488-010-0319-7
77. 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;4:50-65. doi: 10.1186/1748-5908-4-50
78. Polit DF, Beck CT. *Nursing Research: Generating and Assessing Evidence for Nursing Practice*, 9th ed. Philadelphia, PA: Lippincott Williams & Wilkins, 2012.
79. Gans D, Hadler MW, Chen X, et al. Impact of a pediatric palliative care program on the caregiver experience. *J Hosp Palliat Nurs* 2015;17:559-565. doi: 10.1097/NJH.0000000000000203
80. Steele RG. Experiences of families in which a child has a prolonged terminal illness: Modifying factors. *Int J Palliat Nurs* 2002;8:418-434. doi: 10.12968/ijpn.2002.8.9.10687
81. Haley JM, Walker GA. Discovering the strength of parents whose children are at end of life. *J Hosp Palliat Nurs* 2016;18:139-148. doi: 10.1097/NJH.0000000000000225
82. Linton JM, Feudtner C. What accounts for differences or disparities in pediatric palliative and end-of-life care? A systematic review focusing on possible multilevel mechanisms. *Pediatr* 2008;122:574-582. doi: 10.1542/peds.2007-3042

83. Widger K, Brennenstuhl S, Duc J, Tourangeau A, Rapoport A. Factor structure of the Quality of Children's Palliative Care Instrument (QCPCI) when completed by parents of children with cancer. *BMC Palliat Care* 2019;18:23. doi: 10.1186/s12904-019-0406-9
84. Gustavsson S. Improvements in neonatal care: Using experience-based co-design. *Int J Health Care Qual Assur* 2014;27:427-438. doi: 10.1108/IJHCQA-02-2013-0016
85. Gustavsson S, Gremyr I, Kenne Sarenmalm E. Designing quality of care – Contributions from parents. *J Clin Nurs* 2016;25:742-751. doi: 10.1111/jocn.13050
86. Tsianakas V, Robert G, Maben J, et al. Implementing patient-centred cancer care: Using experience-based co-design to improve patient experience in breast and lung cancer services. *J Support Care Cancer* 2012;20:2639-2647. doi: 10.1007/s00520-012-1470-3
87. Bond S. Response to "Using satisfaction to measure the quality of palliative care: a review of the literature" *J Adv Nurs* 2003;43:533-534. doi: 10.1046/j.1365-2648.2003.02750_3.x
88. Stiel S, Pastrana T, Balzer C, et al. Outcome assessment instruments in palliative and hospice care: A review of the literature. *Support Care Cancer* 2012;20:2879-2893. doi: 10.1007/s00520-012-1415-x
89. Hinds PS, Menard JC, Jacobs SS. The child's voice in pediatric palliative and end-of-life care. *Prog Palliat Care* 2012;20:337-342. doi: <https://doi.org/10.1179/1743291X12Y.0000000035>
90. Feudtner C, Feinstein JA, Zhong W, Hall M, Dai D. Pediatric complex chronic conditions classification system version 2: Updated for ICD-10 and complex medical technology dependence and transplantation. *BMC Pediatr* 2014;14:199. doi: 10.1186/1471-2431-14-199
91. Mooney-Doyle K, Deatrick JA. Parenting in the face of childhood life-threatening conditions: The ordinary in the context of the extraordinary. *Palliat Support Care* 2016;14:187-198. doi: 10.1017/S1478951515000905
92. Mongeau S, Champagne M, Liben S. Participatory research in pediatric palliative care: benefits and challenges. *J Palliat Care* 2007;23:5-13.
93. Hinds PS, Burghen EA, Pritchard M. Conducting end-of-life studies in pediatric oncology. *West J Nurs Res* 2007;29:448-465. doi: 10.1177/0193945906295533
94. Children's Hospital of Philadelphia. Exempt Research 2019. Available from: <https://irb.research.chop.edu/exempt-research>. Accessed November 22, 2019
95. Hulley SB, Cummings SR, Browner WS, Grady DG, Newman TB. *Designing Clinical Research*, 3rd ed. Philadelphia, PA: Lippincott Williams and Wilkins, 2006.
96. Perrin JM. Health services research for children with disabilities. *Milbank Q* 2002;80:303-324. doi: 10.1111/1468-0009.t01-1-00005

TABLES AND FIGURES

Table 1: Example EXPERIENCE@Home Measure Items and Weights

Table 2: Additional Study Measures

Figure 1: Domains of Quality PPHC@Home

Table 1: Example EXPERIENCE@Home Measure Items and Weights

Item	Weighted Score	Domain
The care team treats my child's physical symptoms so that my child has as good a quality of life as possible.	61.08	Physical aspects of care: Symptom management
The care team gives me enough information to make informed health care decisions about my child's care at home.	23.84	Communication between family and care team
The care team helps adapt our home to better support my child's current care needs.	13.38	Care coordination

Table 2: Additional Study Measures

Measure	# Items (Estimated Completion Time); Scoring	Hypothesized Relationship to EXPERIENCE@Home	Evidence for Relationship Based on Supporting Studies
Overall satisfaction with care (“Overall, how satisfied are you with your child’s home-based care over the last week?”)	1 item (<1 minute); 10-point scale (1 = Completely Dissatisfied; 10 = Completely Satisfied)	Better care experience will correlate with higher satisfaction with care	Used in 2 instrument development studies in PPC (29, 31)
Desire for Care Improvement (64) (“I would like to see improvement in my child’s home-based palliative or hospice care”)	1 item (<1 minute); 10-point scale (1 = Strongly Disagree; 10 = Strongly Agree)	Better care experience will correlate with lower desire to improve care	Used in multi-national study to measure overall satisfaction with care and desire for improvement (64)
Kessler-6 Psychological Distress Scale (65)	6 items (1-2 minutes); Reported as mean and total score (range: 0-24)	Better care experience will correlate with lower parental distress	Used in study of distress and resilience in parents of children with cancer (66)
One item from the Family Management Measure – Family Life Difficulty Subscale (67) (“Taking care of our child’s condition is often overwhelming.”)	1 item (< 1 minute); 10-point scale (1= Strongly Disagree; 10= Strongly Agree)	Better care experience will correlate with lower perceived difficulty of family life	Used in studies of parents of children with cancer (68, 69)
Child’s Health Status (“How would you rate your child’s health today?”)	1 item (<1 minute); 10-point scale (1=Worst Health; 10=Best Health)	N/A – will be assessed at baseline and re-test to account for changes in child’s condition	Used in analysis of parental anxiety and social support in pediatric serious illness (70)

