8-9-2022

Influence of Social Support and Family Resources on Workload, Capacity, and Depressive Symptoms in Parents of Children with Medical Complexity

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Recommended Citation

Lawrence, Patricia R., "Influence of Social Support and Family Resources on Workload, Capacity, and Depressive Symptoms in Parents of Children with Medical Complexity." Dissertation, Georgia State University, 2022.
doi: https://doi.org/10.57709/29692999

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This dissertation, INFLUENCE OF SOCIAL SUPPORT AND FAMILY RESOURCES ON WORKLOAD, CAPACITY, AND DEPRESSIVE SYMPTOMS IN PARENTS OF CHILDREN WITH MEDICAL COMPLEXITY by PATRICIA R. LAWRENCE was prepared under the direction of the candidate's dissertation committee. It is accepted by the committee members in partial fulfillment of the requirements for the degree of Doctor of Philosophy in Nursing in the Byrdine F. Lewis College of Nursing and Health Professions, Georgia State University.

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3. Lawrence, P.R., Feinberg, I., & Spratling, R. (2021). The relationship of parental health literacy to health outcomes of children with medical complexity. *Journal of Pediatric Nursing, 60, 65-70.* doi:10.1016/j.pedn.2021.02.014
4. Spratling, R., Chambers, R., Lawrence, P., & Faulkner, M. S. (2021). Best Practices with Use of Feeding Tubes for Children at Home. *Pediatric Nursing*, 47(1), 7-10.
5. Lawrence, P., Chambers, R., Faulkner, M. S., & Spratling, R. (2020). Evidence-Based Care of Children with Tracheostomies: Hospitalization to Home Care. *Rehabilitation Nursing,* 46(2): 83-86. doi:10.1097/RNJ. .0000000000000254
6. Spratling, R., Faulkner, M. S., Chambers, R., Lawrence, P., Feinberg, I., & Hayat, M. J. (2020). Establishing Fidelity for the Creating Opportunities for Personal Empowerment: Symptom and Technology Management Resources (COPE-STAR) Intervention. *Journal of Advanced Nursing*, 76(12), 3440-3447. doi:10.1111/jan.14552

Presentations since 2018

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2. Delaney, A., Riker, C., Green, S. & Lawrence, P. March, 2021. "Seven Ways for the APRN to Impact Change". Presentation for annual *National Association of Pediatric Nurse Practitioners* Conference (Virtual conference).
3. Lawrence, P. "Use of Theory Derivation to Understand Caregiver Workload and Capacity to Care for Children with Medical Complexity", February, 2020. Podium presentation at Georgia Nursing Leadership Coalition Annual Conference, Atlanta, GA.
4. Lawrence, P., Chambers, R., Faulkner, M., Feinberg, I., & Spratling, R. "Role of Health Literacy in Creating Education Modules for Caregivers of Children Requiring Medical Technology", August, 2019. Invited podium presentation at CHOA Nursing Research Symposium, Atlanta, GA.

ABSTRACT

INFLUENCE OF SOCIAL SUPPORT AND FAMILY RESOURCES ON WORKLOAD, CAPACITY, AND DEPRESSIVE SYMPTOMS IN PARENTS OF CHILDREN WITH MEDIAL COMPLEXITY

by

PATRICIA R. LAWRENCE

Children with medical complexity (CMC) are a small but growing population with chronic, sometimes life-limiting health conditions and high health care utilization. Their parents experience social and financial hardships and describe care as demanding. Research examining parental mental health is limited. The purpose of this study was to examine relationships among family resources, social support, parents' perceived workload to care for their CMC and their perceived ability to do the work, and how this impacts family burden and parental depressive symptoms.

A non-experimental, cross-sectional, correlational design was conducted in a national sample of 106 parent participants of CMC, recruited using social media. Pearson's correlations and multiple regression was conducted to examine relationships among the major variables. Parent participants were largely white (84.9%) biological (91.5%) mothers (98.1%) ranging in age from 23 to 47 years. The majority were married/partnered (86.8%) and college educated (37.7%) with incomes of $50,000 or more (61.3%). Nearly 62% of parents reported clinically significant depressive symptoms.

Higher family resources were associated with less workload (r = - 0.47, p <.001), increased capacity (r = .54, p < .001), and fewer depressive symptoms (r = - 0.56, p < .001) while more social support was associated with greater capacity (r = .44, p < .001). Higher workload was associated with lower capacity (r = - 0.33, p <.001). Workload and capacity significantly predicted family burden (Adj. R^2 = .515, F (2, 103) = 56.717, p = <.001). When workload exceeds capacity, it predicted both parental depressive symptoms (Adj. R^2 = .203, F (1, 104) = 27.714, p = < .001) and family burden (Adj. R^2 = .340, F (1, 104) = 54.996, p = < .001). Together, social support, family resources, workload, and capacity explained 32.4% of the variance in depressive symptoms and 56.5% of the variance in family burden, with family resources being a strong predictor in both models.

In this largely homogeneous sample of parents with CMC, depressive symptoms were common, family burden was significant, and both were explained by inadequate resources and high workload. Results emphasize the importance of care coordination support for families, as well as universal screening for social support, family resources, and depressive symptoms.

INFLUENCE OF SOCIAL SUPPORT AND FAMILY RESOURCES ON WORKLOAD, CAPACITY, AND DEPRESSIVE SYMPTOMS IN PARENTS OF CHILDREN WITH MEDIAL COMPLEXITY

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PATRICIA R. LAWRENCE

A DISSERTATION

Presented in Partial Fulfillment of Requirements for the
Degree of Doctor of Philosophy in Nursing in the Byrdine F. Lewis
College of Nursing and Health Professions,
Georgia State University

Atlanta, GA

2022

ACKNOWLEDGEMENTS

My PhD journey would not have been possible without the support of so many individuals. Dr. Regena Spratling provided countless hours of mentoring and guidance. I am deeply grateful that she served as my advisor and dissertation chair. She provided steadfast support and the willingness to take me under her wing. I sincerely appreciate the guidance from my dissertation committee which included Dr. Melissa Faulkner and Dr. Mathew Gayman. My entire committee had high expectations of me at each step, and I will carry the standards they set for me for the rest of my career. Dr. Aycock made every PhD student feel valued. Dr. Raeda Anderson helped me to understand, love, and appreciate the power of multiple regression.

Dr. Susan Kelley deserves special recognition because she had a significant hand in my decision to attend Georgia State University. I am extremely grateful for my family at the Heart Center at Children’s Healthcare of Atlanta. Their flexibility with my work schedule, along with their support, made this possible. The rest of my village includes many friends and family members. You know who you are because I relied on you so very often. My husband, Alexander was my rock for the last four years: he cooked, cleaned, and carried the weight. My daughters Marin and Sophia encouraged me when I thought about calling it quits. I could not have done this without the three of them.

