Stress and Coping in Parents of Children with Medical Complexity

By

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Dissertation

Submitted to the Faculty of the

Graduate School of Vanderbilt University

in partial fulfillment of the requirements

for the degree of

DOCTOR OF PHILOSOPHY

in

Nursing Science

December 18, 2021

Nashville, Tennessee

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DEDICATION

This research is dedicated to the parents of children with medical complexity.
You allowed me to share in such a personal part of your lives.
I hope I described your experiences with the same courage, strength, compassion and love
you conveyed to me. I am a better person for it and am eternally grateful.

ACKNOWLEDGMENTS

I could not have achieved this amazing milestone without the help of an entire village that has supported me throughout this journey. I would like to first thank the Vanderbilt University PhD in Nursing Science program and all the faculty for providing me the foundation to build a strong and enduring research career. Funding provided by the Vanderbilt Student Achievement Research Award was pivotal to supporting the study and for that I am extremely grateful. To Dr. Natalie Truba for her work in getting the study off the ground, you were monumental.

This dissertation was guided by some very special people. First, my Dissertation Chair, Dr. Mary Jo Gilmer has encouraged me, pushed me and believed in me even when I didn't believe in myself. Without her this work would not have been possible. My dissertation committee, Dr. Terrah Foster Akard, Dr. Mary S. Dietrich, and Dr. Tracy Hills provided insight that questioned and challenged my thinking, making this research even more worthwhile. I would also like to thank Kemberlee Bonnet and Dr. David Schlundt for sharing their qualitative research expertise with me and teaching me along the way.

My friends and family have gone above and beyond to help compensate for my absences when my attention was focused on reaching the finish line. For this I am eternally grateful. To my mom, dad and siblings, you made me believe from the start if you put your heart into something and you want it bad enough you can accomplish anything. To my beautiful sons and daughter, who have grown to be kind, considerate, compassionate, and often hilarious young men and an amazing young woman….oh the places I know you will go. I hope I have shown you that even when things are difficult it is ok to take the road less traveled and make your own path if you do it with strong conviction and certainty that you can accomplish great things.

Lastly to my husband Mike, who has been there through thick and thin, keeping the boat afloat at all times even despite some rough waters, you will never know how much your love and support have meant to me. You were always the one who was certain I could accomplish this no matter what, you were my greatest cheerleader. I hope you know I will always do the same for you.

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Introduction

Statement of Problem

The population of children with medical complexity (CMC) is growing secondary to numerous healthcare advances.[1] Development of therapies and treatments, early detection and increased access to care have improved long-term survival from previously fatal conditions. This progress has allowed parents to provide the majority of their children's multifaceted care at home.[2] Challenges facing parents providing this specialized health care include adverse effects on their mental well-being, poorer physical health, financial stressors, and higher prevalence of anxiety and depression.[3] Parenting capabilities may be compromised.[4] A paucity of research currently exists addressing possible associations of parent and child characteristics, parents' frequency and difficulty of stress, and types of coping strategies used by parents of children with medical complexity (CMC).

The majority of the care of the CMC population is provided by their parents in the home.[2] Parents of CMC are put into the position of not only being the child's guardian, but also the medical caregiver. Research has shown providing care may be an especially difficult situation for parents with respect to their child's illness, not encountered daily by parents of healthy children.[5] There are emotional, psychological and physical impacts on parents of CMC, affecting their day-to-day functioning and overall well-being. Emotions ranging from fear, anxiety, depression, and post traumatic symptoms[6] have been identified in parents of CMC. The following paragraphs explain the variables of focus in this dissertation.

Since 1988, the Maternal and Child Health Bureau has referred to CMC as children with special health care needs (CSHCN). They were defined previously as "children who have or are at an increased risk of a chronic, physical, developmental, behavioral or emotional condition and require health care and related services beyond that required of children generally."[7] Recently, a focus on CMC as a sub-set of the CSHCN population encompasses children with more intense health care needs. For example, support for a child with a single health issue such as a speech delay, may not require the same support as a child with spastic quadriplegia accompanied by other disorders.[1] For purposes of this dissertation, CMC will be defined operationally as children who have more than one chronic condition, requiring care in a midwest hospital's Complex Care Clinic.

Lazarus and Folkman in 1986 proposed that stress occurs when people feel demands put on them from external situations (environment) that are viewed by the person to exceed their current resources.[8] Parents of children with medical complexity often suffer from high levels of emotional and physical stress.[2] They are faced with the extraordinary challenge of being a parent, while providing necessary medical care to their child. Travel required for specialized health care on a routine basis can create an increased level of stress for parents.[9] Additional impact from negative stressors to this unique parent population also may include emotional, psychological, social, and financial struggles.[10]

Coping strategies are frequently changing conscious efforts to adapt or solve an internal or external stressful demand.[8] They are active ways of responding to threatening situations. Coping strategies are ways parents adapt to the stressors of raising a CMC. Parents of children with medical complexity have reduced levels of stress when there are increases in helpful coping strategies.[11] Coping strategies are often divided into three different types: problem-focused coping strategies that aim to solve the problem or do something to alleviate the stress, emotion-focused coping strategies that aim to reduce or manage the feelings of the stress, and dysfunctional coping that employs avoidance, denial and self-blame.[11] Research demonstrates when CMCs exhibited more severe limitations and their parents used higher levels of problem-focused coping, they had decreasing levels of depression, but when using more emotion-focused coping strategies, the parents exhibited increasing burden felt from caring for their CMC over time. Dysfunctional coping strategies often lead to maladaptation to their children's medical complexity.[12]

Purpose of Study

The purpose of this study was to explore parental perceptions of frequency and difficulties of stressful events and coping strategies when caring for CMC. The research included use of surveys to examine potential associations between child and parent characteristics, frequency and difficulties of stressors, and types of coping strategies. A qualitative component provided additional meaning to the data collected in the surveys and facilitated further understanding of the factors that influence coping strategies used by parents of CMC.

Specific Aims

The aims of this study included:

Aim 1: Describe levels of frequency and difficulty of parental stress when caring for children with medical complexity.

Aim 2: Describe types of coping strategies (dysfunctional, problem-based and emotion-focused) used by parents of children with medical complexity.

Aim 3: Describe associations among children's characteristics (gender, age, race and date of diagnosis) and parent characteristics (gender, race, age, income, marital status and level of education) and types of coping strategies (dysfunctional, problem-focused and emotion focused) used by parents of children with medical complexity.

Aim 4: Examine the impact of caring for children with medical complexity on parents through qualitative interviews.

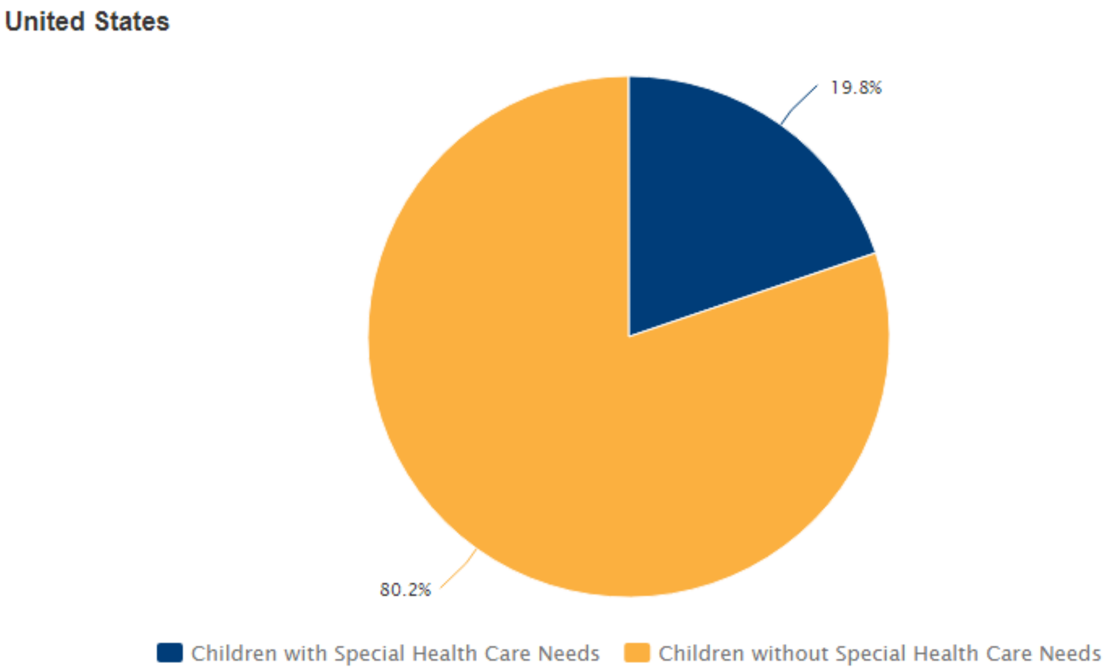
Significance

The Health Resources and Service Administration estimates that CMC represent almost 20% of all children in the United States (See Figure 1).[7] The care for these children continues to be substantial and wide-ranging. Parents provide over 1.5 billion hours of care in their homes every year. Parents' responsibilities include routine daily child care coupled with technical[13] and nursing care.[14] Parents are faced with the tasks of balancing family life[15] and work responsibilities,[16] while coordinating and/or providing their child's acute care.[17] Parents who provide this level of multifaceted care often have a higher incidence of depression,[18] PTSD,[19] anxiety,[2] and stress[20] due to social, emotional and financial strains that accompany having CMC.[21]

Children with Medical Complexity (CMC) encompass a diverse group. Every child is unique in their disease, medical and physical needs, and limitations, yet the care provided from the perspectives of their parents is often similar.[22] Care needs of the children may include administering complex medications, suctioning, bowel care, oxygen and catheterization. Definitions of CMC describe children with extensive chronic health problems that affect multiple organ systems and result in functional difficulties, high health care utilization, and often the need for or use of medical technology.[17, 23]

Figure 1

Estimates of Numbers of Children with Medical Complexity



A national profile reports that the cost of caring for CMC comprise as much as one-third of health care spending of all children (nearly $100 billion).[23] However, the cost of care provided by parents in their own homes has not been calculated. In an analysis of care of CMC from 2009-2010, replacing the parents with a home health aide would have cost $6400 per child each year, for a total of $35.7 billion. Wages lost by the parent providing the care were also in excess of $17 billion per year.[4] The families of CMC had three times the health care expenses of healthy children.[3]

Parents who provide care to CMC, especially those who rely heavily on medical technology, can often feel overwhelmed, and their sense of responsibilities are never-ending.[24] Institutional care to has shifted to care in the home setting.[25] Gonzalez et al. found that parents who provide home care are often emotionally drained and their quality of life scores are lower than those of parents with healthy children.[26] Studies have shown these parents often suffer from feelings of isolation,[27] exhaustion,[28] anxiety and depression.[29] Identifying associations among parental and child characteristics and parents' levels of stress and use of coping strategies may provide a foundation for further work to provide adequate support to parents when they voice challenges or barriers accompanying their child's illness trajectory.