Figure 1: Domains of Quality PPHC@Home

Physical	Psychological & Emotional	Structure & Processes	Communication & Relationships
<ul style="list-style-type: none">• Symptom management• Information about treatments of symptoms	<ul style="list-style-type: none">• Psychological & emotional care for the child• Psychological & emotional care for the parent• Caregiver support at child's end of life	<ul style="list-style-type: none">• Knowledge/skills of care provider• Access to care team• Coordination of care• Continuity of care	<ul style="list-style-type: none">• Relationship between family and care team• Communication between family and care team• Communication at end of life

APPENDIX

The appendix contains the following 4 documents pertinent to the proposed research project (as described in the *Research Strategy*):

- A. The EXPERIENCE@Home Measure (Aim 1)
- B. The interview guide for parent participants (Aim 2)
- C. The weekly meeting guide for pediatric palliative care providers (Aim 3)
- D. The co-design session guide for the pediatric palliative and hospice care stakeholder group (Aim 3)

Appendix A: Aim 1: The Final EXPERIENCE@Home Measure

Please choose the answer that best fits your experience with your child's home-based palliative or hospice care team over the past week.

Your answers will help the care team to better understand if your child's and family's most important needs are being met, and to guide the care team to better address those needs.

Note: For all questions, the care team is your child's home-based palliative care or hospice team (that is, the nurses, physicians, social workers, chaplains, child life therapists, art therapists who care for your child and family in your home).

Use this scale for all questions:		Strongly Disagree	Disagree	Neutral	Agree	Strongly Agree	Does not apply
1	The care team treats my child's physical symptoms so that my child has as good a quality of life as possible.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2	The care team helps me to feel confident in managing my child's symptoms at home without needing to go to the hospital.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3	My care team provides the care or guidance we need to keep my child at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4	I trust the care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5	The care team works together with me and my child to make decisions about my child's medical care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6	The care team asks for my opinions and concerns about my child.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7	The palliative care or hospice team works closely with my child's other providers (for ex, neurology, cardiology, oncology, primary care) to support our family's goals for my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8	The care team gives me enough information to make informed health care decisions about my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9	I have regular access to on-call services from our care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10	I have access to a provider who can help me coordinate my child's care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11	The care team considers all of my child's needs.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12	The nurses have the knowledge, skills, and experience to support my child's palliative or hospice care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13	The doctors and nurse practitioners have the knowledge, skills, and experience to support my child's palliative or hospice care at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
14	The care team provides the information and strategies to effectively manage my child's pain and other physical symptoms at home.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15	The care team helps adapt our home to	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

	better support my child's current care needs.						
16	The care team provides opportunities for my child to express his/her feelings, such as fears, worries, and hopes.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
17	The care team provides support for <u>my other children's</u> feelings and emotions.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
18	The care team provides support for <u>my</u> feelings and emotions, including fear, worry, sadness, and hope.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
19	The care team helps me hope for the best possible outcome for my child's health, while also helping me prepare if that outcome doesn't happen.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
20	The care team has provided or directed me to resources that support my needs as my child's caregiver.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
21	I can talk about my child's end of life with the care team.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
22	The care team has talked with me about my child's last weeks of life and what they may be like.	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

APPENDIX B: Aim 2: Interview Guide for Parent Participants

General impressions about the EXPERIENCE@Home measure:

1. I would like to ask about your overall experience with completing the EXPERIENCE@Home Measure.
 - a. Can you tell me briefly what you think is the purpose of this tool?
 - b. Overall, what do you think about the tool?
 - c. Overall, are the questions important? Why or why not?
 - d. Overall, are the questions useful? Why or why not?
 - e. Overall, how well do you feel the questions apply to your family's experience? Why?

Characteristics of the Measure:

2. Barriers and Facilitators
 - a. Can you tell me about what made completing the EXPERIENCE@Home Measure easy?
 - b. Can you tell me about what made completing the EXPERIENCE@Home Measure difficult?
 - i. Please describe any things that got in the way of completing the EXPERIENCE@Home Measure.
 - c. *For parents who did not complete the study:* Could I ask what made you decide to stop participating in the study?
 - i. Is there anything we could have done differently?
3. Relative Advantage
 - a. Do you feel that the EXPERIENCE@Home Measure could help you better communicate your family's experiences and needs at home with your care team? Why or why not?
 - b. Do you feel that the EXPERIENCE@Home Measure could improve the care you receive in your home from your care team?
 - c. How many of the items that you disagreed or strongly disagreed with on the EXPERIENCE@Home Measure would you expect your care team to fix or address?
 - d. *(If Applicable)* How does the EXPERIENCE@Home Measure compare to other surveys or tools that you have completed to better understand your family's needs or experience at home?
4. Adaptability
 - a. What kinds of changes or alterations would need to be made to the EXPERIENCE@Home Measure so it will be more useful for families?
 - b. If we were to ask you to complete the EXPERIENCE@Home Measure regularly, how often do you think it would be useful for you to complete it?
 - i. What could we change about the EXPERIENCE@Home Measure to make you more willing to complete it regularly?

Caregiver Characteristics:

5. Personally, what might make you hesitant to use the EXPERIENCE@Home Measure?
6. Personally, what might make it easier for you to use the EXPERIENCE@Home Measure?

Conclusion:

7. Do you have any additional comments that could help us better understand why you liked (or did not like) the EXPERIENCE@Home Measure?