But it is the parents of children with medical complexity who participated in this research study who I wish to thank the most. My hope is that this work encourages meaningful conversations and ideas that lead to the very best lives for their children. My “north star” has always been to do the right thing for children and their parents. So that is the direction in which I will keep moving.

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LIST OF ABBREVIATIONS

CAVC	Complete atrioventricular canal
CCHD	Critical Congenital Heart Disease
CCM	Cumulative Complexity Model
CDC	Centers for Disease Control and Prevention
CES-D	Center for Epidemiologic Studies Depression Scale
CHD	Congenital Heart Disease
CMC	Children with Medical Complexity
CoA	Coarctation of the aorta
DORV	Double outlet right ventricle
FaMM	Family Management Measure
FMSF	Family Management Style Framework
FRS-R	Revised Family Resource Scale
IAA	Interrupted aortic arch
IOF-R	Revised Impact on Family Scale
PA	Pulmonary atresia
PRQ2000	Personal Resource Questionnaire 2000
TA	Truncus arteriosus
TAPVR	Total anomalous pulmonary venous return.
TGA	Transposition of the great arteries
TOF	Tetralogy of Fallot
PVS	Pulmonary vein stenosis
VSD	Ventricular Septal Defect

CHAPTER 1

INTRODUCTION

Children with medical complexity (CMC), whose medical diagnoses vary in number and type, represent patients with high health care utilization due to their chronic, sometimes life-limiting health conditions that require a host of health care services (Cohen et al., 2011; Kuo et al., 2011). Although CMC represent less than one percent of all children in the United States (Berry et al., 2014), their care costs are disproportionally high, accounting for health care spending between $50-$110 billion annually, with up to 80 percent of this cost due to hospitalizations (Cohen et al., 2012; Lassman et al., 2014). Despite these costs, CMC health outcomes remain poor (Kuo et al., 2018; Shumskiy et al., 2018). While studies focused on CMC health outcomes are increasing, research examining the broader impact on the family, and parental mental health in particular, is limited. Understanding factors contributing to parental depressive symptoms is particularly important given evidence for worse health outcomes in children whose parents have poor mental health (Pierce et al., 2019) as well as evidence of higher pediatric health care use in depressed mothers (Brooks et al., 2015). Poor parental mental health may limit one's capacity to care for CMC who are more medically complex and who require frequent, active care and continuous monitoring at home, but these relationships have not been examined.

The care that CMC require by parents is demanding and stressful. In addition to the time required to coordinate and provide direct care to CMC, parents need to manage complex medication schedules, as well as medical equipment such as feeding tubes, tracheostomies, oxygen, and/or ventilators (Rehm, 2013). Time is needed to attend numerous appointments with health care specialists and these office visits are not always conveniently located near their home. Despite the time parents take to provide care to their CMC, these patients are more likely to have several unmet medical needs (Kuo et al., 2011).

This workload, sometimes referred to in the scientific literature as “demands” or “burdens”, is directly related to the daily care that parents provide to their CMC (Javalkar et al., 2017; Mooney-Doyle & Lindley, 2020). One systematic review found that parent demands, which are due to the complex treatment regimens and frequent appointments needed by their CMC, were associated with more illness-related parenting stress (Cousino & Hazen, 2013). This pile-up of demands affects family functioning (Mooney-Doyle & Lindley, 2019). The workload related to caregiving is *in addition* to other routine daily parental responsibilities such as caring for other children, laundry, meal preparation and cleaning, while sometimes maintaining a job outside the home.

Parents of CMC are negatively impacted financially, and studies describing the experiences of parents of CMC consistently report the financial hardships that go hand-in-hand with the time required to care for CMC (Kuo et al., 2014). For example, parents caring for CMC report costly out-of-pocket expenditures, and more than half of parents stop working outside the home to care for their CMC (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019). More than half of families experience financial problems which was found to be significantly associated with poverty in one study (Mooney-Doyle & Lindley).

Parents of CMC experience social hardships, and many parents describe the negative impact when social support is lacking, including feeling isolated and poorly understood by others (Caicedo, 2014; Mesman et al., 2013). Families of CMC are more likely to report little expectation of receiving any help from family or friends when assistance is needed (Thomson et al., 2016). Understaffed and underfunded community and home care services make the scarcity of social support that much more difficult for parents of CMC (Berry et al., 2014; Kuo et al., 2011). Parents of CMC describe the loss of free time to socialize or make friends, and the inability to get breaks from caring for their CMC (Whiting, 2013). Indeed, the complicated nature of caring for CMC places parents at increased risk for experiencing stressors associated with the workload that CMC require at home, in addition to the social and economic stressors that these parents are already known to experience (Pinquart, 2018; Thomson et al., 2016).

Social support improves coping and parenting self-efficacy and deters caregiver burden and depressive symptoms in some pediatric patient populations (Leahy-Warren et al., 2012; Tak & McCubbin, 2002). Therefore, given the link between stress and depression (Hammen, 2004), it is reasonable to anticipate that the workload associated with caring for CMC places parents at increased risk for depressive symptoms, which may in turn affect the health and well-being of CMC. Moreover, social support and family resources, which includes financial resources, may play a role in easing the burdens and depressive symptoms experienced by these families. However, these relationships require further exploration. Examining these relationships will provide a foundational understanding of how social support and family resources influence parental workload, parental depressive symptoms, and impact families of CMC. A deeper understanding has the potential to inform policies and influence the creation and availability of resources for CMC and their families. Moreover, a better understanding of these relationships will assist researchers to create targeted interventions that can minimize workload and strengthen capacity so that families are optimally prepared for caring for their CMC at home.

This study used the definition of CMC presented in a seminal work by Cohen et al. (2011) which describes CMC as 1) having one or more chronic clinical conditions which can be lifelong and severe such as critical congenital heart disease (CCHD); 2) having substantial health service needs such as multiple surgeries; 3) having functional limitations, such as requiring a nasogastric tube for feeding or oxygen supplementation; 3) having high health care use including the involvement with two or more subspecialty services over time.

This study included CMC with the diagnosis of CCHD to provide a thread of consistency and homogeneity in a group whose medical diagnoses are known to be heterogeneous (Kuo & Houtrow, 2016). Congenital heart disease (CHD) affects 8 out of 1,000 infants each year in the United States and is the most common type of birth defect (Van Der Linde et al., 2011). Approximately 1 in 4 infants with CHD is diagnosed with more *severe* CHD, known as critical congenital heart disease (CCHD) (Centers for Disease Control, 2020). This terminology is based on their need for expert care and specialized surgical intervention within the first year of life in order to survive. Many children with CHD fit the criteria for medical complexity based on the definition by

Cohen et al. due to their increased risk for neurodevelopmental morbidity, limited physical activity, increased health care utilization, health care costs, and the presence of other comorbidities (Razzaghi et al., 2015).