Chapter 2

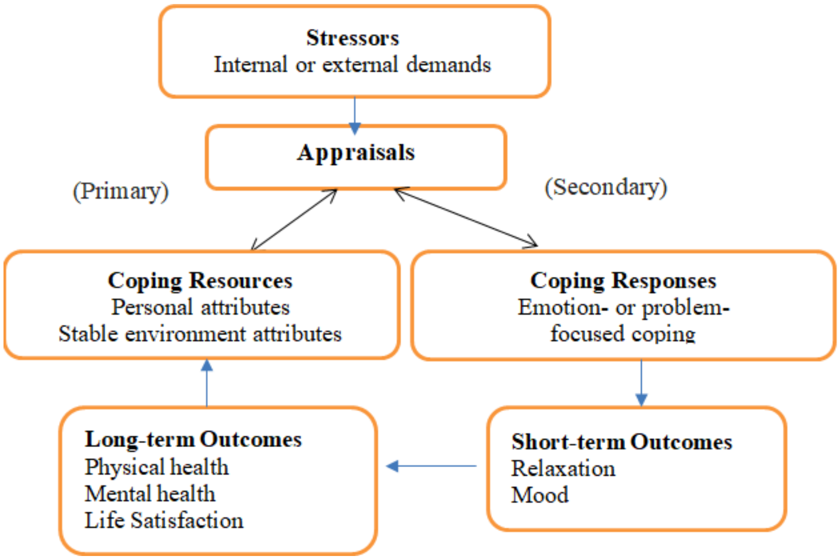
Theoretical Framework and Literature Review

Theoretical Framework

A lack of an accepted framework to address parental and child characteristics associated with parents' levels of stress and coping strategies exists in the CMC population. Various coping strategies are used by parents as they respond to stress in caring for their CMC as described in the Transactional Model of Stress and Coping (Figure 1).[8] Parents' levels of stress influence outcomes of their types of coping strategies.

Figure 2

The Transactional Model of Stress and Coping

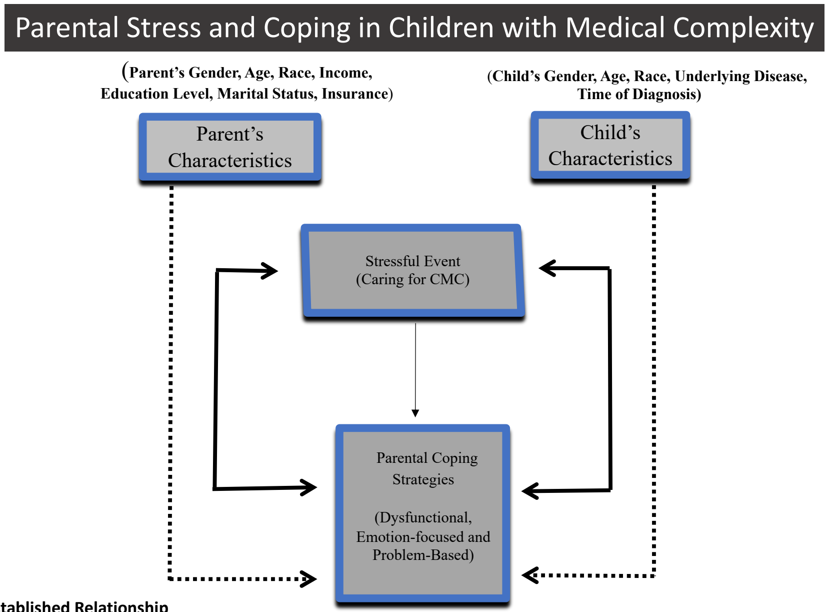


The Transactional Model of Stress and Coping developed by Lazarus and Folkman (Figure 2) describes the coping strategies of problem-focused coping and emotion-focused coping.[8] Problem-focused coping is the ability to directly manage the stressor, in a more positive way; whereas emotion-focused coping is a reaction to the stressor, in a more negative way.[8]

Through incorporating this important framework, an adapted model of Parental Stress and Coping in Children with Medical Complexity guides this study as illustrated below (Figure 3).

Figure 3

Parental Stress and Coping in Children with Medical Complexity



*Solid Line indicates Established Relationship
*Dotted Line indicates Proposed Relationship

Folkman S, Lazarus RS, Gruen RJ, DeLongis A. Appraisal, coping, health status, and psychological symptoms. J Pers Soc Psychol. 1986;50(3):571-9

Building on the Transactional Model of Stress and Coping,[5] the adapted theoretical model helps to define and direct inquiries of parental coping in the CMC population. The model depicts the association of parental characteristics and child characteristics and how they may affect the stressor (Figure 3). The stressor is identified as the act of caring for the CMC. The emphasis is on what direct effect the constellation of parental and child characteristics has on coping strategies. The model is wide-ranging in scope, looking at these characteristics collectively, the relationship to stress and the outcome of coping strategies. This model highlights CMC, but the application may be relevant to other chronic illnesses in childhood. The versatility of the model could be applied to different groups of caregivers affected by stress as well (Figure 3). To provide clarity for the reader, both conceptual and operational definitions are described below.

Critical Analysis of Relevant Literature

The literature examining coping strategies of parents of children with medical complexity was reviewed. Due to the lack of literature investigating stress and coping strategies in parents of children with complex medical needs, the review was expanded to encompass other relevant studies including those of parents of children with any chronic medical condition. This included studies that involved at least one group of primary caregivers of children who had an intellectual, physical impairment and/or a chronic illness. Studies were also included that addressed the impact of stress that caring for their child may have on the parent's ability to cope.

Parental characteristics of age, gender, educational level, income, marital status, race and child demographics of gender, age, race, and type of insurance are understudied as they relate to parental coping strategies of CMC. Limited studies investigate how child disease characteristics, the etiology of the underlying disease, and date of diagnosis influence parents' coping strategies. Rarely has the combination of both parental and child characteristics and their potential relationship to parental coping been studied together. Variability in these factors may be associated with a diversity of coping strategies used by parents. Different child diseases present different prognoses and degree of care needed. Disparities in neurocognitive levels and treatments could also affect parental coping.[30] For purposes of this study, child demographic and childhood disease characteristics will be combined under the construct of child characteristics and considered along with parental characteristics as they affect parental coping.

The initial search yielded a total of 38 articles. After closer examination, 16 of the studies were excluded, with 22 remaining for analysis. Synthesis of the review of literature includes 19 studies focused on parents of children with complex medical needs.[31-50] The remaining 3 studies targeted parents of children with cystic fibrosis,[51] autism spectrum disorder,[52] and complex congenital malformations, known as VACTERL Association.[53] The following paragraphs describe the study designs, sampling, methodology, measurements, descriptions of any theoretical frameworks identified and limitations of the reviewed literature.

Study Aims and Designs of Reviewed Literature

Eight cross-sectional studies reviewed included several different foci. The three initial studies described parental caregiver burden of children with chronic conditions,[31, 40, 47] with one of those focusing on predictors,[40] and two on the association of psychosocial factors with caregiver burden.[31, 40, 47] Two cross-sectional studies addressed specifically parents' coping strategies of CMC.[6, 54] One investigated the association with parental stress and family's quality of life among parents of children with intellectual disabilities,[41] one examined support needs and coping strategies among mothers of children with Autism Spectrum Disorder (ASD)[50] and finally an investigation of a structural model of family empowerment for families of children with special needs was described.[39] Five of the reviewed studies were reviews of literature addressing coping trends,[50] health and well-being,[35] positive parenting aspects,[34] special health care needs at home,[46] and chronic sorrow of parents of children with medical complexity.**[36]** Two studies were retrospective designs investigating differences in perceived needs of parents of children with ASD[52] and challenges of caregivers of medically complex children.[45] One longitudinal study described humor styles among parents of children with disabilities. [38] Finally, a mixed-methods study focused on improving transitions in care for CMC.[31]

Sampling. The majority of studies used convenience sampling with the exception of two qualitative studies that recruited using purposive[32, 43] and snowballing sampling techniques.[43] The caregivers surveyed included on average 70% female, and 30% male, though 3 studies only described "caregivers" with no differentiation of gender.[38, 43, 49] Parents were primarily Caucasian, with a range of socioeconomic and educational levels represented in the samples. The reasons some participants declined study participation was not readily described across studies. Parents and caregivers with a desire to provide information on their experiences may have been more likely to share and complete study questionnaires, focus groups and interviews than those who were more inclined to remain private; thus, all studies may not be generalizable to all CMC parents.

Methodology. Eight studies used quantitative approaches to investigate stress and coping in parents of children with chronic illness, or complex medical complexity.[31, 37, 40, 41, 47, 50, 51, 54] Five qualitative studies included discussions of parental challenges,[49] impact of caring for children with complex medical needs,[43] how to facilitate continuity of care,[48] coping strategies[32] and finally adaptation to their children's illness.[53] One study used a mixed-methods approach.[37] The quantitative approach enabled comparison of stress and coping strategies between different caregivers and association with socio-demographic variables across studies. However, the qualitative data provided richness and meaning that may not have been identified by collection of the quantitative data alone.

Measurements

Quantitative measurement. Among the eight quantitative studies, [31, 37, 40, 41, 47, 50, 51, 54] numerous instruments were used to assess stress and coping in parents of children with chronic illness. Of these, only three measures were used in more than one study. The Zarit Burden Interview Measure,[31, 40, 47] Parenting Stress Index-Short Form (PSI-SF) [41, 50] the and Brief Cope (B-Cope) were all used in more than one study.[37, 51] It is important to note that the Brief-Cope in the study by Curran was not used at all 4 research time points but rather only T2 and T4.