APPENDIX C: Aim 3: Weekly Meeting Guide with PPC Providers

Parent Study ID Number:	
Meeting Date and Time:	
Attendees:	

Evidence Strength & Quality

1. What information from this report did you already know about the family?
2. What information from this report is new (that you didn't know about the family)?
3. What information do you feel is clinically actionable over the short-term for this family?
 - a. In other words, what do you feel you can improve, in the next days to weeks, through clinical interventions or changes to the child and family's plan of care?
 - b. How confident are you that these interventions or changes can be done successfully?
4. What information do you feel is clinically actionable over the long-term for this family?
 - a. In other words, what do you feel you can improve, in the next weeks to months, through clinical interventions or changes to the child and family's plan of care?
 - b. How confident are you that these interventions or changes can be done successfully?
5. What information do you feel is not clinically actionable for this family?
 - a. In other words, what do you feel will not be changed, no matter how many changes you make to the child and family's plan of care?

Adaptability

6. If you could change one thing about the EXPERIENCE@Home Measure to make it more clinically useful, what would that one thing be?

Process

7. Given these results, what would your immediate next clinical action be? Please describe one action.

Conclusion

8. Do you have any additional comments that could help us better understand why you think the EXPERIENCE@Home Measure is clinically useful or not?

APPENDIX D: Aim 3: Quarterly Co-Design Sessions with PPHC Stakeholder Group

Co-Design Session Number:	
Meeting Date and Time:	
Attendees:	

Evidence Strength & Quality

1. What is your impression of the purpose of this measure?
2. What information do you feel is clinically actionable over the short-term for this family?
 - a. In other words, what do you feel can be improved, in the next days to weeks, through clinical interventions or changes to the child and family's plan of care?
 - b. How confident are you that these interventions or changes can be done successfully?
3. What information do you feel is clinically actionable over the long-term for this family?
 - a. In other words, what do you feel can be improved, in the next weeks to months, through clinical interventions or changes to the child and family's plan of care?
 - b. How confident are you that these interventions or changes can be done successfully?
4. What information do you feel is not clinically actionable for this family?
 - a. In other words, what do you feel will not be changed, no matter how many changes are made to the child and family's plan of care?
5. What information might be important to for us to evaluate over time (that is, if we were to administer this measure regularly, like every week or every month, with families)?
 - a. Please select 3 items that would be important to understand over time.

Barriers and Facilitators

6. What do you see as potential barriers to providers successfully acting on EXPERIENCE@Home Measure results?
 - a. Family-level (for example, things that the family can control in the home)
 - b. Provider-level (for example, things the providers – whether the PPC team or home-based team – can impact in the home)
 - c. Organizational-level (for example, things that the organization can control or impact)

- d. System-level (for example, regulations, laws, federal or state-level funding)
- 7. What do you see as potential facilitators to providers successfully acting on EXPERIENCE@Home Measure results?
 - a. Family-level (for example, things that the family can control in the home)
 - b. Provider-level (for example, things the providers – whether the PPC team or home-based team – can impact in the home)
 - c. Organizational-level (for example, things that the organization can control or impact)
 - d. System-level (for example, regulations, laws, federal or state-level funding)
- 8. What structures and processes at CHOP and in the community already exist to support successfully acting on EXPERIENCE@Home Measure results?
- 9. What new structures and processes at CHOP and in the community are needed to support successfully acting on EXPERIENCE@Home Measure results?

Relative Advantage

- 10. How does the EXPERIENCE@Home Measure compare to existing measures or tools you know of (if any)?
- 11. How does the priority of evaluating a family's experience with care at home compare to other priorities within the team/organization?

Adaptability

- 12. If you could change one thing about the EXPERIENCE@Home Measure to make it more clinically useful for CHOP providers and for families, what would that one thing be?

Conclusion

- 13. Thinking “outside the box” for a minute, what else might we consider as we implement the EXPERIENCE@Home Measure? Please be broad in your thoughts about this.
- 14. Do you have any additional comments that could help us better understand why you think the EXPERIENCE@Home Measure is clinically useful or not?

CHAPTER 5: SUMMARY AND FUTURE DIRECTIONS

SUMMARY OF FINDINGS

Children with life-shortening, serious illnesses are living longer with significant, medically complex care needs (1-5) and are increasingly being cared for primarily at home by their families.(2-4, 6) Pediatric palliative and hospice care in the home (PPHC@Home) supports these children and families through coordinated medical and social services primarily in the home, integrated with care in the community, outpatient, and hospital settings.(3, 4, 7)

While PPHC@Home has been associated with many benefits, such as improved child and family quality of life (8-11), death in the preferred location (8, 12, 13), better pain and symptom management (9, 14, 15), and family satisfaction with care (9, 10, 12, 16), children and families are not receiving consistently high-quality PPHC@Home. The provision of PPHC@Home in the U.S. is impacted by multilevel factors on the child and family, health system, and state and federal levels.(17) Consequently, the systematic measurement of child and family outcomes at home is necessary to ensure that care teams are consistently meeting children's and families' most important needs and priorities throughout the illness experience.

In order to accurately and effectively measure outcomes in PPHC@Home, instruments should possess 4 minimum characteristics: importance/relevance, usability/actionability, feasibility, and scientific acceptability.(18-21) Outcomes measurement in PPHC@Home, however, is challenged by several issues. For example, we know what domains of care are important for PPHC@Home, including excellent pain and symptom management (22-25), child- and family-centered care and decision-making (9, 22, 24, 26), inclusion of

siblings in care (22, 23), consistent, high quality communication between family and providers (9, 22, 23, 25, 26), family education and preparation for the end of life (25), good psychosocial and spiritual care (24, 26), and care coordination and management.(9, 24, 26) We do not, however, know which of these domains are *most* important for families, and thus, which are most important to measure and to direct limited health care resources. Additionally, existing instruments are often lengthy (27-29) and have not been validated for use in PPHC@Home.(28, 29) Many family-reported palliative care measures also survey respondents retrospectively.(8-10, 12, 16, 27-29) All of these issues may potentially limit the ability of the instrument to produce clinically-actionable data that could help providers improve care for children and families at the time care is provided. An instrument was needed to measure the most important PPHC@Home domains, that produces real-time, actionable information for clinicians, that can be administered routinely and without undue burden on families and providers, and that produces accurate measurements in a PPHC@Home population.