Significance of the Problem

Much of the existing literature on CMC and their parents has largely focused on describing drivers of CMC care costs, poor CMC health outcomes, and the demands and stressors that parents experience in providing necessary care for these children. Research examining CMC comprehensive care coordination programs that assist parents have demonstrated remarkably better CMC health outcomes and parent satisfaction due to improved communication between providers (Avritscher et al., 2019; Mosquera et al., 2014), suggesting that parental workload and capacity to manage the workload are relevant concepts to improving the health and well-being of these families. Although studies examining CMC health outcomes are growing, research examining the broader impact on CMC parents has been limited. Parents of CMC carry enormous burdens over the life course of their children. The workload that parents encounter, in addition to the challenges in parental capacity to manage the complexities of CMC care, may contribute to poor parental mental health and have a deleterious impact on the CMC and family, particularly when parental workload outweighs capacity. Adequate social support and family resources may attenuate the effects that workload and capacity have on parental mental health. However, there is a lack of research examining these relationships.

Purpose of the Study

The purpose of this study was to examine relationships among family resources, social support, workload and capacity, depressive symptoms in parents and impact on the family of children with medical complexity. In addition, the relationship between parental workload, parental depressive symptoms, and a number of sociodemographic and CMC clinical factors were explored.

Research Questions

For English-speaking biological, adoptive, or foster parents (single or partnered) who are 18 years of age or older and who consider themselves to be the primary caregiver of a child 6 months through 5 years of age with critical congenital heart disease (CCHD) as defined by the Centers for Disease Control (Centers for Disease Control, 2020) who fits the definition of CMC by Cohen et al. (2011):

1. What is the relationship of social support or family resources to parental workload or capacity?
2. What is the relationship between parental workload and capacity?
3. What is the relationship of parental workload or capacity to parental depressive symptoms or impact on the family of children with medical complexities?
4. What are the relationships among social support, family resources, parental workload and capacity, impact on the family and parental depressive symptoms?
   4a. Do social support, family resources or parental capacity moderate the relationship between parental workload and parental depressive symptoms or impact on the family?

5 What are the relationships among the number of prescribed CMC medications, number of specialists who care for CMC, number of hours per week of care coordination and number of hours per week providing direct care and parental workload or parental depressive symptoms?

Theoretical Framework

The theoretical framework used for this study was an integrated model derived from the Family Management Style Framework (Knafl et al., 2012) and the Cumulative Complexity Model (Shippee et al., 2012). This revised model, using the steps for theory derivation outlined by Walker and Avant (2019), incorporated the concepts of workload and capacity from the Cumulative Complexity Model (CCM) to enhance the dimensions of effort and ability within the Family Management Style Framework (FMSF). The revised model, as seen in Figure 1, provides richer meaning for understanding the relationship between the concepts of parental workload and parental capacity described in the CMC literature and offers a new and insightful way of explaining the potential effects that parental workload and capacity have on CMC, their parents, and the family, particularly when parental workload outweighs parental capacity.

The FMSF provides a framework for understanding how families respond to, manage, and incorporate a child's chronic illness into everyday family life. According to the Family Management Style Framework, a family's social network and family resources contribute to the ease or difficulty in how a family manages a child's chronic illness (Knafl et al., 2012). Therefore, the concepts of social support and family resources, which have been described in the CMC literature as distinct challenges that impact parental workload and capacity, are included in the theoretical model. Included in the FMSF are three major components within which are eight dimensions for understanding how families manage the care of children with chronic illnesses (Knafl et al., 2012). One of the eight dimensions, known as management mindset, refers to a parent's view of their ability to manage the illness, as well as whether the disease is easy or difficult to manage. This dimension is similar to the constructs of workload and capacity from the CCM. However, the CCM goes further to posit that when an imbalance exists between workload and capacity, specifically when workload outweighs ability, poor health outcomes can occur (Shippee et al., 2012). Similarly, the FMSF includes individual and family outcomes in the framework, since the FMSF aids in understanding how individual family members, and the family as a whole, are affected when poor adaption to a chronic illness exists (Knafl & Deatrick, 2003). Given that theoretical frameworks have rarely been included in studies focused on parents and their CMC, this framework fills an important gap in the CMC literature.

In the derived theoretical framework, presented in Figure 1, relationships have been established between the concepts of family resources, social support, parental workload and capacity and parental depressive symptoms and impact on the family in a number of categorical, pediatric chronic health conditions. However, few have examined these relationships in CMC whose care has been described as more complex and time consuming. Vessey et al. (2017) demonstrated a relationship between family resources (family finances) and its impact on families of children with cerebral palsy undergoing orthopedic surgery. Lower perceived social support was associated with more maternal mental health problems in at-risk mothers of premature infants (White-Traut et al., 2017). A relationship exists between parental workload and capacity wherein demanding and complex home care regimens for CMC may reduce parental capacity due to fatigue and stress, a phenomenon that has been described in studies involving CMC (Caicedo, 2014; Cousino & Hazen, 2013). Gibson-Young et al. (2014) established a relationship between parental capacity and a number of health and family outcomes in children with chronic asthma. Finally, a relationship exists between family management (which includes the dimensions of parental workload and capacity) and parental depressive symptoms in children with autism spectrum disorder (Kim et al., 2016).

Although relationships between these variables have been examined in patients with categorical chronic illnesses, studies examining these relationships are lacking in the CMC literature. Therefore, the model in Figure 1 was derived to test these relationships. In addition, this study considered a number of FMSF variables (family resources, social support, and parental capacity) to evaluate their moderating effects on the relationship between parental workload and parental depressive symptoms and impact on the family, depicted in Figure 2.