The Zarit Burden Interview[40] is a self-report scale that evaluates a respondent's perception of stress based on the caregiver's experience.[55] It includes 22 five-point Likert-type items using a response scale ranging from 0 (never) to 4 (always). The sum of 22 items totals a score ranging from 0 to 88. Higher scores indicate a greater level of burden.[55]

The PSI-SF is a 36-item sub-scale that assesses parental stress.[41] The items are grouped into 3 sub-scales. The *difficult child* measures a child's self-regulatory abilities as perceived by the parent, the *parent-child dysfunctional interaction* assesses parental dissatisfaction of interactions with the child, and the degree to which parents find it unacceptable. The third sub-scale is *conflicts with a partner and life-restrictions due to child-rearing.* It is related to feelings of loss of control, and/or dissatisfaction. The Cronbach's alpha for the total score was 0.92.[41]

The B-Cope is a 28-statement instrument that represents 14 groups of stress coping strategies.[51] These statements are grouped into 14 sub-scales and each one relates to a particular coping strategy. Each item is evaluated according to a four-item scale (never-sometimes-often-always) ranging from 1 to 4 points. Th e split-half reliability for the 14 subscales is 0.86.[51]

Socio-demographic variables. Most instruments used to gather information about the socio-demographic variables of the parents and/or children were author developed. The only tool with tested reliability and validity administered to participants was the Sociodemographic Variables Questionnaire

(Q-SV).[47] The Q-SV was developed to focus on family caregivers of children with chronic diseases including but not limited to their age, sex, marital status, level of education, and monthly family income. In addition, it included characteristics of the ill child such as sex, age, diagnoses and length of time since the chronic illness was identified.[47]

Qualitative measurement. Five qualitative studies utilized semi-structured interviews and focus groups.[43,32,38,48,49] A wide array of open-ended questions, items and probes were selected to explore several topics relevant to parenting CMC. None of the qualitative interview questions focused specifically on stress and coping strategies. All focus groups were conducted in person by the principal investigators (PI's), except for Zanello et al., where three participants requested phone interview only.[48]

There were several qualitative methods used to analyze transcripts from the interviews and focus groups. Varying from a thematic approach,[43] researchers used content analysis, directed approach,[48] and analysis of an established framework.[32] Aims of the qualitative methods were to explore, categorize and interpret the data to maximize an accurate understanding of participants' experiences.

Theoretical Framework Analysis of Current Literature

Theoretical frameworks describe the concepts, variables and relationships to be examined in a research study.[56] The majority of studies reviewed did not clearly explain a theoretical basis. Six studies of the 22 reviewed described theoretical perspectives, two using the Family Adjustment and Adaptation Model (FAAR model).[45] The model was chosen in both instances as it placed an emphasis on family capacity and potential for growth.[41, 57] Two studies used Lazarus and Folkman's Transactional Model of Stress and Coping as theoretical foundations as it deals with both the process and context of stressful situations.[51, 58] A separate study built on the Double ABX Model as family adaptation was identified as its core concept.[39] It is also described as the effort made by the family to reach a new level of equilibrium.[59] Lastly, the Conceptual Framework of Coping Strategies by Burr and Klein,[60] was used to guide conceptualization.[58] Consistent themes were recognized in the theoretical foundations, coping, and adaptation; however, there was no single identified theoretical framework for stress and coping in parents of CMC. The following paragraphs synthesize the specific concepts focused on as part of a review of this literature, with the study limitations and finally, the subsequent identification of gaps as found in this critical analysis of the current literature.

Study Concepts of Focus in Literature Review

Stress. There is an increased demand on parents when raising children with medical complexity as families are faced with their developmental and functional limitations. Parents of CMC have unique types of stressors, facing daily challenges and dealing with perpetual feelings of crisis.[58] Parents who have a greater level of social support have been identified as having less stress and enhanced mental health overall.[38] Parents described the support of other families in similar situations as significantly decreasing their level of stress.[49] Adaptive coping strategies that were problem-focused and that reflected positive thinking were found to be most helpful in decreasing stress; while mothers' describing unmet needs were found to increase maternal stress.[50]

Coping Strategies. The use of effective coping strategies has been established as an important mechanism of managing parental stress and well-being.[50] Chronic illnesses in children challenges parents' resources and the use of positive coping strategies.[46] Emotion-focused coping strategies were related to greater parental stress, while problem-focused coping strategies such as information seeking and acceptance were frequently used coping strategies by both parents.[32]

Characteristics. Mothers and fathers often experience caring for their CMC very differently. Unfortunately, often parents are studied as a couple, or only the mothers are examined, as they are more frequently the care provider.[5] Though studies examined varied in results with some finding significantly higher stress in mothers,[58] others showed no statistically significance differences.[34] Differences in coping strategies between genders remain unclear, and continued investigation is necessary. The relationships between SES and specific parental coping strategies are similarly understudied. Parents of CMC from lower SES were more likely to suffer from financial implications of needing to quit their jobs to provide long term care, especially in homes where the CMC required mechanical ventilation.[45] No studies specifically investigated how insurance plans meet the needs of the families who are providing care at home to their CMC.[45]

Study Limitations of Reviewed Literature

The common challenges identified in the reviewed stress and coping studies in parents of children with chronic illnesses included small, homogenous sample populations. Most of the studies examined were cross-sectional designs, limited to only one longitudinal design out of 22 reviewed. In all studies, findings were subject to recall bias and how well caregivers understood the questions. This may not allow for generalizability across patient populations.

None of the studies had a comparative group of parents with healthy children. Across the studies, no one measure was identified as the “gold standard” of measurement of stress or coping strategies. One study specifically cited a limitation of their research lacking in utilization of measures that addressed resilience, coping strategies and social skills.[41] Study samples also may have been overrepresented by parents who are mentally stable and emotionally adjusted that would choose to participate as subjects in research studies versus those who were not (inherent response bias).

Gaps in the Literature

The primary gap identified in the review of literature is the lack of studies of child and parental characteristics in parents of CMC associated with parents’ stress and types of coping strategies used. Cross-sectional, correlational studies are needed to evaluate the associations among variables as presented in the adapted conceptual framework. The goal of this study was to address the gaps in the literature needed to build a case for later intervention studies and test the suggested framework, using a sample of parents of children with medical complexity. Because child and parental characteristics have been understudied in parents of CMC, a long-term goal would be to address these characteristics with further examination in studies across the lifespan. Longitudinal studies would fill a gap and provide insight into development of interventions designed to support families faced with challenges consistent with caring for children with medical complexity.

Discussion

Factors that influence the degree to which parents use various coping strategies are not fully understood and need to be examined to build a foundation that will guide future research and enhance the quality of care in this vulnerable population. Previous work on coping strategies of parents of CMC have focused on some demographic characteristics of the parent.[45] A review of the literature did not find a study that investigates the constellation of both parent and child characteristics and their relationships to the level of stress and coping strategies by parents of CMC. To develop effective interventions, an understanding of these factors that place parents of CMC at higher risk for dysfunctional coping is crucial. Knowledge gained from research exploring characteristics of both parents and children that may affect parental coping strategies will contribute to the sparse information currently available and establish a basis for a program of research intended to explore this phenomenon further, develop therapeutic interventions, and improve quality of life and overall adaptation in parents of CMC.

Definition of Terms

Children with Medical Complexity (CMC)

Conceptual Definition. Children with medical complexity (CMC) are those with persistent illnesses that cause mental, emotional, and physical difficulties, often leading to use of advanced medical equipment coupled with an increase in the use of the health care system. CMC may have a constellation of medical diagnoses but have higher risks of gaps in care, needing pre-emptive, wide reaching, and a highly organized level of care.[61]

Operational Definition. Any child with a medical diagnosis that requires care at a midwest hospital Complex Care Clinic.

Parent

Conceptual Definition. A person who is responsible for rearing a child, regardless of whether it is the father, mother, protector, or guardian.

Operational Definition. The person, regardless of gender or biological relationship to the child, who identifies themselves as the primary caregiver.

Stress

Conceptual Definition. Stress is a “particular relationship between the person and the environment that is appraised by the person as taxing or exceeding his or her resources and endangering his or her well-being."[8]

Operational Definition. The results of the Pediatric Inventory for Parents-Short Form (PIP-SF).

Coping Strategies

Conceptual Definition. Coping strategies are the "specific efforts, both behavioral and cognitive, that people adopt to master, tolerate, reduce, or minimize stressful events. Two major categories of coping strategies are widely recognized: problem-focused coping strategies (efforts to do something active to alleviate stressful circumstances) and emotion-focused coping strategies (efforts to regulate the emotional consequences of stressful or potentially stressful events)."[8]

Operational Definition. The results of the Brief-Cope. (B-Cope).

Child Characteristics

Conceptual Definition. The child characteristics to be examined are inclusive of some demographics and some disease variables and include gender, age, race, diagnosis, and use of medical technology.

Operational Definition. The child's characteristics will be determined utilizing the information from the demographic form provided by the parents at time of their survey completion.

Parent Characteristics

Conceptual Definition. The parents' characteristics to be examined include gender, race, age, income, marital status and level of education.

Operational Characteristics. The parent's characteristics will be determined utilizing the information from the demographic form provided by the parents at time of their survey completion.

Chapter 3

Methodology

Research Design and Assumptions

This dissertation used a mixed-methods, descriptive cross-sectional study design. The first phase of the study was used a quantitative survey approach; the second phase used a qualitative approach comprised of semi-structured individual interviews.

Assumptions for the study included:

1) Caring for a child with medical complexity is a stressful experience.
2) Parents may utilize a variety of coping strategies to deal with the demands of caring for a child with medical complexity.
3) Variables such as parents' gender, age, race, income, and level of education play a role in the level of stress and coping strategies used by parents.
4) Variables such as children's gender, age, race and data of diagnosis play a role in the level of stress and coping strategies used by parents.

Participants and Setting

Parents of children with medical complexity (CMC) who receive care at a pediatric hospital in the Midwest were invited to participate in the study. Parents were eligible if their child was between 1 month and 21 years of age and had or were at increased risk for chronic physical, developmental, behavioral, or emotional conditions. Any biological or legal guardians of the CMC, or any full-time caregivers were eligible. Any parent that had a decreased cognitive ability as determined by the child's primary care team was excluded. Parents who were unable to communicate in English were also excluded.

Procedures for Study Recruitment

The study received Institutional Review Board approval prior to recruitment. Data collection occurred January to April 2021. The private investigator (PI) met with the medical director, nurse manager, physicians, nurse practitioners, staff nurses and social workers in the Pediatric Complex Care

Clinic and discussed details of the study, including the proposed aims, inclusion and exclusion criteria, and study procedures. The clinic team members agreed to help assess study interest in potential participants during their scheduled visits.

Complex care providers who participated in the information session provided recruitment brochures to parents while the parents were waiting for the visit to begin. Brochures included a description of the study project and eligibility criteria for participation during clinic appointments. Because of COVID restrictions to clinic visits, providers occasionally provided the information to families via their telehealth appointments. When parents voiced interest in participating in the study during these appointments, the Complex Care Team notified the PI.

The PI confirmed parents' study eligibility in collaboration with the complex care psychologist and clinic manager. The PI contacted interested parents by phone or email to share more information about the study, confirmed eligibility, and answer any questions. When eligible parents expressed continued interest, the PI described the study in more detail and answered any questions. The PI also informed potential participants that they were eligible to receive a $25 Amazon gift card in appreciation for their time to participate in the study. After obtaining verbal consent, the PI then emailed a link to a REDCap (secure web-based application for building and managing online surveys and databases)[62] electronic written consent and the accompanying surveys to the parent within 24 hours of contact, with a request for surveys to be completed within one week. The PI sent a reminder email through REDCap for surveys not completed within 5 days.