In response to this need, my dissertation research focused on developing a family-reported instrument to measure experiences with PPHC@Home provided in the U.S. using a multi-method, multi-stakeholder approach. In **Aim 1** of the project (Paper 1), I explored how parents rated and prioritized PPHC@Home domains using a discrete choice experiment with traditional MaxDiff scaling with parents whose children have received PPHC@Home services. By asking parents to provide their ratings of the most and least important domains, across sets containing different combinations of domains, I was able to calculate a rank-ordered list of parent-prioritized domains of PPHC@Home. Highest-rated domains included *Physical Aspects of Care: Symptom Management*,

Psychological/Emotional Support for the Child, and Care Coordination. Lowest-rated domains included *Spiritual and Religious Aspects of Care* and *Cultural Aspects of Care*. I found few significant differences between sub-groups of parents in my exploratory analysis, although I did find that parents who had other children rated the *Psychological/Emotional Aspects of Care for the Sibling(s)* domain significantly higher than parents who did not have other children. Additionally, bereaved parents rated the *Caregiver Support at the End of Life* domain significantly higher than parents who were currently caring for their child.

In **Aims 2 and 3** (Paper 2), I followed a 4-phase instrument development process. I first built the instrument's foundation on 20 domains of high-quality PPHC@Home, which were based on broad frameworks of high-quality palliative and hospice care and the peer-reviewed literature, as well as on feedback from a PPHC stakeholder panel. In the item identification and development phase, I first conducted a comprehensive review of the literature for items within existing measures, practice standards and guidelines, and studies that evaluated PPHC@Home outcomes. After reviewing this list of 100 items with an expert panel of PPHC stakeholders, including providers and parent advocates, I ended up with 75 items for evaluation in the next phase. In the item prioritization and reduction phases, I first asked a sample of 34 interdisciplinary PPHC providers and parent advocates to prioritize the 75 items using a discrete choice experiment with Bandit MaxDiff scaling. Top-ranked items included *I trust the care team*, *Access to on-call services from palliative care or hospice team*, and *Care team helps me do the best for my child*. Lowest-ranked items included *Care team helps prepare my child for school*, *Care team keeps me informed about their arrival time*, and *Care team helps with*

arranging transportation. I retained the top-ranked items from each domain for a total of 42 items for further evaluation by parents.

I then asked a sample of 47 parents whose children have received PPHC@Home to complete a similar discrete choice experiment with Bandit MaxDiff scaling with the 42 remaining items. Leveraging both domain and item importance scores, we calculated weighted item scores and retained the top 20 items across domains. Top-ranked items by weighted score included *Care team treats my child's physical symptoms so that my child has as good a quality of life as possible, I feel prepared to treat my child's symptoms at home*, and *My child can easily get necessary care*.

Finally, in the cognitive interviewing phase, I recruited 11 parents of children who have received PPHC@Home to participate in cognitive interviews. During these interviews, I asked parents to describe their interpretation of each item, to discuss any problems with wording and clarity, and to help us identify missing items. I retained or revised the 20 items per parents' recommendations. Based on parents' recommendations, I also added 2 items: *The care team provides support for my other children's feelings and emotions* and *The care team has provided or directed me to resources that support my needs as my child's caregiver*. The current 22-item EXPERIENCE@Home (**Ex**periences of **P**alliative and Hospice Care for Children and **C**aregivers at Home) Measure will undergo further evaluation in my post-doctoral fellowship project.

In **Aim 4** (Paper 3) – an F32 grant application to the NINR – I proposed next steps to evaluate the validity, reliability, acceptability, and feasibility of the EXPERIENCE@Home Measure, administered in real-time, from the perspective of parents whose children are

currently receiving PPHC@Home using survey and interview methods. I also proposed to evaluate the instrument's clinical utility from the perspectives of PPHC providers through weekly interdisciplinary team meetings and quarterly co-design sessions.

DISCUSSION

Through a phased multi-method, multi-stakeholder project, I developed a comprehensive yet concise instrument that measures aspects of PPHC@Home that are the most important to parents of children with serious illnesses and other PPHC stakeholders. My goal is that this instrument, which I have named the EXPERIENCE@Home Measure, can ultimately guide clinical care and care improvement initiatives.

Several general observations resulting from this dissertation study merit further discussion. First, we knew from the literature that PPHC@Home is associated with many positive outcomes (8-12, 16); we knew less about what aspects of this care are most valuable from the families' perspective. In my study, parents prioritized the domains of PPHC@Home, and rated the domains *Physical Aspects of Care: Symptom Management*, *Psychological/Emotional Support for the Child*, and *Care Coordination* as particularly important for the care of their children, as described in Paper 1. In Paper 2, parents prioritized items related to the physical care of their child (e.g., *Care team treats my child's physical symptoms so that my child has as good a quality of life as possible; I feel prepared to treat my child's symptoms at home*) and access to PPHC@Home (e.g., *My child can easily get necessary care*). In the cognitive interviews, parents spoke of the importance of the PPHC@Home team for helping their child enjoy a good quality of life and to remain at home through the receipt of expert symptom management, information,

and resources. Parents also described the value of the emotional support they and their children, including the ill child and their other children, received from their providers, particularly at the end of life.