Figure 1

Theory for Understanding Parental Workload and Capacity to Care for Children with Medical Complexity

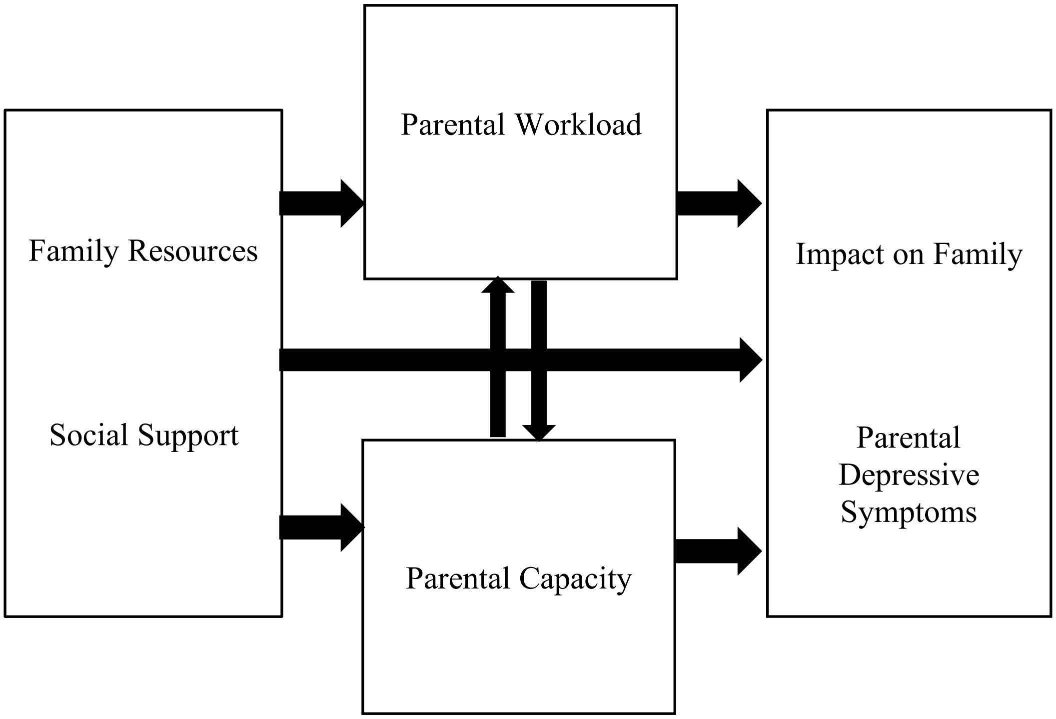
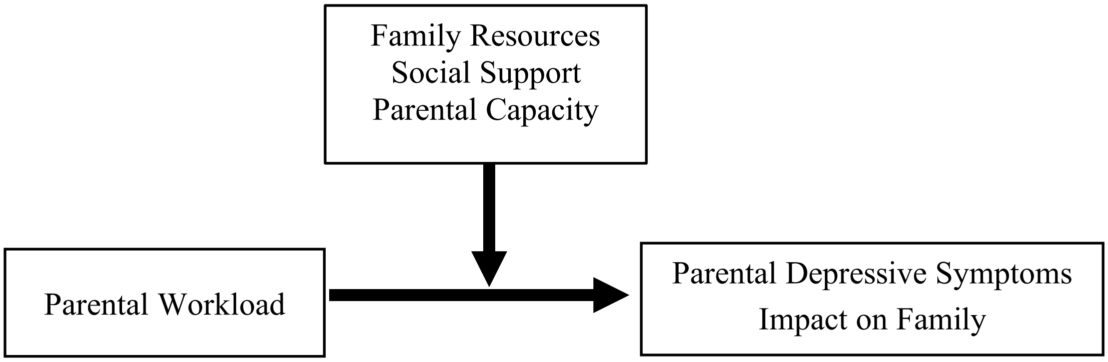


Figure 2

Testing for Moderation Effects



Abbreviated Review of Literature

Children with Medical Complexity: Definition and Prevalence

Children with medical complexity (CMC) have become an important population of research focus, given the growing numbers of CMC as well as their poor health outcomes, high health care utilization, and costs (Cohen et al., 2018; Cohen et al., 2011). The number of CMC has grown as a result of improved survival rates of conditions that were once fatal (Cohen et al., 2018; Kuo et al., 2011). The definition of CMC was derived from a need to further define a smaller subset of children with special health care needs who are especially medically fragile and whose health care requirements are the most intensive (Cohen et al., 2011). The term CMC, often used interchangeably with other terms including "medically complex", is becoming more widely used to signify the chronicity of illness, as well as the *time, resources and expertise* needed by parents and health care providers to manage their care and achieve optimal wellness (Cohen et al., 2018). Cohen et al. (2011) describes CMC as 1) having one or more chronic clinical conditions which can be lifelong and severe such as critical congenital heart disease (CCHD); 2) having substantial health service needs such as multiple surgeries; 3) having functional limitations, such as requiring a nasogastric tube for feeding or oxygen supplementation; 3) having high health care use including the involvement with two or more subspecialty services over time.

As the number of CMC has grown, so too has their medical complexity: a 2010 secondary analysis from the Nationwide Inpatient Sample analyzing more than 61 million pediatric hospitalizations over 15 years demonstrated increasing medical complexity of inpatient children (Burns et al., 2010). Most noteworthy references about CMC describe this population as being small, representing less than 1% of all children within the United States (Kuo et al., 2011). Their disproportionate care costs, described as totaling 40% of all pediatric hospital costs (Kuo et al., 2011; Simon et al., 2010), are in large part due to the complexity of their care which is influenced by a number of characteristics. For instance, CMC require inpatient and outpatient care from a variety of specialists, rely on expensive medications and therapies, require surgeries, procedures and life-sustaining technologies, and experience acute illnesses and exacerbations of chronic illnesses that require unique medical and surgical management (Cohen et al., 2011). These costs to the family, CMC and the system are driven higher when CMC care is not well-coordinated and when CMC parents are over-taxed because care coordination support is unavailable (Altman et al., 2018; Hofacer et al., 2019).

The following sections describe what is known about parental workload to care for CMC, parental capacity to care for CMC, and depressive symptoms of parents caring for CMC. These concepts have been selected based on CMC literature describing them as important to families and clinicians who care for this patient population (Barnert et al., 2018; Barnert et al., 2019; Fayed et al., 2019). The mental health of CMC parents, and depression in particular, has been identified as a research priority given the lack of research in this area, as well as the impact that compromised parental mental health can have on children (Barnert et al., 2019; Woolf-King et al., 2017). Finally, the theoretical model used for this study suggests that social support and access to family resources can either support or hinder parents' efforts to meet the challenges brought about by their child's health conditions (Knafl & Deatrick, 2003). Thus, family researchers have recommended these concepts be included when research is focused on understanding the intersection of family life and chronic health conditions (Knafl et al., 2012).

Factors Associated with Workload to Manage CMC Care

Workload refers to the enormous demands placed on parents to manage the day-to-day care of CMC. A number of terms have been used in the scientific literature to describe these demands, including "challenges", "stressors" and "burdens" when describing the day-to-day experiences of parents to manage care (Javalkar et al., 2017). In addition, parents feel on their own to navigate what they describe as a complex health care system that is unsupportive, siloed, fragmented, and confusing (Allshouse et al., 2018; Cady & Belew, 2017), compounding the demands they experience.