During the initial contact with the parents, this PI assessed interest in their potential participation in the semi-structured telephone interviews. Those who agreed were provided further detail. The PI requested a telephone call with the parent for approximately 20-30 minutes, at their convenience. The PI explained open-ended questions would be asked regarding the parents' experiences of caring for their CMC. The inclusion of a $50 Amazon gift card as a thank you for their time was also discussed.

Strategies to Ensure Human Subjects Protection

Approval of the study and the study protocols were obtained from the Vanderbilt Institutional

Review Board (IRB), and the Midwest pediatric hospital where the study took place prior to the onset of any study activities. Electronic consent was obtained prior to starting any study procedures. Potential participants were informed that quantitative data collected would be de-identified and reported in aggregate, ensuring parent participants that their answers would not be linked to any of their identifying information. Qualitative data were also de-identified, and permission was obtained to use any quotes in future manuscripts or publications. The study did not include any hazardous procedures, situations, or material. There was minimal risk to the participants with possible emotional distress being the primary concern for the parents due to the sensitive nature of their child with medical complexity.

Data Collection Methods

Procedures

Survey. After completing written e-consent, parents advanced to the study surveys. The surveys included demographic questions, a questionnaire assessing the frequency and difficulty of potentially stressful events, and a measure of coping strategies used by parents of CMC. All surveys totaling approximately 60 questions and were loaded into REDCap and participants reported completion taking 25- 30 minutes in total. REDCap was programmed to flag missed items. It has the capability of ensuring each question is completed or is intentionally skipped by the participant before advancing to the subsequent items. This minimized randomly missing items within each of the measures.

Interview. After informed consent was obtained parents completed semi-structured interviews one-on-one with this PI on the telephones. Interviews were recorded and then stored as de-identified files in a locked folder on the PI's password protected computer. The files were uploaded to Vanderbilt Box for qualitative coding with the Vanderbilt Qualitative Core. The interviews consisted of about 10 open ended questions, and interviews lasted between 26-96 minutes.

Instruments

Stress and Coping Measures

Aligned with the previously discussed theoretical framework guiding this study, the two surveys chosen for this study were also based on Lazarus and Folkman's theoretical framework. The Pediatric

Inventory for Parents Short Form (PIP-SF) questionnaire measured frequency and difficulty of potentially stressful events.[63] The Brief Cope (B-Cope) questionnaire measured coping strategies used by parents of CMC.[64] These measures have excellent psychometrics and have been commonly used in other studies focusing on stress and coping.[65] Selection of these instruments further established their psychometrics in a new population of caregivers not previously studied.

Parental Stress Measure. The purpose of the Pediatric Inventory for Parents Short Form (PIP-SF) was to assess difficulty and frequency of potentially stressful events of parents coping with the chronic illness of their child.[65] The original survey includes 42 events and typically requires 20-30 minutes for completion. Internal consistency was excellent with a Cronbach's alpha of 0.80-0.96.[65] To decrease participant burden, researchers developed a short form (PIP-SF) with 15-events which has been used for over a decade.[66] The survey lists 15 events, and parents are asked to rate frequency of each event on a five-point scale, ranging from 1 (never) to 5 (very often). The same events are then repeated, and parents are asked to rate difficulty of each event on a five-point scale ranging from 1 (not at all) to 5 (extremely). To complete this survey parents are asked to keep the previous week in mind and respond to each event twice. Internal consistency of the short form in this study was excellent with a Cronbach's alpha of 0.96, which was congruent with previous PIP-SF scores of 0.95[63] and 0.96[66] respectively.

Coping Measure. The 28-item Brief-Cope (B-Cope) was adapted from the COPE Inventory[64] and assessed coping responses to stress. The B-Cope focuses on understanding the frequency with which people use different coping strategies in response to various stressors. Its development was based on extensively used theoretical frameworks, including Carver and Scheier's Behavior and Self-Regulation model[67] and Lazarus and Folkman's Transactional Model of Stress and Coping.[8] Responses are rated on a 4-point integral scale ranging from 1 (I didn't do this at all) to 4 (I did this a lot). It includes 14 subscales for which psychometric properties are described. Three composite subscales measuring emotion-focused, problem-based and dysfunctional coping have demonstrated usefulness in clinical research.[64] Reliability of the measure for this study was acceptable with Cronbach's alpha of 0.90, higher than previous B-Cope scores of .68-.73.[68] [69]

Demographics

Participants completed questions about personal and child characteristics using the author-developed Demographic Data Form. Data collected on parental characteristics included parents' gender, age, marital status, education, income, and race. Characteristics of the child included age, gender, race, etiology of their underlying disease, time since diagnosis and insurance status.

Parent Interviews

Parent participants provided information describing self and child characteristics through a demographic survey. This PI conducted individual, semi-structured telephone interviews with participants using the parental interview guide (See Appendix A) developed in collaboration with a content expert and the Qualitative Research Core at Vanderbilt University (VU-QRC). The guide consisted of open-ended questions with probes intended to explore several aspects of caring for their CMC. Specifically, we examined the parent experiences, perceptions, and self-care habits of caring for their CMC

All telephone interviews were audio-recorded and transcribed verbatim using Rev.Com, an IRB-approved transcription service. The PI completed field notes to ensure accuracy of transcripts and note tone of voice or hesitancy that may not have been conveyed in the transcripts. Transcripts were assigned unique identification numbers and stored in password protected and encrypted electronic files.

Data Analysis

Quantitative

Data analysis for this study was conducted using IBM SPSS (Version 27.0). Descriptive statistical and graphical methods were used to describe sample characteristics and measure scores as well as to evaluate observed data distributions for appropriate analytic approaches. Frequency distributions summarized nominal and ordinal data; means and standard deviations were used for normally distributed continuous data, medians and inter-quartile ranges for skewed distributions. Associations of parental and child characteristics with the B-Cope scores were assessed using Spearman Correlations (continuous characteristics) and Mann-Whitney tests (ordinal or nominal characteristics). All tests of statistical significance maintained a Type I error rate of 0.05 ($p<.05$).

Qualitative

This PI completed coding and analysis of the qualitative data with the VU-QRC, using an inductive thematic approach as described by Jamshed.[70] The qualitative approach allowed critical themes to develop and be interpreted inherently by the data.[71] The initial coding system was based on the parental interview guide and refined after careful immersion in the data. Data were coded collaboratively with the PI and a second coder from the VU-QRC. Coders met to further discuss the coding system until agreement was reached. Management of transcripts, quotations, and codes was completed, using Microsoft Excel 2021 and SPSS version 27.0. Qualitative data were de-identified, but parents granted permission for quotes to be used in future manuscripts or publications.

A coding system used in this study was organized into seventeen categories. The categories included medical history/status, hospital-specific experiences, team interactions, experiences, attitudes and belief, navigating the health system experience, barriers and facilitators, emotion and cognition, life impacts, coping, settings of care, suggestion/needs, timeframe, diagnosis status, world impacts, changes over time, and noteworthy quotes. Subcategories were developed through the coding process for each of the main seventeen categories. One hundred and six codes were developed and were used to code 1900 quotes from parents (See Appendix B for the coding sheet).

Chapter 4

Results

Sample Characteristics Parents: Quantitative Survey

Fifty participants were approached and agreed to enroll in this study. Seven parents only completed the demographic survey and did not complete the other measures in this study (PIP-SF and B-Cope). Thus, 43 participants were included in the analysis sample. Demographic characteristics of the sample are summarized in Table 1. Parents were primarily mothers (n=31,72.1%) with a median of 41.8 years of age (IQR=31,48). The majority were white (n=35, 87.5%) and slightly more than half were married (n=23, 53.5%). Twenty-five (60%) reported earning an associate degree or higher. The majority of parents reported household incomes of the lowest or highest bracket (<$39,999: n=12, 32.4%; and >$80,000: n=15, 40.5%). Genders of the CMC included 27 girls (62.8%) and 16 boys (37.2%) with a median age of 8.8 years (IQR=5,15). The majority of the children with medical complexity were diagnosed either in utero or at birth with a median age of 0 months. Twenty-five parents (62.5%) had public insurance coverage, 88% of those having Medicaid, while 15 parents (37.5%) either had private insurance or were self-pay (those making payments out of pocket).

Table 1

Summaries of the Sample Demographic Characteristics (N=43)

	N	Median (IQR) Min,Max	
		Parent	**Child**
Age (years)	**43**	41.8 (34, 48) (19, 60)	8.8 (5, 15) (2, 22)
Time Since Diagnosis (Months)	**43**		0.0 (0.0, 2.3) -5, 139
		n (%)	
Gender	**43**		
Male		12 (27.9)	16 (37.2)
Female		31(72.1)	27 (62.8)
Ethnicity	**40**		
White		35 (87.5)	
Other		5(12.5)	
Parent Education	**42**		
Prior to High School Completion		1(2.4)	
High School or GED		16 (38.1)	
Associate Degree Completion		9 (21.4)	
Baccalaureate Completion		12 (28.6)	
Masters Completion		4 (9.5)	
Marital Status	**43**		
Single		8 (18.6)	
Married		23 (53.5)	
Divorced		4 (9.3)	
In a relationship		5 (11.6)	
Living with a partner		3(7.0)	
Household Income	**37**		
<$39,999		12(32.4)	
$40,000-$59,999		6(16.2)	
$60,000-$79,999		4(10.8)	
$80,000-$99,999		3(8.1)	
>$100,000		12(32.4)	
Type of Insurance	**40**		
Private/Self-Pay		15 (37.5)	
Public		25 (62.5)	
If, public Insurance*	**25**		
Medicaid		22(88.0)	
Medicare		2(8.0)	
Other		2(8.0)	
I don't know		1(4.0)	

*Check all that apply

Table 2 describes PIP Frequency scores and PIP Difficulty scores (Mean of 43.4 vs 36.6 respectively). This pattern indicates that parents reported both frequency and difficulty of their stressful events comparably.

Table 2

Summaries of the Pediatric Inventory for Parents (N=43)

Score	**Means (SD)**	**Median (IQR)**
Pediatric Inventory for Parents (PIP)		
PIP Frequency	43.4 (16.2)	44.0 (32.0, 57.0)
PIP Difficulty	36.6 (15.4)	38.0 (23.0, 49.0)

The B-Cope scores (Table 3) showed the use of dysfunctional coping, problem-based coping and emotion-focused.

Table 3

Summaries of the Brief-Cope (N=43)

Brief-Cope (B-Cope)	**Means (SD)**	**Median (IQR)**
Dysfunctional Coping	NA*	17.0 (13.0, 21.0)
Problem-Focused Coping	13.1 (4.2)	14.0 (11.0, 16.0)
Emotion-Focused Coping	22.0 (6.4)	22.0 (17.0, 28.0)

* The distribution was severely skewed.