We also know, however, that families face many significant challenges in caring for their children at home. As one parent reflected on during the cognitive interviews: "And that's such a big thing when you're trying to manage from home, right? Just think about you're not equipped with the gazillion computers, the fancy medical equipment, and you're trying to manage it from home..." From my integrative review, I learned that children and families are challenged with unequal access to high-quality PPHC@Home because of the variability of program size, funding, staffing, and provider knowledge and experience.(17) Previous studies have also found that insufficient access to high-quality PPHC@Home may lead to potentially avoidable hospitalizations (15, 30), inadequate pain and symptom management (15, 23), and parental stress and distress.(31) In Aim 3 of my study (Paper 2), parents participating in the cognitive interviews often spoke of challenges with physical symptoms that resulted in hospital admissions; problems with accessing appropriately-trained and experienced hospice, palliative, and home care nurses who serve children; feeling unprepared to manage acute crises in the home; and the chronic stress, anxiety, and exhaustion associated with caring for their child at home. These barriers to accessing palliative care and hospice providers who serve children has been previously described in the literature.(32, 33) Many hospice nurses who primarily care for adults, for example, lack adequate training, experience, and comfort with caring for seriously-ill children and their families.(34) Caring for seriously-ill children requires unique skills, including the need for developmentally-appropriate pain and symptom

assessment skills; the unique dynamics of young families and siblings, which presents unique challenges to communication and decision support; and the incredible emotional burden carried by parents, siblings, and providers.(35-39) More in-depth evaluation and, ultimately, future intervention research is critically needed to better understand how to address these critical gaps in care for children and families.

Moreover, through the Courageous Parents Network, I recruited participants from several states, including Pennsylvania, Massachusetts, Texas, and Vermont. Although my overall sample was too small to examine differences based on geographic location, from the cognitive interviews, it appears that parents living in different regions have starkly different experiences. For example, I interviewed several parents from Massachusetts, which has a robust state-funded PPHC@Home program.(12) These parents often spoke positively about the regular home visits they received from an interdisciplinary team of nurses, social workers, and therapists (including massage, child life, occupational, art). Many spoke of the regular access they had to their specialists at the hospital, as well as the advantages of having a hospice program integrated within the same organization as their palliative care program. In contrast, parents from other geographic areas often described their struggles with finding adequate home nursing support, accessing pediatric-trained hospice providers, and uncertainty about who to contact when they had questions or concerns about their child's care. Future research is greatly needed with a larger number of geographically-diverse sample of parents to more fully understand these potential differences in care experiences.

Ultimately, PPHC@Home plays a critical role in the lives of many children with serious illnesses and their families, and yet, we remain significantly challenged in our

understanding of how best to provide this care to families. This project represents an important step toward advancing our knowledge of what matters most to families, and toward ensuring that we are providing care that is adequately meeting families' most important needs and priorities.

STRENGTHS AND LIMITATIONS

The strengths of this project included a rigorous multi-method approach, leveraging the strengths of innovative methods not traditionally used in PPC research. First, I used a DCE with MaxDiff scaling. The recent movement in patient-centered health care and decision-making has increased interest in choice experiments for quantifying attributes of particular products or services that are most important and salient to patients and families, given trade-offs across different products or services.(40) My project, however, is the first known use of the DCE with MaxDiff scaling technique for instrument development in health care. MaxDiff provides several specific advantages for this study over other quantitative methods for obtaining patient and family preferences (e.g., Likert scale ratings, ranking or ordering lists of items) because these other methods: 1) allow participants to give many items a similar rating, making it difficult to discriminate between items (e.g. every item is equally important to include); 2) allow different participants to use the scale differently (e.g., scale use bias); and 3) may produce inaccurate estimates due to the cognitive demands of ranking or comparing numerous items within the same task.(41, 42) While qualitative methods, such as interviews and focus groups, can also identify aspects of health care that participants deem to be valuable, these methods do not provide us with a way to compare the value of each aspect in relation to other aspects. Conversely, using the DCE for this project allowed us to accurately, reliably,

and efficiently evaluate, quantify, and rank-order stakeholders' values and priorities regarding PPHC@Home.(40)

Second, I used cognitive interviews to provide preliminary information on the instrument's content validity and clarity. Cognitive interviewing is particularly valuable when questions are complex, sensitive, difficult, or potentially intrusive (43), and this technique has been used in recent years in other areas of palliative care research.(44-46) Cognitive interviewing may be particularly valuable in PPC research, where there are oftentimes concerns about the potential vulnerability of participants and the burden of participation on seriously-ill children and on their families.(47) Beyond the benefit of standard pilot testing, cognitive interviewing may be especially useful in PPC research because, first, it helps to minimize problems with questionnaires to make "the best use of data collected from every study participant," particularly in PPC settings where sample sizes are often small (48); second, it provides information specific to the palliative care context that may impact the validity of instruments but that may not arise in standard pilot testing of survey instruments; and third, it helps to carefully match the instrument to the capabilities of the population of interest, particularly in palliative care research where the burden of research participation should always be carefully considered.

Third, this project incorporated the perspectives of multiple stakeholders. From my literature review, I reviewed over 200 peer-reviewed papers and other published standards and guidelines about palliative and hospice care quality and outcomes. I compiled evidence from guidelines and standards of practice from palliative and hospice care and pediatric leaders such as the National Quality Forum (49), the National Hospice and Palliative Care Organization (50), and the American Academy of Pediatrics.(51) I

also reviewed quality measures from the field of adult hospice and palliative care, including the CAHPS-Hospice Survey (52) and the Bereaved Family Survey.(53) Finally, I compiled evidence from previous research studies conducted in the field of PPHC, and sought the input from interdisciplinary providers, parent advocates, bereaved parents, parents currently caring for their child, and other stakeholders such as expert researchers.

Several general limitations to this project also warrant discussion. First, the cross-sectional design of the project allowed me a snapshot of families' needs and priorities only at one point in time; it is likely that families' needs and perspectives change over the course of their child's illness experience. Although I included bereaved parents and parents currently caring for their children, I did not measure experiences of parents over time and into bereavement, and, thus, I cannot address how experiences change through the illness and caregiving trajectory. Future work will be needed to better understand how care needs and priorities in PPHC@Home evolve over time. Additionally, my study included a sociodemographically homogenous sample of parents, which is similar to challenges faced in PPC and hospice-based research.(54-57) Notably, I lacked adequate representation from fathers, racial and ethnic minorities, non-Christian religious backgrounds, lower socioeconomic groups, and non-traditional families (e.g., single-parent families, grandparent caregivers). I therefore cannot generalize findings to the greater population of families who receive PPHC@Home, although I did include parents of children with many different medically-complex conditions who received care from different PPHC@Home programs from various geographic regions of the U.S.