Workload due to Direct Care

Parents of CMC face a number of demands when caring for these medically fragile children. One common challenge that CMC parents encounter is in providing most of the direct home care to their children, which often requires specialized skills, medical equipment use, and complex medication schedules (Mooney-Doyle & Lindley, 2020;

Rempel et al., 2012). Nearly half of CMC receive health care at home by one or more family members, and almost 12% of CMC receive 21 or more hours of direct care each week at home by a family member (Romley et al., 2017). Researchers have calculated that approximately 5.6 million special health care needs children receive an annual 1.5 billion hours of family-provided care at home (Romley et al.). Outlining the time required of parents to care for their CMC is important when trying to understand its relationship to parental mental health and well-being, since more caregiving time has been associated with decreased quality of life for parents (Lawoko & Soares, 2003), and because caregiver demands are associated with more illness-related parenting stress (Cousino & Hazen, 2013).

Workload due to Care Coordination

In addition to providing a majority of direct, hands-on care, parents of CMC spend considerable time coordinating CMC care, with some parents describing this aspect of workload as a full-time job (Cady & Belew, 2017). Care coordination provides links between CMC, their family, and all health care team members, providing continuity over time and across health care settings. Therefore, accounting for care coordination workload is important because it has the potential to mitigate crisis-driven overtreatment of the CMC when done thoroughly, but this effort requires time and skills for which parents are not typically trained. Parents are expected to provide a level of care coordination that is typically provided by nurses or nurse practitioners in program-provided complex care programs (Ruggiero et al., 2019). Since care coordination services are often unavailable to most CMC parents (Hofacer et al., 2019; Mooney-Doyle & Lindley, 2020), parents must quickly adopt these skills. Moreover, community primary care providers and pediatric specialists do not typically integrate care across the array of providers involved in CMC care (Berry et al., 2014), further emphasizing the need for parents to take on this task and do it well.

Studies examining care coordination efforts have found that parents spend anywhere from 2 hours per week (Kuo et al., 2011) and up to 6.6 hours each week to coordinate care (Caicedo, 2014; Mooney-Doyle & Lindley, 2020), with 66.47% of CMC parents reporting no assistance with care coordination activities (Hofacer et al., 2019). Although effective care coordination has been demonstrated to improve CMC outcomes ((Mosquera et al., 2014), parents are left to manage this workload on their own without guidance and support.

Workload due to Health Care Access and Utilization

Parents experience workload by spending a considerable amount of time taking their CMC to health care appointments. In one study, parents and their CMC attended a median of 11 to 15 medical specialty clinics in one year (Kuo et al., 2011), while Cohen et al. (2012) found that CMC had a median of 13 different outpatient physicians providing necessary care from 6 distinct medical specialties. Some parents describe needing to travel frequently to their CMC's health care specialists, sometimes hundreds of miles away, because specialists are part of an urban, tertiary-care center (Cady & Belew, 2017). The location, number of appointments, and frequency of visits has an impact on the financial health and wellbeing of parents of CMC. A number of studies have underscored the financial impact that numerous outpatient visits and prolonged hospital stays have on families due to lost wages from interrupted work hours (Mooney-Doyle & Lindley, 2019; Thomson et al., 2016; Vessey et al., 2017).

Despite the time parents spend utilizing health care services, their CMC are still *twice as likely* to have one or more unmet need when compared to children without a complex medical condition (Kuo et al., 2011). In particular, needs such as some prescriptions and preventive dental and vision care went unmet, with more than 5.4% of CMC having 5 or more unmet medical service needs in the prior year. Kuo et al. reported 33% of parents of CMC had difficulty in accessing nonmedical services such as early intervention and childcare when compared to parents of children who were less medically complex. These studies, most of which are descriptive, underscore the workload that parents experience on a daily basis to access, manage, and coordinate the care of their CMC. Yet, despite the effort required by parents to do so, some needs of CMC go unmet.

Capacity to Manage CMC Care

Capacity refers to the ability, knowledge, readiness, or competency needed to manage a health condition (Shippee et al., 2012). Parents of CMC are required to manage the care of their child which may include complex treatment or medication regimes, complicated diets, and tracheostomy or feeding tube care (Caicedo, 2014; Romley et al., 2017). Indeed, mastering these demands is expected of parents (Knafl et al., 2013). However, parents receive little or no training in preparation for providing complex care (Barnert et al., 2019; Spratling, 2017).

Few studies have addressed the concepts of parental ability, competence, and readiness to manage the complex care that CMC require, and those that have examined this issue have mostly been descriptive and qualitative. A meta-synthesis of qualitative studies examining parents' experiences in caring for children with special health care needs described parents feeling overwhelmed and unprepared to care for their chronically ill child, requiring that parents quickly learn the skills necessary to provide complex care at home with little guidance (Nygard & Clancy, 2018). While parents willingly take on the care coordination and primary caretaker role, they describe frustration at the lack of resources necessary to meet these expectations (Cady & Belew, 2017). One qualitative study with 22 parents of children with congenital heart disease (CHD) found that nearly 32% of parents lacked the ability and knowledge to care for their child at home (Ni et al., 2019). Rempel (2012) interviewed parents of children with single ventricle physiology who described feeling like professionals after needing to quickly learn how to provide specific care to manage the needs of their chronically ill children. These studies emphasize the lack of ability, knowledge, and readiness experienced by parents of children with a variety of chronic conditions.

In an effort to reduce hospitalizations for CMC, Nelson et al. (2016) explored modifiable factors that influenced parents to bring their CMC in for acute care episodes that led to inpatient stays. Interviews with 35 parents of CMC found that parents “seek help when they pass a comfort threshold” for caring for their CMC at home (p. 584). Parents in the study expressed their desire for learning more about their CMC’s condition to assist in guiding them in how to respond to concerning symptoms with the hope of preventing unnecessary acute care visits. In addition, parents thought that having expanded access to health care providers that know their CMC would help them with making decisions about when to seek acute care more appropriately, suggesting that improving family capacity to navigate these situations by increasing parental knowledge and provider access would decrease hospitalizations and their associated costs.

Two quantitative studies measuring perceived parental ability to manage a child's chronic health condition were identified in the scientific literature, both using the Family Management Measure (FaMM). One of the six FaMM subscales measures parents' perceptions of their competence to manage their child's condition. Gibson-Young et al. (2014) examined maternal behaviors in managing their children's chronic asthma and found that when mothers had a lower perceived ability to manage chronic asthma, the number of child hospitalizations significantly increased. This finding suggests that some health care utilization patterns and child health outcomes are related to parents' knowledge and confidence to manage the disease (Gibson-Young et al., 2014). Zhang, Wei, Shen, and Zhang (2015) studied various aspects of family management in 399 Chinese caregivers whose children have a chronic health condition. These researchers found that caregivers living in urban areas, caregivers with a higher family income, and higher maternal caregiver education were the greatest predictors of a caregiver's ability to confidently manage their child's chronic condition, noting that this finding was consistent with other studies demonstrating an association between higher family income and better adaptation to managing a child's chronic illness.