Associations of demographic characteristics with the B-Cope scores are shown in Table 6. Caucasians reported statistically significant higher dysfunctional coping scores (median = 18 vs. 13 respectively, p=.036) than participants with other ethnic backgrounds. Furthermore, compared to participants who self-paid for health care or had private insurance, those with public insurance utilized statistically significantly higher dysfunctional and problem-based coping scores (Dysfunction: median = 18 vs. 13, p = .036; Problem-Based: median = 14 vs. 11, p = .025, see Table 4. Correlations of the Brief Cope with parental and child characteristics is shown in Table 5. While not achieving statistical significance, consistent with the pattern seen for insurance, the strongest correlation observed for the continuous or ordinal variables with the coping scores was the inverse relationship of income with the reported use of problem-based coping (r_s = -.30, p = .068).

Table 4

Summaries of Brief-Cope (B-Cope) Scores by Demographic Characteristics

		Dysfunctional Coping	**Problem-Based Coping**	**Emotion-Focused Coping**
	N	**Median (IQR) Min,Max**	**Median (IQR) Min,Max**	**Median (IQR) Min,Max**
Gender (child)		*p value=.216*	*p value=.733*	*p value=.344*
Male	16	*16(13,20)*	13(11,17)	*21(17,28)*
		12,34	6,24	*10,32*
Female	27	18(13,27)	14 (10,15)	25(18,29)
		12,35	6,17	12,30
Ethnicity		*p value=.036*	*p value=.984*	*p value=.226*
White	35	18(13,21)	13(10,16)	22(17,26)
		12,35	(6,18)	(10,32)
Other	5	13(12,15)	14(11,16)	29(18,30)
		12,16	17,30	9,17
Marital Status		*p value=.364*	*p value= .077*	*p value=.238*
Single	8	18(13,21)	15.5(11,18)	24(17,30)
		12,34	9,24	17,31
Married	23	15(13,19)	12(7,15)	22(14,26)
		12,30	6,17	10,32
Divorced	4	18(16,26)	18(13,18)	29(23,30)
		16,27	12,18	22,30
In a relationship	8	20(14,27)	14(12,16)	22(17,24)
		12,35	6,18	10,29
Type of Insurance		*p value=.036*	*p value=.025*	*p value=.075*
Private/Self-Pay	**15**	13(12,19)	11(7,15)	18(12,26)
		12,30	6,17	10,30
Public	25	18 (14,21)	14(12,17)	24(18,29)
		12,35	6,18	10,32

Table 5

Correlations of Parental and Child Characteristics with Brief Cope (N=43)

	Coping (B-Cope)		
Parental and Child Characteristics	**Dysfunctional Coping**	**Problem-Based**	**Emotion-Focused**
Child Age	.02 (.891)	-.06 (.686)	-.11 (.497)
Time Since Diagnosis	.02 (.887)	-.13 (.405)	.04 (.793)
Parent Age	-.01 (.943)	-.20 (.215)	-.08 (.633)
Educational Level	.09 (.569)	.24 (.130)	.14 (.367)
Yearly Income	-.19 (.249)	-.30 (.068)	-.05 (.784)

~Note: Values in cells are r_s (p-value), *p<.05 **P<.01
~Higher scores indicate more dysfunctional coping, problem-based coping or emotion-focused coping.

Sample Characteristics of Parents: Qualitative Survey

Fifteen of the 43 (38%) participants completed the qualitative interviews. Demographic characteristics are summarized in Table 3. Parents were primarily mothers (n=11, 73%) with a median of 41.8 years of age. Most parents were white (n=13, 86%), married (n=8, 53.5%), and had earned an associate degree or higher (n=9; 60%). The household income of participants varied, with several falling either in the lowest income bracket or the highest (<$39,999: n=7, 47%; and >$80,000: n=7,47%). Child gender included 5 girls (33%) and 10 boys (66%) with a median age of 8.8 years. Eighty-seven percent of the CMC were diagnosed either in utero or at birth with a median age of 0 months. (Table 6).

Table 6

Summaries of the Interview Sample Demographic Characteristics (N=15)

	N	**Median (IQR) Min, Max**	
		Parent	**Child**
Age (years)	**15**	41.8 (34, 48) (19, 60)	8.8 (5, 15) (2, 22)
Time Since Diagnosis (Months)	**15**		0.0 (0.0, 2.3) -5, 139
		n (%)	
Gender			
Male		4 (27)	10 (67)
Female		11 (73)	5 (33)
Ethnicity			
White		14 (93)	13 (87)
Other		1 (7)	2(13)
Parent Education			
Prior to High School Completion		3 (20)	
High School or GED		7 (47)	
Associate Degree Completion		1 (6)	
Baccalaureate Completion		4 (27)	
Marital Status			
Single		3 (20)	
Married		8 (54)	
Divorced		2 (13)	
Living with a partner		2 (13)	
Household Income			
<$39,999		7 (47)	
$40,000-$59,999		1 (6)	
$60,000-$79,999		0	
$80,000-$99,999		4 (27)	
>$100,000		3 (20)	

Table 7

Major Themes with Sub-Categories Illustrating Impact on Parents Caring for CMC

Major Themes	**Sub-categories**
Navigating the health care system experiences and interactions	Perceived health care system capabilities Perceived health care system inadequacies Staff Interactions Lack of Resources Wish I would've known
Emotion and Cognition	Expectations Desire for Normalcy Hope Social Isolation Guilt
Coping Strategies	Problem-Focused Organization Sense of Control Advocate Emotion-focused Religious/Spiritual Self-Care Reframing Purpose Resilience Acceptance

Qualitative Results

All parents described a high burden of caregiving related to their child's chronic medical complexity. Three major themes emerged from the data: 1) navigating health care system experiences and interactions, 2) emotions and cognition and, 3) coping strategies. They were each divided into subcategories as illustrated in Table 7. Each theme will be presented using exemplar quotes.

Theme 1. Navigating health care system experiences and interactions

The first theme refers to participants' discussions of their perceptions as they traveled through the health care system with their children. Each parent verbalized both positive and negative aspects of the health care system, and these comments were further categorized into capabilities of the health care system, staff interactions, inadequacies of the health care system, lack of resources, and things they wished they would have known along their child's medical journey.

1.1 Perceived Healthcare System Capabilities. Overall perceptions of the healthcare system that were positive appeared to enable participants to deal with their child's illness more effectively. One mother of a 2-year-old boy said, "I think just having the medical team we had definitely helped me to cope with everything. They also helped me if there were decisions to be made or we needed opinions…just having somebody to talk to was so helpful." Another mother of a 12-month-old boy verbalized appreciation for the level of education they were given prior to their discharge: "They didn't let us fall through the cracks on anything. We felt like we were taught everything we needed to know before we left."

1.2 Perceived Healthcare System Inadequacies. Parents described difficulty recovering from some instances that occurred during in-patient stays, citing extreme disappointment and an overall disregard for the parents' needs during a tumultuous time. One mother of a 6-year-old boy tearfully shared, "Yeah, they didn't care about my feelings and my wants and needs, and some of them, their lack of knowledge, lack of experience, I felt really damaged my son. He had a lot of problems, but they were only problems because of things that those nurses and doctors did to him. Like, they messed up. They would give him the wrong medicine or they... He has just had brain surgery and they ripped the pillow out from under him. The RT was not able to intubate him, and that caused a lot of scar tissue, things like that, that actually damaged him." A mother of a 22-month-old girl spoke to the consistency that a parent gets used to when being on a unit for an extended period. Specifically she said, "It was in the NICU, and then when she was I think maybe eight or nine months old, they transferred her to the intermediate care unit, and that was very difficult as well because it was hard to go from a unit where they followed her and the nurse was there checking, doing vitals every hour to a unit where they would only check her vitals like twice a day. That was really hard for me to go from that. A father of a 6-year-old girl shared, "I just felt like a child with all of my daughter's complex needs, it'd be nice if there was a physician that kind of jumped in there and helped guide as well. Because we just kind of had to learn everything on our own. And yeah. So that would have been nice. And we did, I mean, we found out about, and I forget what it's called now, like a care conference, do you know what I'm talking about?"

1.3 Staff Interactions. The parents who felt like they had great care providers for their children readily pointed out the good in the interactions. One father of an 8-year-old-boy stated, "There is a great person in Complex Care, who can answer any question I have so I don't hesitate to call her." A mother of an 11-year-old boy said, "I can't ask for better people than the care team in complex care and his pediatrician." Other staff interactions affected parents negatively, such as this mother of a 22-month-old girl stating, "And we didn't get along with the nurses on that unit. Yeah. I didn't get along with a lot of the nurses, but that's just the way it goes, but the ones I didn't get along with didn't know how to do their job."

1.4 Lack of Resources. Several parents spoke freely of the inability to maintain employment of their in-home staff and what a huge loss the aides and nurses were to all the members of the family. A mother of a 15-year-old girl stated, "You are constantly losing your home health nurses, but they become a member of your family… even to my typical daughter. I kind of equate it to a lot of men coming in and out of her life.” One of the fathers of an 8-year-old boy shared, “A huge missing piece that I have found is someone to help you navigate all the systems. Like someone who shared about navigating systems of support, like financial assistance, social security, early intervention, all those things. Things still come up I have never heard of, if staff didn’t have time to do it what about another parent who has gone through these things and come out on the other side?”

1.5 Wish I would’ve known. Several parents reported concerns or issues they wished they would have known. These comments varied from parents depending on their medical experiences. Parents voiced wishing health care providers fully educated them on their CMC’s diagnosis and thoughts for prognosis. The parents reporting hoping for more complete and comprehensive home medical care instruction prior to their child’s discharge and making sure they would make sure they have everything they could potentially need at home. Lastly, they voiced wishes of reciprocal communication during their child’s time of diagnosis. Parents voiced wishing they could have told physicians how they made them and their children feel without compromising their care. One mother of a 4-year-old boy stated:

> When my son got his wheelchair, like I said before, no one said make sure to get a wheelchair accessible van. I kind of wish we knew that before. I mean, I know that sounds so dumb... But you don't think about it and so you feel like, I kind of wish like someone said You should think about getting a van for when he is in a big wheelchair. Because I never thought about that, he was in a stroller…Just little things like that but a lot of things me and my husband just kind of roll with it and we figure it out and we make it happen somehow, but might take us a little while, but we'll get it. So, it's just the little things.

A mother of an 11-year-old boy shared, “I feel like I wish the physicians wouldn’t have been so matter of fact like this is what you are going to deal with. I know they think they know but they didn’t. And if they would try to show you there could be a light at the end of the tunnel instead of letting you feel like it could only be dark … I know it sounds weird, but they treated us kind of like we were invisible even when we were no different than parents of normal kids, but like we didn’t deserve their time because our kid couldn't be fixed in their minds."

Theme 2. Emotion and Cognition

The second theme was described as responses to things parents dealt with daily. Subcategories included expectations, desire for normalcy, hope, social isolation, and guilt.