DIRECTIONS FOR FUTURE RESEARCH

Future research with larger, more socio-demographically- and geographically-diverse samples is needed to examine how parents' priorities for care change over time. Additionally, several other research questions remain. For example, how valid and reliable is the EXPERIENCE@Home instrument for measuring family-reported care experiences in a large sample of families currently receiving PPHC@Home? How feasible and acceptable will it be to use this instrument to measure experiences with care in real-time (that is, at the time care is being provided)? How clinically-actionable is the information produced by this instrument for clinicians caring for children and families in the home setting? My proposed postdoctoral work (F32: *The EXPERIENCE@Home Project*) will address some of these questions, in that I will examine the psychometric properties of the EXPERIENCE@Home Measure in a larger parent sample, including test-retest reliability over two administrations of the instrument. I will also evaluate the validity of the measure by examining the associations between the EXPERIENCE score and outcomes like satisfaction with care and parental distress. Lastly, I will examine the clinical utility of the EXPERIENCE@Home Measure, including the ability of the instrument to capture novel, clinically useful information that helps health care teams tailor their care to individual children and families.

Additional research beyond my post-doctoral fellowship project is also needed to evaluate the feasibility and utility of regular administrations of the tool over time (that is, how feasible will it be for families to complete this instrument routinely every week, every month, or every quarter? How useful will this instrument be for evaluating and monitoring

trends in family-reported outcomes over time?). Additionally, how responsive is the measure to changes in the child's condition, care needs, and care quality over time?

Other questions include the applicability of the EXPERIENCE@Home Measure outside of the clinical care setting. Although the primary goal of this instrument is to improve the care experience of individual patients and families, I expect that this instrument can also be used in the future to provide data to inform local and national program design and care improvement efforts. And finally, from a policy standpoint, the EXPERIENCE@Home Measure also could be used in the future to provide information on the effectiveness of PPHC-relevant policy and regulations. For example, there is a current NINR R01-funded research project to evaluate the effectiveness of pediatric concurrent care in the U.S.(58) The investigators are using Medicaid claims data that will provide valuable information about the effectiveness of concurrent care on hospice length of stay, hospice transitions, and non-hospice symptom management. It will not, however, be able to evaluate how children and families are experiencing care and what care needs may remain, even when receiving concurrent care. In the future, the EXPERIENCE@Home Measure could support similar evaluations of state and federal regulations like concurrent care by allowing us insight into how families' experiences with care are impacted by such regulations over time.

CONCLUSION

PPHC@Home varies across programs and geographic areas, likely leading to differences in patient and family outcomes. Systematically measuring how children and families experience PPHC@Home is critical for ensuring that children's and families'

most important needs and priorities are met, informing allocation of health care resources, future research priorities, and future improvement efforts.

Through this dissertation study, we have a better understanding of what aspects of care are most important to families receiving PPHC@Home. Moreover, I developed the first known family-reported instrument to measure PPHC@Home experiences in the U.S. using an innovative, rigorous, multi-method, multi-stakeholder approach. Finally, I have proposed the next steps for the evaluation and implementation of this newly developed instrument. This project aligns closely with the National Institute of Nursing Research's strategic research plan (59) and funding priorities (60), and the position of other leaders in health care like the American Academy of Pediatrics (51), the National Quality Forum (49), the National Hospice and Palliative Care Organization (50), and the Institute of Medicine (61), to advance high-quality, evidence-based community-focused palliative and hospice care, particularly for children and families. By developing an instrument to measure families' experiences with PPHC@Home, we now have a way to understand and, ultimately, help improve the care we provide in the home to all children with serious illnesses and their families.

BIBLIOGRAPHY

1. Feudtner C, Feinstein JA, Satchell M, Zhao H, Kang TI. Shifting place of death among children with complex chronic conditions in the United States, 1989-2003. *JAMA* 2007;297:2725-2732. doi: 10.1001/jama.297.24.2725
2. Feudtner C, Kang TI, Hexem KR, et al. Pediatric palliative care patients: A prospective multicenter cohort study. *Pediatr* 2011;127:1094-1101. doi: 10.1542/peds.2010-3225
3. Friebert S, Williams C. NHPCO'S Facts and Figures: Pediatric Palliative and Hospice Care in America. In: Alexandria, VA: NHPCO, 2015.
4. Nageswaran S, Radulovic A, Anania A. Transitions to and from the acute inpatient care setting for children with life-threatening illness. *Pediatr Clin North Am* 2014;61:761-783. doi: 10.1016/j.pcl.2014.04.008
5. Lindley LC, Lyon ME. A profile of children with complex chronic conditions at end of life among Medicaid beneficiaries: Implications for health care reform. *J Palliat Med* 2013;16:1388-1393. doi: 10.1089/jpm.2013.0099
6. Feudtner C, Silveira MJ, Christakis DA. Where do children with complex chronic conditions die? Patterns in Washington State, 1980-1998. *Pediatr* 2002;109:656-660. doi: 10.1542/peds.109.4.656
7. Kaye E, Rubenstein J, Levine D, et al. Pediatric palliative care in the community. *CA Cancer J Clin* 2015;65:315-333. doi: 10.3322/caac.21280
8. 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;18:143-150. doi: 10.1089/jpm.2014.0285
9. Hays RM, Valentine J, Haynes G, et al. The Seattle Pediatric Palliative Care Project: Effects on family satisfaction and health-related quality of life. *J Palliat Med* 2006;9:716-28. doi: 10.1089/jpm.2006.9.716
10. Gans D, Kominski GF, Roby DH, et al. Better outcomes, lower costs: Palliative care program reduces stress, costs of care for children with life-threatening conditions. *UCLA Center for Health Policy Research* 2012. doi:
11. Goldhagen J, Fafard M, Komatz K, Eason T, Livingood WC. Community-based pediatric palliative care for health related quality of life, hospital utilization and costs lessons learned from a pilot study. *BMC Palliat Care* 2016;15:73. doi: 10.1186/s12904-016-0138-z
12. Bona K, Bates J, Wolfe J. Massachusetts' Pediatric Palliative Care Network: Successful implementation of a novel state-funded pediatric palliative care program. *J Palliat Med* 2011;14:1217-1223. doi: 10.1089/jpm.2011.0070
13. Niswander LM, Cromwell P, Chirico J, Gupton A, Korones DN. End-of-life care for children enrolled in a community-based pediatric palliative care program. *J Palliat Med* 2014;17:589-591. doi: 10.1089/jpm.2013.0576
14. Lindley LC, Kieim-Malpass J. Quality of paediatric hospice care for children with and without multiple complex chronic conditions. *Int J Palliat Nurs* 2017;23:230-237. doi: 10.12968/ijpn.2017.23.5.230