Findings from these quantitative studies are important, since it is widely reported that CMC have poorer health outcomes and their families experience a number of burdens when caring for their CMC. The paucity of research examining the concept of parental capacity to meet the demands necessary to manage CMC care is notable, emphasizing the need for further examining the relationships between workload and capacity and their impact on CMC and parent health outcomes, as well as the CMC family as a whole. Fayed et al. (2019) described several target areas that deserved attention when evaluating CMC initiatives. One of the areas, all of which were developed in conjunction with parents of CMC, included outcomes of understanding parental ability to manage CMC health and parental ability to keep up with the demand of care, suggesting that parental workload and capacity are not well understood, yet are important concepts of interest to both researchers and parents of CMC.

Influence of Financial Hardship on Parents

Parents of CMC experience financial strain. High health care costs for CMC are due in large part to the complexity of CMC conditions which often require unique inpatient and outpatient medical and nursing management from one or more health care specialists, in addition to expensive medications, therapies, procedures and life-sustaining technologies (Berry et al., 2013). However, treatments, inpatient stays and outpatient appointments with specialists have a ripple effect that affect the financial status of these families due to the associated non-reimbursed, out-of-pocket expenses. Caring for CMC also frequently impacts the careers of these parents or spouses (Mandic et al., 2017), which further effects the financial health of these families over time. Moreover, CMC condition complexity is increasing over time (Burns et al., 2010) which leads to more deductibles and non-reimbursed medical expenses and causes further personal economic hardship and financial insecurity for these families (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019; Vessey et al., 2017).

Several studies have described the financial strain that CMC experience. In one of the first studies describing the financial challenges experienced by families of CMC, Kuo et al. (2011) used data from the 2005-2006 National Survey of Children with Special Health Care Needs using a sample size of 324,323 CMC. Researchers found that 46.3%

of parents paid more than $1,000 in the prior year toward out-of-pocket medical expenses, with 48.7% of parents reporting a lack of income to cover the medical expenses of their CMC, and another 56.8% of parents reporting financial difficulties directly related to the costs of unreimbursed CMC care. Moreover, 54.1% of families required that one family member stop working to provide direct care at home for their CMC. When compared to a cohort of families of children with asthma alone, nearly 50% parents of CMC reported financial difficulties, similar to those experienced by the most socioeconomically disadvantaged asthma families (Thomson et al., 2016), emphasizing that parents of CMC are especially hit hard financially.

Families of children with severe cerebral palsy spent between $193 and $7,192.71 *per hospitalization* for hip or spine surgery (Vessey et al., 2017). These families experienced financial strain with more than 75% of families needing additional funds to cover their child's expenses and costs related to missed work hours amounting to, on average, just over $1,000 per hospitalization. This study illustrates that while health insurance covers the majority of hospital-related medical costs, it doesn't cover all expenses that are especially associated with travel requirements to more distant health care centers for expert, regional care. These expenses include costs for transportation, lodging, food, and dependent care for those children that remain at home. These costs are in addition to the lost parent work hours that occur when accompanying their CMC.

Some parents of CMC experience poverty as a result of the lost income that comes from stopping work outside the home in order to care for their CMC (Mooney-Doyle & Lindley, 2019). This study also found that families reporting financial difficulties were also more likely to experience poverty. Similar to other study findings, nearly 40% of these families spent $1000 or more per month toward non-reimbursed medical expenses. It is clear from these studies, some of which use large, nationally representative samples, that the financial burden CMC parents experience is substantial, increases the likelihood of experiencing poverty, and appears to be directly related to the care that their CMC require. What is less clear are the relationships between resources, parental workload, and parental mental health, particularly when family financial resources are inadequate. While the financial costs to parents are inextricably linked to the unavoidable time and resources required to independently care for their medically complex child at home, few studies have examined relationships between financial resources needed to provide care and parental workload and parental mental health. Exploring these relationships are important given that parents have reported a reduced quality of life partially due to the financial difficulties associated with caring for their chronically ill child (Lawoko & Soares, 2003).

Influence of Social Support on Parents

Parents play a significant role in the day-to-day lives of their CMC. However, 64% of parents reported difficulty finding time for social activities, 61% had difficulty in finding energy for social activities, 44% had difficulty getting support from others, and 35% of parents felt isolated in one study (Caicedo, 2014). Social support has been associated with higher maternal self-efficacy and less postnatal depression in first-time mothers (Leahy-Warren et al., 2012), however these relationships are poorly understood in parents of CMC. Among parents of children defined as chronically ill with congenital heart disease (CHD), perceived social support was positively associated with parental coping, suggesting that social support may be a factor influencing the resiliency of parents caring for children with CHD (Tak & McCubbin, 2002). Parents of children with complex chronic conditions who report family support experience less caregiver burden (Toledano-Toledano & Domínguez-Guedea, 2019). Yet not enough is known about the relationship between social support and CMC and family outcomes. Understanding the role of social support is especially important in the CMC population given the numerous burdens their parents are known to experience.

Several small, qualitative studies have uncovered important themes relevant to social support of parents with children with complex health needs. Hudson et al. (2014) found that parents and health care providers both agree that family support acts as a protective factor in preventing hospitalizations and emergency room visits in children with chronic complex health conditions. Parents commonly describe family, friends, and health care professionals as important sources of social support in caring for their children with complex health needs (Whiting, 2014), whereas some parents identify support services that they need *more* of, or have no access to, in order to ease the intensity of CMC care responsibilities (Woodgate et al., 2015). Parents have noted that despite the many obstacles they encounter in providing care to their CMC, social networking and online support groups provide another avenue for sharing advice on how to ease the workload (Cady & Belew, 2017). Noting that a lack of social support can place further strain on parents, Thomson et al. (2016) explored the prevalence of social hardships among parents of CMC compared with parents of children with asthma and found that more than 50% of families caring for CMC reported social hardship, defined as not having anyone to ask for help or from whom to borrow money. Taken together, these studies help in beginning to shape our understanding of the toll that exists for parents of CMC when social support is lacking. These studies stress the important role that social support plays in improving coping and parenting self-efficacy and in deterring caregiver burden and depressive symptoms in some patient populations. However, further study is needed to examine these relationships in parents whose children require more intensive, complex care over the course of a lifetime.