2.1 Expectations. Parents reported not having appropriate expectations. Understanding that some illnesses were progressive, parents searched for medications to slow this process. Parents looked for specialists to help prevent recurrent infections. They looked for avenues to help their children communicate. They verbalized wanting to provide their children with the happiest, most normal lives. One mother of a 15-year-old girl explained:

> Supposedly, they've seen a little bit of gain in the hands use. It says that when you have Rett, the dendrites in the brain, when they go to send messages, they described it if you put your hands with your fingers stretched out towards each other and your fingers are the dendrites, they communicate with each other but if you have Rett syndrome, you try to put your hand into a fist. Your dendrites have curled in. They can't communicate very well. Supposedly this trofinetide, which is actually insulin growth factor, IGF one is the post to regrow those dendrites.

Parents reported feeling like their expectations were not the outcome they hoped, and they viewed unmet expectations as another loss. They were always quick to clarify how much they loved their children, but life would never be what they expected for their families. One mother of a 17-year-old boy reflected on the differences with "normal families" and how their marriage would be impacted as well. She stated, "Yep. I mean he's amazing, but he has a lot of issues and will never be ok to work or live on his own. So that's a lot when we think about it not that we wouldn't want him forever. It's just out of the norm of what you think your kids growing up and moving on and then having that time with your husband. I'm sorry that sounded selfish. You know what I mean."

2.2 Desire for Normalcy. Parents reported a desire for social normalcy. They described wanting to have a life like those around them, being out in the world openly without fear of further illness, or the impact their child may have on other people. A mother of a 15-year-old girl reported, "It's like people are afraid of children with chronic illness. Which I will never understand. Don't they deserve more care and compassion?" A father of a 6-year-old son said "We just make sure we include her in things. If we are watching TV we make sure he is up on the couch with us." Parents also stated despite the limitations of their children, they wanted them to have the same opportunities as other children. A mother of a 15-year-old girl stated, "We want her to be able to enjoy the ocean and the beach just like everyone else." A mother of a 5-year-old girl felt similarly. She stated, "She's in preschool and goes to the sitters in the afternoon after the school bus drops her off…just normal kid stuff home with us on the weekends."

2.3 Hope. Some parents shared that their situations did not lead them to a place of despair, but instead being hopeful for things in the future. One mother of a 6-year-old boy stated, "I don't want to make it sound all sad… there is so much joy and hope too. He's an amazing little guy." A mother of a 22-month-old boy shared, "My everyday life has changed but by my doing. I wrote a children's book to bring awareness to his condition and that has opened so many amazing opportunities. I'm so hopeful for the future!"

2.4 Social Isolation. The parents spoke repeatedly about how their focus is always taking care of their child. They verbalized not having meaningful relationships with others when they cannot empathize with their situations. One mother of a 4-year-old boy stated, "I think the thing that hurt the most was when he got his trach. That was a kind of turning point…. They took away his voice I never got to hear his voice. And no one else could know what that was like." Parents frequently reported on the social isolation their CMC felt from their peers. Parents further explained that it is just as important for the children to belong as it is for the parents to feel like they are a part of something as well. They felt often overlooked by others. A mother of a 15-year-old girl explained, "I think she has been invited to one special needs birthday party in her whole life…but guess what she was the only one who couldn't get in and out of the water by herself…It's awkward so you feel stupid and weird…like don't worry about me I will be over here in the pool while you guys are relaxing and talking."

2.5 Guilt. Parents often portrayed the feeling of guilt. Parents discussed wondering if there was anything they did to contribute to their child's medical diagnosis. They also verbalized guilt for feelings of frustration towards their child. Parents specifically described instances when they put in a great deal of effort preparing their child for routine daily activities. Parents verbalized being afraid something could happen to their child, and not being there for them. One mother of a 13-year-old girl said, “I can’t leave her with an aide all the time…you want to take her and include her but sometimes it backfires…you feel like you are damned if you do and damned if you don’t…Another mother of a 15-year-old girl stated, I would feel guilty all the time and feel terrible even though it is such a strain for us to take her places.” A mother of a two-year-old son voiced the difficulty of being away. She explained, “When it is your child, you want to be there. If anything happened, I wanted to be there. I wanted to be the one to take care of him.” A mother of a 15-year-old girl discussed how as parents they needed to be prepared in all aspects of life. She stated, “I don’t know if she will outlive us. My husband and I had to have a plan/trust set up in the event we die first…what will happen to her.”

Theme 3. Coping strategies

The third category that emerged reflected parents reported coping strategies. Overall, there were five coping strategies reported by parents including avoidance and anticipation of the future, but the three most common strategies were problem-focused, emotion-focused and reframing.

3.1 Problem-Focused.

Problem-focused coping can be defined as taking control of the stress, seeking assistance in the situation, or removing oneself from the stressful situation. Many parents discussed the need for their life to be organized in every aspect and not just in terms of their child’s medical care.[22]

3.1.1 Organization. The idea of organization was a frequent comment. Parents spoke about the need to have a system of organization and how it would make life slightly less difficult. One mother of a 15-year-old-girl stated, “I have to plan everything. There is a lot of planning all the time… I feel like I have a very tight schedule. It’s a lot sometimes.” A mother of a 13-year-old girl stated, “Vacation took so much planning and sending supplies ahead…it was exhausting… but I never know when she will take her last breath, so I want her to have the experiences.”

3.1.2 Sense of control. Some parents felt they often lacked control of their situation. Examples of loss of control included declines in their own health and their child’s health, emotions they could not control as things always “unexpectedly” came up, or traumatic episodes such as PTSD. One mother of a 6-year-old son shared, “I have had trouble accepting it and moving on from the initial trauma of it. It’s hard for me to give up control and hard to cope with the drastic difference in my life before my son and now.” A mother of a 4-year-old son stated, “It's still just doctor appointment after doctor appointment for him and for me. My health has gone downhill quite a bit because I just am overstressed, overwhelmed with everything, so physical health and mental health is just downhill. If it's not an appointment for me, it's an appointment for him. There's little time for fun or leisure.”

3.1.3 Advocate. Parents verbalized the need to be their child’s advocate. The parents felt without their constant presence, bad things could happen to their children. They worried about things like the safety of their daughters having male providers or their daughters being near men at school. They reported even leaving their children in the care of medical providers was anxiety- provoking. The parents were left feeling no one could provide care for their child as well as they could. One mother of a 6-year-old boy voiced her concern, “…And he can’t communicate very well, so we are his voice. We know him so well we can read his facial expressions and … make sure he is comfortable. A mother of a 13-year-old girl stated, “Most parents don’t have to think about the fact that as a parent of a daughter who is non-verbal who is now developing into a woman would have to be concerned about getting shots to prevent STDs, but that’s my life… what if I leave her and something happens to her? I could never forgive myself.”

4. 1 Emotion-Focused

Emotion-focused coping can be described as escape and avoidance strategies.[8] Many parents stated their belief in God was strong despite their previous traumas.

4.1.1 Religious. Many parents spoke of their spiritual connection with a greater being. One mother of a 1-year-old son shared “…Honestly God almighty is my biggest support. When you have that kind of relationship and know he is in control, I could do nothing about the outcomes of my son.” Some participants reported that their prayers helped them cope with the difficulties they experienced. A mother of a 6-year-old daughter stated, “Faith has helped me have the sense of acceptance for what life is rather than fighting it.”

4.1.2 Self-Care. Parents take care of themselves in various ways. Many of the parents stated they practiced good self-care. Parents explained their primary reason for taking care of themselves was for their child. They reported activities such as working out and eating right. They prioritized self-care to carry their CMC up the steps or require less sleep to stay up longer with their child at night. They spoke of needing to maintain their overall health as much as possible when coupled with their other obligations. One mother of a 15-year-old girl stated, "I probably take care of myself more now because I know she is going to need me forever."

A mother of a 2-year-old boy shared, "It seems like I do more self-care now than when I had him…. A lot of special needs parents get lost in their children and I am not going to do that."

5.1 Reframing

All parents shared positives of their life in caring for their CMC. All parents expressed wishes for their children to be healthy, but also voiced appreciation for who their CMC had become, and the lessons they had learned from them.

5.1.1 Purpose. Many parents verbalized what a sense of purpose their child provided for them, from making them better people, helping them realize their own dreams and opening doors to new opportunities. A mother of a 15-year-old girl said, "I think she has shaped me into the person that I am…I feel like not everybody is a caretaker and natural caregiver…There are things I have done that I never would have done prior to having her." One mother of a 7-year-old-boy shared, "Honestly, I don't know, for me personally, I have achieved so much more since I've had him. I've changed as a person and I've grown as a person in very positive ways. I feel like before, I wouldn't say I was a bad person, but I don't know if that makes sense. He kind of pushed me, I'm going to college, I'm getting a degree. So I'd say he's made me a better person. I don't think I was a bad person before but I don't know, he's just kind of pushed me to achieve a lot, I wanted to do it for him, but for myself too." A mother of a 2-year-old boy stated, "I can honestly say since my son was born it has been 10 times better with him…Having him has changed everything…I have a reason to be…"

5.1.2 Resilience. The resilience these parents exhibited in their descriptions of caregiving was apparent, but difficult for them to acknowledge as a personal quality. A mother of a 15-year-old girl denied doing anything differently than what anyone else would do. She stated, “I think that I do what anybody would do in the same situation…it could happen to anybody. She’s the hero, I’m not.” Similarly, a mother of a 2-year-old son stated, “I think that when I am put under pressure, I don’t fold…it can be overwhelming…just being strong for him and putting him first is my number one priority.” Only one mother of a six-year-old boy stated being resilient as a reason she could provide constant care for her CMC when she said, “I think I am a pretty resilient person anyway dealing with past trauma and stuff in my life has set me up to be stronger.”

5.1.3 Acceptance. Parents of CMC stated that their personal acceptance helped make caring for their CMC less difficult…A mother of a 6-year-old son reported, “I think what makes it a little bit easier is how happy my son is.” “I think it may be different for kids who knew a different life. This is the only life my son has ever known and …he’s happy.” A mother of a 4-year-old girl said “what your every-day attitude is to having a child with a complex medical condition, you can either feel sorry for yourself or put one foot in front of the other and make the best of it.” The mother of a 2-year-old son spoke candidly as she ended her story “…I feel like he is a blessing. He belongs in this family. He chose this family.”

Chapter 5

Summary of Findings, Implications, and Directions for Future Research

The purpose of this study was to examine stress and coping in parents of children with medical complexity (CMC). This study investigated perceptions of frequency and difficulty of stressful events experienced by parents in caring for CMC. The study also explored relationships between parental and child characteristics and coping strategies reported by parents. The impact on parents of caring for their children with medical complexity was also addressed. This study differs from previous studies because it examined associations of several variables concurrently. Stress and coping were operationalized with two instruments in the survey portion of the study, with further depth and meaning added with the qualitative data. This chapter will summarize key findings based on specific aims and strengths and limitations identified in the study. Lastly, implications and directions for future research will be discussed.