15. Thienprayoon R, Marks E, Funes M, et al. Perceptions of the pediatric hospice experience among English- and Spanish-speaking families. *J Palliat Med* 2016;19:30-41. doi: 10.1089/jpm.2015.0137
16. Knapp CA, Madden VL, Curtis CM, et al. Partners in Care: Together for Kids: Florida's model of pediatric palliative care. *J Palliat Med* 2008;11:1212-1220. doi: 10.1089/jpm.2008.0080
17. Boyden JY, Curley MAQ, Deatrick JA, Ersek M. Factors associated with the use of U.S. community-based palliative care for children with life-limiting or life-threatening illnesses and their families: An integrative review. *J Pain Symptom Manage* 2018;55:117-131. doi: 10.1016/j.jpainsymman.2017.04.017
18. Conway PH, Mostashari F, Clancy C. The future of quality measurement for improvement and accountability. *JAMA* 2013;309:2215-2216. doi: 10.1001/jama.2013.4929
19. Dy SM. Measuring the quality of palliative care and supportive oncology: principles and practice. *J Support Oncol* 2013;11:160-164. doi: 10.12788/j.suonc.0017
20. Kuhlthau KA, Mistry KB, Forrest CB, Dougherty D. Advancing the science of measurement in pediatric quality of care. *Acad Pediatr* 2014;14:S1-3. doi: 10.1016/j.acap.2014.06.016
21. National Quality Forum. The ABCs of Measurement. In: Washington, DC: National Quality Forum, 2018. Available from: https://www.qualityforum.org/Measuring_Performance/ABCs_of_Measurement.aspx. Accessed June 30, 2020.
22. Andresen EM, Seecharan GA, Toce SS. Provider perceptions of child deaths. *Arch Pediatr Adolesc Med* 2004;158:430-5. doi: 10.1001/archpedi.158.5.430
23. Contro N, Larson J, Scofield S, Sourkes B, Cohen H. Family perspectives on the quality of pediatric palliative care. *Arch Pediatr Adolesc Med* 2002;156:14-19. doi: 10.1001/archpedi.156.1.14
24. Donnelly JP, Downing K, Cloen J, et al. Development and assessment of a measure of parent and child needs in pediatric palliative care. *J Pain Symptom Manage* 2017;55:1077-1084. doi: 10.1016/j.jpainsymman.2017.12.484
25. 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
26. Weidner NJ, Cameron M, Lee RC, et al. End-of-life care for the dying child: What matters most to parents. *J Palliat Care* 2011;27:279-286. doi: <https://doi.org/10.1177/082585971102700404>
27. Vollenbroich R, Duroux A, Grasser M, et al. Effectiveness of a pediatric palliative home care team as experienced by parents and health care professionals. *J Palliat Med* 2012;15:294-300. doi: 10.1089/jpm.2011.0196
28. Widger K, Tourangeau AE, Steele R, Streiner DL. Initial development and psychometric testing of an instrument to measure the quality of children's end-of-life care. *BMC Palliat Care* 2015;14:1. doi: 10.1186/1472-684X-14-1
29. Zimmermann K, Cignacco E, Eskola K, et al. Development and initial validation of the Parental PELICAN Questionnaire (PaPEQu): An instrument to assess parental experiences and needs during their child's end-of-life care. *J Adv Nurs* 2015;71:3006-3017. doi: 10.1111/jan.12741
30. Thienprayoon R, Lee SC, Leonard D, Winick N. Hospice care for children with cancer: Where do these children die? *J Pediatr Hematol Oncol* 2015;37:373-377. doi: 10.1097/MPH.0000000000000331