Parental Depressive Symptoms

While the workload that parents face in providing direct care for CMC has been well described, little is known about the mental health consequences that this workload has on parents. Although a great deal more is known about the health outcomes of CMC, and while a focus on different aspects of parent health is beginning to emerge in the CMC literature, less is known about mental health, and depression in particular, in parents (Barnert et al., 2019).

Depression is more common among parents caring for chronically ill children. Studies have examined depression or depressive symptoms among parents whose children have single, categorical, chronic illnesses, but few were identified in the CMC population. For example, in parents of children with cerebral palsy, epilepsy, diabetes or renal disease, depressive symptoms ranged from mild in 37% of parents, to moderate and severe depressive symptoms in 38% of parents (Khanna et al., 2015). A systematic review and meta-analysis by Cohn et al. (2020) examined 26 publications assessing a variety of parental health outcomes including depression in parents whose children have a variety of chronic illnesses and found 35% of parents with chronically ill children met cutoff points for clinical depression compared to just 19% of parents with healthy children. Depression is more common among parents of children with intellectual and developmental disabilities (Scherer et al., 2019). Among parents of children with critical congenital heart disease, a systematic review and meta-analysis demonstrated an increased risk of mental health problems in the weeks and months following cardiac surgery, with 25 to 50% of parents reporting depressive symptoms (Woolf-King et al., 2017).

Only one study was identified that examined physical and mental health in parents of CMC in 84 English and Spanish-speaking parents in Florida (Caicedo, 2014). In this study, more than 50% of parents felt frustrated, anxious, sad, and angry over a five-month period, suggesting that these emotions are chronic in nature for parents of CMC. Evidence from small, cross-sectional studies as well as systematic reviews examining the mental health of parents of children with single, categorical, chronic diseases demonstrates that parents are at increased risk for depressive symptoms. However, there is a significant shortage of research specifically examining depressive symptoms in parents whose children are considered to be the most medically complex, and whose care is associated with a number of stressors, including financial burdens and a lack of social support.

Summary

Parents of CMC, including those diagnosed with CCHD, are at risk for mental health morbidity including depression (Woolf-King et al., 2017). A lack of social support, in addition to financial strain, have been described in the literature as significant hardships for these parents. Using a derived theoretical model to guide the exploration of these relationships, this study examined family resources and social support and their relationship to parental workload, parental capacity, parental depressive symptoms, and impact on the family. Findings from this study will contribute to greater knowledge about the concepts of parental workload and parental capacity to manage the care of their CMC with CCHD and how workload and capacity are influenced by family resources and social support, particularly when workload outweighs capacity. Developing a more thorough understanding of these relationships will provide insights for developing future interventions aimed at alleviating parental workload and enhancing their capacity to care for CMC.

Description of Student's Role in Study Development

This dissertation document represents the sole work by the student, including development of the study design, conduction of the study including enrollment and data collection, and completion of all aspects of the dissertation study. Study completion includes analysis of the data and interpretation of the results. The student is first author on all three manuscripts in accordance with the requirements for the three-manuscript dissertation option at Georgia State University School of Nursing. All three manuscripts have been included in this document as appendices A, B, and C.

CHAPTER 2

PRESENT STUDY

This chapter is a summary of the methods used for this study, as well as the most important findings of this study. The third appended, data-based manuscript entitled “Social Support and Family Resources Influence Workload and Capacity in Parents of Children with Medical Necessity” includes the methodology used to answer a portion of the research questions addressed in the third manuscript as part of this three-manuscript dissertation.

Methodology

Design

A non-experimental, cross-sectional, correlational design was used to examine relationships among family resources, social support, workload and capacity, impact on the family, and depressive symptoms in parents of children with medical complexity. This study also explored the relationship between parental workload and a number of sociodemographic and CMC clinical factors.

Sample and Setting

All aspects of this study were conducted remotely. The target population for this study were parents of CMC whose medical diagnosis included critical congenital heart disease (CCHD) between the ages of 6 months and 5 years of age. The definition of CMC by Cohen et al. (2011) was used. According to this definition, CMC have one or more chronic clinical conditions that are severe and expected to be lifelong. Therefore, the diagnosis of CCHD was required for eligibility into the study to provide a thread of consistency and homogeneity in a group whose medical diagnoses are known to be heterogeneous (Kuo & Houtrow, 2016).

Inclusion criteria for this study were: (1) parents (biologic, foster or adoptive parent) of a child with medical complexity which included the diagnosis of critical congenital heart disease according to the definition by the CDC (Centers for Disease Control, 2020); (2) age 18 years of age or older; (3) parents who self-identified as the primary care provider for their CMC; (4) able to read, speak and understand English; (5) child was a singleton birth (not a twin or triplet), aged 6 months through 5 years of age; and (6) had electronic access to all study questionnaires and instruments through a hand-held device, laptop, or desktop computer. A non-random (convenience and snowball) sample of 106 parents who met the inclusion criteria were recruited through the use of a digital flyer shared through local and national social media platforms and CMC parent support groups which allowed for a potentially more diverse sample of CMC parents.

Selection of CMC between the age of 6 months through 5 years of age provided an acceptable age range to allow for an adequate sample size, and during which timeframe a large proportion of CCHD surgical procedures take place. At the same time, this age range excluded the newborn period during which is known be a very stressful time for parents of a new child with medical complexities, especially those who require surgical intervention for CCHD (Woolf-King et al., 2018). Exclusion criteria included: (1) parents unable to read or speak in English since several measures used in this study only exist in English; (2) parents whose CMC was not a singleton birth, since multiple births may be associated with a perceived higher workload while also needing to manage CMC care.

Sample Size Calculation

A power analysis using G*Power (Faul et al., 2009), a free software program used to calculate statistical power, was used to determine the sample size for this study based on the specific aims and research questions of this study. Power analysis was conducted using bivariate correlation for research questions 1, 2, and 3, and using multiple regression for research questions 4 and 5. Using a moderate effect size, a power of .80, and an alpha level of .05, the sample size was determined by selecting the largest calculated sample size which is 92. After accounting for a 15% incomplete participant survey rate, the final target sample size was 106. A moderate association was anticipated based on findings from a similar study examining perceived maternal effort/workload and perceived maternal ability/capacity and the relationship to the health outcomes of their children with chronic asthma (Gibson-Young et al., 2014).

Protection of Human Subjects

Prior to initiating this study, Internal Review Board (IRB) approval was obtained from Georgia State University. The study was conducted remotely using Qualtrics, which is a secure, password-protected survey platform. While no harm from participating in this study was anticipated, the student PI acknowledged potential feelings of emotion that may be uncomfortable for some participants. Information and resources on depression were shared with all eligible participants, regardless of their depressive symptoms score. The purpose, overall procedure of the study, and potential risks and benefits of study participation were explained in the consent. Both the digital flyer and the consent form indicated that participation was voluntary, and parent participants could discontinue the survey at any time without repercussions, even after they had consented to the study.