Aim 1: Frequency and Difficulty of Parental Stressors

The first aim of this study was to describe the frequency and difficulty of parental stress when caring for children with medical complexity. The Pediatric Inventory for Parents-Short Form (PIP-SF)[63] was used to assess parental perceptions of stressful events. The PIP-SF [63] presents the parents with fifteen potentially stressful situations and describes parental perceptions of the frequency and difficulty of each situation. Our study demonstrated that parents of CMC perceive both frequent and difficult stressors when caring for their child. These results are consistent with previous studies describing stressors in parents of CMC being higher than that of parents of healthy children and will be described below.[72, 73]

Parents of CMC are often the sole medical provider in the home.[46] Parents of CMC have previously reported constant fear that their child may get sick or die.[36] Uncertainty about their child’s future was also identified as a major concern.[74] Parents of CMC have been shown to often neglect their own physical, mental and emotional well-being.[31] This was congruent with our PIP-Difficulty (PIP-D) findings, demonstrating a need to address these factors in parents of CMC. In a study by Bray et al., peer-to-peer support groups decreased parents' level of psychological distress and had a positive influence on their ability to cope with having a child with a disability.[75] Support groups may be an important intervention to consider with this population.

Aim 2: **Coping Strategies**

The second aim of this study was to describe types of coping strategies (dysfunctional, problem-based, and emotion-focused) used by parents of children with medical complexity. The Brief-Cope (B-Cope)[64] was used to assess coping strategies. The B-Cope[64] presents the participant with 28 items to measure effective and ineffective ways to cope with a stressful life event.

Parents used a variety of coping strategies to deal with the impact of caring for their CMC which included emotion-focused, problem-focused and dysfunctional coping. According to Lazarus and Folkman, the use of emotion-focused and problem-focused coping are ways to reevaluate the stressor and may potentially be helpful coping mechanisms.[8] However, problem-focused coping is usually associated with a more positive outcome, while emotion-focused coping is associated with a more negative outcome.[76]

A systematic review found that problem-focused coping strategies improved caregiving well-being.[73] Emotion-focused coping strategies in parents of children with epilepsy or cerebral palsy were shown to have a negative effect on their overall quality of life.[73] Dysfunctional coping includes mechanisms such as avoidance and denial that were identified in parents of traumatic brain injury patients.[77] The results of our study indicated that the use of problem-focused coping was reported the most often, including strategies such as planning, use of social support and active coping.

Aim 3: Parental and Child Characteristics

The third aim of this study was to describe associations among children's characteristics which were gender, age, race underlying diagnosis and date of diagnosis concurrently with parent characteristics which were gender, race, age, income, marital status, level of education, insurance and types of coping strategies (dysfunctional, problem-based and emotion focused) used by parents of children with medical complexity.

Although statistical significance was not achieved, associations indicated that there was a positive correlation between educational level and problem-based coping. The more advanced level of education of the parent was associated with increased reporting of problem-based coping. The parents who were younger in age and had a lower income were also more likely to use problem-based coping but cause and effect was not established in the study. The possibility that younger parents with lower incomes have not cared for their CMC as long as older parents may suggest time since diagnosis could lead to development of poor coping strategies.

Associations among parental characteristics such as gender, age, income, education, and marital status did not show which coping strategies were more effective in dealing with stressors. In our study, survey results were associated with higher dysfunctional coping. This finding is not generalizable though, with the lack of ethnically diverse subjects. Participants with public insurance also utilized more dysfunctional coping compared to those with private insurance or who were self-pay. Interpretation of this finding is difficult with such a small sample size, but previous research has demonstrated that having a child with chronic medical conditions may affect the family financially, emotionally, and physically.[10, 78, 79] It is unknown whether this is a causal relationship, but a child's health may mandate parents to stay at home, in turn decreasing income, increasing social isolation, and making it more difficult for parents to focus on their own self-care.

Aim 4: **Impact of caring for CMC**

The fourth aim of the study was to examine the impact of caring for children with medical complexity on parents through semi-structured interviews. The findings from our qualitative content analysis described experiences of the parents of CMC as they provided insight into their often-hectic daily lives with great detail. Parents communicated the barriers and facilitators they encountered in their children's health care experiences and their feelings of guilt and social isolation from family and friends. Parents were also able to verbalize feeling hope, happiness, and purpose in their lives. Parents emphasized that their children were always their focus, leading to their own lack of self-care or to the contrary, taking better care of themselves. They stated, "I need to live longer for my child, because who will take care of them when I am gone?"

Parents reported being thankful for their healthcare providers but wishing for different outcomes as their child's trajectories progressed. Research shows that the medical care these parents provide in the home can be overwhelming and their quality of life was reported as poor when compared with parents of healthy children.[26] Yet, they could appreciate and verbalize that no one could take care of their child more appropriately than they could in their own home. They shared positive aspects of their health care team but continued to describe a lack of resources.

Parents explained what a day in their life was like. The parents reported their lives as being hurried and often having feelings of guilt for being unable to spend time with other siblings. They identified having to plan even minor details of their day, to ensure proper care was provided to their CMC. Several parents verbalized concerns of social isolation. Their focus was taking care of their child, with little time or energy to give to anything else. This is similar to previous literature in which parents of CMC have been described as not typically seeking emotional support from others.[80] They less frequently look for friendliness, sympathy, or understanding from other parents of healthy children, citing others' inability to understand what their life looks like.[81] Future research should document parental input about perceptions of community services to better understand how we can identify and support parents who may be at risk for social isolation. Parents reported their lives being fulfilled caring for their CMC. Many parents described the optimism they felt despite the current adversities in caring for their CMC. Research shows that hope has been reported to affect parents' adaptations to their children's chronic conditions.[82]

Summary of Results

Parents cited medical treatments and fear their child might die as their most frequent stressors. The most difficult stressful events reported by parents were trying to find time for their own needs and seeing their child afraid and sad. An event cited as both very frequent and very difficult was the uncertainty they feel for their CMC's future. Qualitative data analysis further validated this finding with a common theme described by parents as being continually or constantly concerned for their children, despite location or what the parents were doing at that time.

Results of this study indicated that problem-based coping is often used in parents of CMC when specific demographic characteristics are present. While not reaching statistical significance, results suggested positive correlations between educational level and problem-based coping. Furthermore, statistically significantly higher problem-based scores were observed for the group with public insurance (suggesting lower income) than those with private/self-pay health care coverage and an inverse association between income and problem-based coping scores was observed. These results are consistent with a study by Akturk and Aylaz that described associations between parents' age, annual income, and coping strategies.[80]

Problem-based coping was used more frequently than other types of coping in younger parents and parents with lower annual incomes in Toledano-Toledano's research.[47] The child's age and time since diagnosis were not evaluated in Akturk and Aylaz'[44] or Toledano-Toledano's study,[45] thus could not be compared to our findings. Examples of problem-based coping include planning, use of social support and active coping. Interestingly, the findings of our study did not indicate maladaptive coping strategies, such as alcohol or drug use, and no signs of overall avoidance of their situations.

Strengths and Limitations

This study has many strengths but must be considered in the context of some limitations. We acknowledge that the small sample size and cross-sectional design are limitations. There is also a risk of selection bias in using a convenience sample. Many of the participants (N=43) were female (N=31), and the study sample included mainly Caucasian participants (N=35). This decreases the generalizability of the findings across populations. Over half of the study population was married (N=23): Consequently, the results may not be generalizable to single parents.

In the qualitative portion of the study, it must be acknowledged that parents may have found discussing their child's medical complexity to be difficult. Recruitment may have been skewed to those who were interested in sharing their experiences. Those who did not agree to participate may have been the most affected by their child's illness. Participants may have responded to self-report survey questions in the way they thought the researcher expected them to answer, leading to response bias. Patients may have delayed seeking care during the pandemic. Parents of children with more severe chronic illnesses may have felt a greater need to participate in a study that could potentially benefit them or others. Therefore, results are not generalizable to all parents of CMC.

Because of the study's cross-sectional research design, differences that may be present at varying ages of the CMC or time from diagnosis cannot all be captured in the parents' report of their experiences. Competing obligations could have affected the participants' responses. The study did not include a comparative group of parents with healthy children. Despite these limitations, this compelling research fills a gap in the literature and serves to inform feasibility of conducting research with this vulnerable population and preliminary understanding of stress and coping as perceived by parents of CMC.

Clinical Implications

Clinicians can be the main facilitators between assessing the needs of parents of CMC and helping aid in their adaptation to their child's illness. In developing an awareness of parents who may be at a greater risk for poor coping, healthcare professionals and support staff may find ways to increase guidance and supportive interventions earlier in their CMC's illness trajectory. Based on parental reports in interviews conducted as part of this study, opportunities for helpful interventions include making recommendations for the most effective modes of transportation for their CMC and identifying helpful financial resources. Parents also sought timely education about potential changes in children's needs, and earlier introductions to support groups.

Nurses developing reciprocal relationships with parents can help educate and prepare parents for caring for their CMC at home. Nurses who have an on-going relationship with parents can consistently provide guidance to promote optimal flexibility and normalcy in everyday life. Physicians can facilitate the development of mutual, respectful relationships with parents of CMC.

At a health care systems level, the addition of multi-disciplinary teams to the complex care clinics could facilitate collaboration and improve holistic, comprehensive care of families. Development of a complex care tool kit for providers could ensure adequate assessments of parents' coping strategies at the time of their child's diagnosis and throughout their child's lifespan. Establishment and initiation of institutional guidelines and standards of care for CMC could guarantee appropriate education and resources are provided for each child despite their diagnosis.

Recommendations for Future Research

Understanding gained from this study exploring how both parental and child characteristics may affect parental responses to the frequency and difficulty of stressful events and which coping strategies are utilized will contribute to the sparse information currently available. Further studies are needed to advance the understanding of how parental and child characteristics may affect the use of coping strategies so that appropriate interventions can be developed for parents of children with medical complexity. Studies describe how parents of children with medical complexity face higher levels of stress than parents of healthy children.[12] Along with the daily demands of parenting, they must understand and manage their child's complex condition, make medical decisions on their behalf, serve as their advocate and the coordinator of care both at home and in medical settings. These issues exist in children with diverse medical complexity, despite their individual diagnosis.

Future work could potentially involve replication of this study, using a longitudinal research design with multiple sites, including a larger sample size, higher percentage of male participants and a more ethnically and culturally diverse population. Additional research should expand to focus on coping strategies used by parents and how they impact the perception of stressful events related to caring for their CMC. Descriptions of parental personality characteristics in addition to parental sociodemographic variables may also add to the understanding of this complex parent and child population. With this foundational understanding, studies may identify beneficial ways to support these vulnerable parents.