31. Verberne LM, Kars MC, Schouten-van Meeteren AY, et al. Aims and tasks in parental caregiving for children receiving palliative care at home: a qualitative study. *Eur J Pediatr* 2017;176:343-354. doi: 10.1007/s00431-016-2842-3
32. Lindley LC, Edwards SL. Geographic access to hospice care for children with cancer in Tennessee, 2009 to 2011. *The American Journal of Hospice & Palliative Care* 2015;32:849-854. doi: 10.1177/1049909114543641
33. Lindley LC, Edwards SL. Geographic variation in California pediatric hospice care for children and adolescents: 2007-2010. *Am J Hosp Palliat Care* 2018;35:15-20. doi: 10.1177/1049909116678380
34. Kaye EC, Gattas M, Kiefer A, et al. Provision of palliative and hospice care to children in the community: A population study of hospice nurses. *J Pain Symptom Manage* 2019;57:241-250. doi: 10.1016/j.jpainsymman.2018.10.509
35. Johnston EE, Rosenberg AR, Kamal AH. Pediatric-specific end-of-life care quality measures: An unmet need of a vulnerable population. *J Oncol Pract* 2017;13:e874-e880. doi: 10.1200/JOP.2017.021766
36. O'Leary N, Flynn J, MacCallion A, Walsh E, McQuillan R. Paediatric palliative care delivered by an adult palliative care service *Palliat Med* 2006;20:433-437. doi: 10.1191/0269216306pm1147oa
37. McConnell T, Porter S. The experience of providing end of life care at a children's hospice: a qualitative study. *BMC Palliat Care* 2017;16:15. doi: 10.1186/s12904-017-0189-9
38. Clarke J, Quin S. Professional carers' experiences of providing a pediatric palliative care service in Ireland *Qual Health Res* 2007;17:1219-1231. doi: 10.1177/1049732307308316
39. Sanders CM. A comparison of adult bereavement in the death of a spouse, child, and parent. *OMEGA* 1980;10:303-322. doi: <https://doi.org/10.2190/X565-HW49-CHR0-FYB4>
40. Muhlbacher A, Johnson FR. Choice experiments to quantify preferences for health and healthcare: State of the practice. *Applied Health Economics and Health Policy* 2016;14:253-266. doi: 10.1007/s40258-016-0232-7
41. Cohen S, Orme B. What's your preference? *Market Res* 2004;16:32-37. Available from: https://in4ins.com/wp-content/uploads/2018/08/i4i_WhatsYourPref.pdf. Accessed June 30, 2020.
42. Sawtooth Software. What can MaxDiff Best-Worst Scaling do for you? 2018. Available from: http://www.sawtoothsoftware.com/index.php?option=com_content&view=article&id=1907. Accessed June 22, 2020.
43. Drennan J. Cognitive interviewing: Verbal data in the design and pretesting of questionnaires. *J Adv Nurs* 2003;42:57-63. doi: 10.1046/j.1365-2648.2003.02579.x
44. Murtagh FE, Addington-Hall JM, Higginson IJ. The value of cognitive interviewing techniques in palliative care research. *Palliat Med* 2007;21:87-93. doi: 10.1177/0269216306075367
45. Ahmed N, Bestall JC, Payne SA, Noble B, Ahmedzai SH. The use of cognitive interviewing methodology in the design and testing of a screening tool for supportive and palliative care needs. *Support Care Cancer* 2009;17:665-673. doi: 10.1007/s00520-008-0521-2
46. Dy SM, Al Hamayel NA, Hannum SM, et al. A survey to evaluate facilitators and barriers to quality measurement and improvement: Adapting tools for implementation research in palliative care programs. *J Pain Symptom Manage* 2017. doi: 10.1016/j.jpainsymman.2017.06.008

47. Tomlinson D, Bartels U, Hendershot E, et al. Challenges to participation in paediatric palliative care research: A review of the literature. *Palliat Med* 2007;21:435-440. doi: 10.1177/0269216307077173
48. Reeve BB, McFatrigh M, Pinheiro LC, et al. Eliciting the child's voice in adverse event reporting in oncology trials: Cognitive interview findings from the Pediatric Patient-Reported Outcomes version of the Common Terminology Criteria for Adverse Events initiative. *Pediatr Blood Cancer* 2017;64. doi: 10.1002/pbc.26261
49. National Consensus Project for Quality Palliative Care. *Clinical Practice Guidelines for Quality Palliative Care*. Richmond, VA: National Coalition for Hospice and Palliative Care; 2018. Available from: <https://www.nationalcoalitionhpc.org/npc/>. Accessed June 5, 2020.
50. National Hospice and Palliative Care Organization (NHPCO). *Standards of practice for pediatric palliative care*. Alexandria, VA: NHPCO; 2019 2009. Available from: https://www.nhpc.org/wp-content/uploads/2019/07/Pediatric_Standards.pdf. Accessed June 12, 2020.
51. Feudtner C, Friebert S, Jewell J. Pediatric palliative care and hospice care commitments, guidelines, and recommendations. *Pediatr* 2013;132:966-972. doi: 10.1542/peds.2013-2731
52. CAHPS Hospice Survey: Frequently asked questions. Centers for Medicare & Medicaid Services, Baltimore, MD 2019 Feb 28. Available from: <https://hospicecahpsurvey.org/en/hospice-specific-faqs2/#Q8>. Accessed June 30, 2020.
53. Kutney-Lee A, Carpenter J, Smith D, et al. Case-mix adjustment of the Bereaved Family Survey. *Amer J Hosp Palliat Med* 2018;35:1015-1022. doi: 10.1177/1049909117752669
54. Hinds PS, Burghen EA, Pritchard M. Conducting end-of-life studies in pediatric oncology. *West J Nurs Res* 2007;29:448-465. doi: 10.1177/0193945906295533
55. Macdonald ME, Chilibeck G, Affleck W, Cadell S. Gender imbalance in pediatric palliative care research samples. *Palliat Med* 2010;24:435-44. doi: <https://dx.doi.org/10.1177/0269216309354396>
56. Knapp CA, Madden VL, Curtis C, Sloyer PJ, Shenkman EA. Assessing non-response bias in pediatric palliative care research. *Palliat Med* 2010;24:340-7. doi: <https://dx.doi.org/10.1177/0269216309351466>
57. Cohen LL. Racial/ethnic disparities in hospice care: A systematic review. *J Palliat Med* 2008;11:763-768. doi: 10.1089/jpm.2007.0216
58. Lindley LC. Effectiveness of concurrent care to improve pediatric and family outcomes at end of life (R01NR017848). University of Tennessee Knoxville NINR, 2018.
59. National Institute of Nursing Research. *The NINR's Strategic Plan: Advancing Science, Improving Lives*. 2016 September. Available from: https://www.ninr.nih.gov/sites/files/docs/NINR_StratPlan2016_reduced.pdf. Accessed June 26, 2020.
60. PAR-19-320: *Palliative Care in Home and Community Settings (R21 Clinical Trial Optional)*. National Institutes of Health. Posted July 31, 2019. Available from: <https://grants.nih.gov/grants/guide/pa-files/par-19-320.html#:~:text=Home%20and%20community%20in%20this,of%2Dlife%20and%20palliative%20care>. Accessed June 27, 2020.
61. Field MJ, Behrman RE. *When Children Die: Improving Palliative and End-of-Life care for Children and their Families*. Washington, DC: The National Academies Press, 2003:419-420.

Available from: <https://www.nap.edu/catalog/10390/when-children-die-improving-palliative-and-end-of-life-care>. Accessed June 5, 2020