Conclusions

Important insight into the experiences of parents of CMC was gained through this research. Results reveal the need for health care providers to assess parents' coping strategies throughout their child's illness trajectory. The parents in this study described both the positive and negative aspects of having a child with CMC. Previous studies have shown that parents of CMC are at higher risk than parents of healthy children in developing compromised emotional,[83] mental,[84] and physical health.[85]

This study identified the reality that many parents of CMC are faced with similar adversities. The need to provide health care for their CMC while fulfilling their parental role was similar across numerous diagnoses. Previous literature has shown the use of problem-based coping strategies to decrease parental stress of CMC.[10, 35, 47] Evaluation of frequency and difficulty of stressful events in parents of CMC, and further understanding of the coping strategies used is crucial to determine the most effective interventions to meet the needs of these families of CMC.

In conclusion, this study was one step to further examine parents' perceptions of experiences related to their children's complex conditions. How well parents cope with their CMC depends on numerous factors, including the parents' perception of the stresses in their lives, and the coping strategies reported. A major finding of this study is that regardless of the child's diagnosis, providers' careful examination of parental coping strategies may have major implications for future work to facilitate beneficial interventions to enhance the overall well-being of the parents. The findings of this study support the need for development of standards of care to assess and address the needs of parents of CMC no matter their child's complex medical diagnosis. In addition, findings illustrate the need for timely referral of parents to individualized and group counseling efforts to strengthen appropriate coping strategies and improve outcomes. Provider awareness and assessment of perceptions of these parents will provide a foundation for understanding beneficial interventions for these vulnerable families.

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Appendix A: Parental Interview Guide

I'd like to start by asking you what your initial thoughts and feelings are after completing the surveys?

> Could you share with me what your life looked like before caring for your child with medical conditions?

- Specifically, what did your everyday life look prior to this?
- What does everyday life look like for (his/her name)?
- Prior to caring for your child with medical complexities, what was your biggest support to help you cope in difficult situations?
- What did you do for self-care before caring for your child with medical complexities?
- Are there any personal characteristics you think have made this any easier for you to deal with? Any more difficult?
- Are there any life events you think have made this any easier for you to deal with? Any more difficult?

Thank you for sharing these thoughts with me. The care of a child with medical complexities is hard to imagine for many parents. We know any medical condition is not something that a parent wants to see their child go through. Many parents have told us that they didn't think they would ever be able to deal with something like this, but some things have helped them along the way. What was hearing about your child's potential or actual medical condition like?

- What does your everyday life look like as your child continues to grow?
- What does everyday life look like for (his/her name) as he/she continues to grow?
- What has been your biggest support since learning of your child's medical condition?
- What do you do for self-care as you care for your child with medical conditions?

Thank you for sharing your experiences with me. With your help, I hope we can support you and other parents going through similar experiences.

Appendix B: Coding Scheme

Code	Label	Definition
1	**Medical History/Status**	**Discussion centers on child's medical history or current medical status**
1.1	Symptoms	Any discussion about child's symptoms
1.1.1	Non motor	Non-motor symptoms such as weight, GI, pain, etc.
1.1.2	Motor/physical function	Discussion centers on child's motor/physical function
1.3	Hospitalization	Hospitalization status (e.g readmit)
1.4	Sequelae	Child experienced new symptoms or complications
1.5	Medication/treatment	Any discussion of child's medication and treatment
1.6	Characterization of treatment	Discussion centers characterization of child's treatment
1.6.1	Impact on health	Impacts of medication/treatment on child's health, symptoms, etc.
1.6.2	Medical equipment	Characterization of equipment or devices needed for home care
1.7	Family history	Any mention of family history
1.8	Other	Other medical history/status not listed above
2	**Hospital-specific experiences**	**Discussion centers on experiences in the hospital**
2.1	Care decisions	Discussion centers on shared decision making, parent preferences, avoiding medical jargon, etc.
2.2	Family perspective	Discussion centers on health team interest in family experience and perspective
2.3	Honesty/transparency	Discussion centers on honesty and transparency of staff
2.4	Assessments	Discusses any assessments that occur during hospitalization
2.5	Education	Discussion centers on education on child's disease, devices, medications, etc.
2.6	Family accommodations	Discussion centers on accommodations provided to family (e.g., Ronald McDonald House)
2.7	Communication	Discussion centers on communication with health team
2.8	Continuity/consistency	Discussion centers on consistency of specific staff interaction
2.9	Family meetings	Discussion centers on family meetings with providers, nurses, and fellows
2.10	Other	Other NICU experiences not listed above
3	**Team Interactions**	**Discussion centers on interactions with specific team members or others**
3.1	Provider/hospital interaction	Parent provides experience of team member interaction
3.1.1	Positive interactions	Describes a positive experience with team members
3.1.2	Negative interactions	Describes a negative experience with team members
3.1.3	Lack of interaction	Parent expresses a lack of interaction with team members
3.2	Type of interaction	Specific team member interaction
3.2.1	Nurse	Interaction with nurses
3.2.2	Physician	Interaction with physician
3.2.3	Home health/respite	Interaction with home health or respite
3.2.4	Other	Interaction with other team not listed above
3.3	Professional presentation	Describes team member demeanor and nonverbal communication. Presenting oneself as a professional.
3.4	Turnover	Turnover of caregivers / team members

4	**Experiences**	
4.1	Education topic	Discussion centers on type and topic
4.2	Method	Education method
4.3	Advocate for self	advocate for child (e.g. not taken seriously)
4.4	Wish had known	Discusses what s/he wished would have known or asked before discharge
4.5	Other	Other discharge preparation experience not listed above
5	**Attitudes and Beliefs**	**Attitudes and beliefs expressed by the caregiver**
5.1	Self perception	Discussion centers on caregiver's view on her/his role in a given
5.2	Health team capabilities	Discussion centers on caregiver's view on health team capabilities
5.2.1	Sufficient	Sufficient capabilities/knowledge
5.2.2	Insufficient	Insufficient capabilities/knowledge
5.3	Causal attributions	Parent provides an explanation of a given outcome related to child's
5.4	Sense of control	Discussion centers on the degree that the parent believes that s/he has control over her/his child's life and health outcomes
5.5	Stigma	beliefs of others
5.6	Social Comparison	Compares family to that of others in same situation or different situation
6	**Navigating health system experience**	**Discussion centers on caregiver experience in navigating the health system**
6.1	Resources	Experiences related to access to resources
6.2	Treatments	Experiences related to treatment (e.g. amount of time etc).
6.3	Appointments	Experiences with follow-up appointments
7	**Barriers and Facilitators**	**Discussion centers on barriers and facilitators to healthcare, outcomes, access, etc.**
7.1	Barriers/challenges	Discussion centers on barriers, challenges, and deficits related to outcomes
7.2	Facilitators	Discussion centers on facilitators/went well
8	**Emotion and Cognition**	**Discussion centers on emotional and cognitive experiences**
8.1	Anger/frustration	Caregiver discusses feelings of anger or frustration
8.2	Sadness/depression	Caregiver discusses feelings of sadness or depression
8.3	Fear/anxiety	Caregiver discusses feelings of fear or anxiety
8.4	Hopeful	Caregiver expresses hopefulness
8.5	Gratitude	Caregiver expresses gratitude
8.6	Disappointment	Caregiver discusses feelings of disappointment
8.7	Shock/confusion	Caregiver discusses moments of shock or confusion
8.8	Trauma/PTSD	Caregiver discusses trauma or PTSD resulting from hospital experience
8.9	Expectations	Caregiver describes her/his expectations (met or unmet)
8.10	Trust	Caregiver discusses feelings of trust (trustful or not trustful)
8.11	Loneliness	Caregiver discusses feelings of being alone or isolated
8.12	Misunderstood/Misinterpretation by others	Caregiver discusses feelings of lack of understanding or misintepretation by others.

8.13	Uncertainty	Caregiver expresses feelings of uncertainty
8.14	Guilt	Caregiver expresses feelings of guilt
8.15	Normalcy	Caregiver expresses feelings of normalcy
8.16	Constant Concern	Caregivers expresses feelings of constant concern
8.17	Faith	Caregiver describes own faith
8.18	Humor	Caregiver describes humor or laughter
9	**Life impacts**	**Discussion centers on life impacts resulting from NICU experience**
9.1	Work	Impacts on work or career
9.2	Relationships/Friendships	Impacts on relationships/friendships/social networks
9.3	Financial	Financial impacts
9.4	Home environment changes	Home environment modification (e.g., accommodate equipment)
9.5	Impact on family members	Ways experience has impacted other family members
9.5.1	Impact on sibling	Impact on siblings
9.5.2	Impact on significant other	Impact on significant other
9.5.3	Impact on the child	Impact on the child
9.6	Neglect own health/needs	Neglects own physical or emotional care for sake of child
9.7	All encompassing changes	Caregiver expresses that experience has changed all aspects of her/his life
9.8	Social activities	Social life activities
9.9	social support	Social support
9.9.1	Supportive behaviors	Supportive behaviors
9.9.2	Unsupportive behaviors	Unsupportive behaviors
9.10	Change in Care Circumstance	Any change in circumstance over time in care (such as death in the family)
9.11	Self-Reliance	Having to rely on only self for help/no ability to rely on others
9.12	Fatigue	Fatigue of caregiver
10	**Coping**	**Discussion centers on comping methods**
10.1	Reframe	(e.g., this is my purpose)
10.2	Problem-based	(e.g., wrote a book)
10.3	Avoidant	medicate e.g food, alcohol etc
10.4	Emotion Based	prayer, meditation, driving, working out
10.5	Anticipating Future	Anticipate future
10.6	Acceptance	Acceptance of situation
11	**Settings of Care**	**Discussion centers on settings of care**
11.1	NICU/PICU	Discussion centers on NICU care
11.2	Outpatient/complex care clinic	Discussion centers on treatment in the outpatient care setting
11.3	Home	Discussion centers on home setting
11.4	Other	Other setting of care not listed above
12	**Suggestions/Needs**	**Discussion centers on caregiver's needs and suggestions**
12.1	Parental Support	Support groups, mentor/someone in similar situation
12.2	Hospital Specific	parent provides suggestion about support during hospitalization
12.3	Navigation of health care system	Support for certain intervention

12	**Suggestions/Needs**	**Discussion centers on caregiver's needs and suggestions**
12.1	Parental Support	Support groups, mentor/someone in similar situation
12.2	Hospital Specific	parent provides suggestion about support during hospitalization
12.3	Navigation of health care system	Support for certain intervention
13	**Timeframe**	**Timeframe for the topic of each quote**
13.1	Before	Context is prior to birth
13.2	hospital	Context is discharge from hospital
13.3	mixed	Mixed timeframe
13.4	current	current life situation
13.5	Future	Context is future of parent or child
13.6	Undetermined	Undetermined timeframe
14	**Diagnosis Status**	
14.1	Before Diagnosis	
14.2	After Diagnosis	
14.3	Mixed	
14.4	Undetermined	
15	**World impacts**	**Any discussion of world events**
16	Change over time	Changes over time with child
17	Noteworthy Quotes	