The informed consent process was conducted online using Qualtrics, and the process explained the purpose of the study, how participants should contact the student PI or the Georgia State University IRB for any questions or concerns, and how data confidentiality would be maintained. In addition, the informed consent included an explanation of procedures that would be followed after data was collected, including incentive details upon completion of all measures, reinforcing that study participation was voluntary.

The confidentiality of all participants was maintained and conveyed throughout the study using a number of strategies. Participant identity remained confidential throughout the entire recruitment process since study participant numbers were assigned in Qualtrics and used on each study record to protect each participant's identity. The student PI maintained a separate, password-protected spreadsheet which contained participants' unique identification numbers associated with each participant's name and email address. The computer used by the student PI was password protected and equipped with a 5-minute inactivity timeout. A separate spreadsheet using participant identification numbers tracked and recorded completion of the study and incentive distribution. Since parents in this study with CES-D scores of 16 or higher are at risk for clinical depression, information about depression and available resources in the form of a letter was included in Qualtrics for all study participants, regardless of their CES-D scores.

Recruitment

Upon receipt of study approval from Georgia State University's Institutional Review Board, participants were recruited nationally using a remote, non-random, snowball sampling technique through multiple sources. Social media platforms including Instagram, LinkedIn, and Facebook groups that provide support to parents of CMC were used to inform potential parent participants of the study using a digital flyer. In addition, local and national parent support groups for children with medical complexity and children with CCHD who often fit the definition of CMC were used to inform potential parent participants with the same digital flyer. Eligibility screening was done individually, either by email, text, or by phone, after the potentially eligible participant contacted the student PI directly by email, text, or by phone. Eligible participants who completed the survey were then asked to inform other potential parent participants about the study by sharing the digital flyer. The email and telephone number of the student PI was provided on all digital flyers for potential participants who had questions or concerns about the study. Participants were recruited and enrolled into the study until the sample size of 106 was reached.

Measures and Instruments

Data collected in Qualtrics included demographic data, participants' responses to five instruments, and the email addresses for participants who wished to receive participation renumeration in the form of a $20 e-gift card of their choosing from Target or Walmart. Descriptions of the demographics survey and five instruments used in this study are provided in the following section.

Demographic Variables

A demographic survey developed by the student PI was used to assess a number of parent and CMC characteristics including age, sex, and race/ethnicity. Additional questions included parents' level of education, employment status, household income, marital/relationship status, residence zip code, if the parent had a car that was dependable, number of other children, the number of hours spent each week providing direct care, coordinating care, and traveling to appointments for the health care needs of their CMC; and the dollar amount that was spent on non-reimbursed costs related to their child's health needs in the last year.

A number of questions related to the CMC included health insurance type, type of critical congenital heart disease (CCHD), other medical diagnoses, medical equipment needs, the number of unpaid home caregivers, number of professional nursing hours received (if any), the number of specialists involved in the care of each CMC, and the number and frequency of medications taken. The Flesch-Kincaid grade level for the demographic survey was 6.2.

Family Resources

Family resources was the first of three independent variables for this study and was measured using the Revised Family Resource Scale (FRS-R) (Van Horn et al., 2001). This 20-item self-report scale measures the perception of available family resources including money to save, money for entertainment, time for self and family, and basic needs such as food, housing, and clothing. Using a 5-point response scale ranging from 1 (*never adequate*) to 5 (*always adequate*), total scores are obtained by summing responses with a potential total score of 20 to 100, with higher scores representing more perceived family resources. Internal consistency reliability for this instrument has ranged from .72 - .84, and validity has been established by exploratory and confirmatory factor analysis that resulted in the revised instrument that was stable using four factors (Van Horn et al.).

Social Support

Social support was the second of three independent variables for this study and was measured using the Personal Resource Questionnaire 2000 (PRQ2000) (Weinert, 2003). This instrument uses15 positively worded items to measure the overall level of perceived social support. Statements such as “there are people who are available if I need help over an extended period of time” and “When I am upset, there is someone I can be with who lets me be myself” are included. Using a 7-point Likert scale ranging from 1 (*strongly disagree*) to 7 (*strongly agree*), total scores are achieved by summing the 15 responses for a potential total score ranging from 15 to 105, with higher scores representing higher levels of perceived social support. Internal consistency reliability for this instrument has ranged from .87 to .93 from different adult samples and has undergone robust construct validity examination (Weinert).

Parental Workload and Capacity

The Family Management Measure (FaMM) measures parents’ perceptions of a chronically ill child’s treatment regimen and its incorporation into day-to-day life (Knafl et al., 2011). Two of the six subscales of the FaMM, used to measure perceived effort (workload) and perceived ability (capacity), were used for this study. The condition management effort (workload) subscale measures parents’ perceptions of the workload needed to manage their childrens’ chronic conditions using 4 items, with higher values signifying greater effort needed to manage the condition. Statements such as “it takes a lot of organization to manage our child’s condition” are included. Internal consistency reliability ranged between .74 for mothers and .78 for fathers (Knafl et al). The condition management ability (capacity) subscale measures parents’ perceptions of their competence to care for childrens’ chronic illnesses using 12 items, and higher values are associated with parents’ view of themselves as more capable of managing the chronic condition. Statements such as “when something unexpected happens with our child’s condition, we usually know how to handle it” are included. Internal consistency reliability for this subscale ranged between .72 for mothers and .73 for fathers (Knafl et al.). Construct validity has been supported by significant correlations with other family functioning measures (Knafl et al.).

Parental Depressive Symptoms

Parental depressive symptoms were assessed by the Center for Epidemiologic Studies Depression Scale (CES-D) (Radloff, 1977). This 20-item instrument uses a 4-point Likert scale ranging from 0 *(rarely or none*) to 3 (*all the time*), asking subjects how often in the past week they have experienced depressive symptoms. A total score is obtained by summing responses. Scores can range from 0 to 60, with higher scores indicating greater depressive symptomatology, and a score of 16 or greater indicating a risk for clinical depression. Internal consistency reliability has ranged from .78 to .89, and the CES-D has established validity, with the instrument having been used across a range of populations (Radloff).

Impact on Family

This study used the revised version of the Impact on Family Scale (IOF-R) to assess parents’ perceived burden of an ill child on the family (Stein & Jessop, 2003). This self-report measure uses 15 items on a 4-point Likert scale ranging from 1 (*strongly disagree*) to 4 (*strongly agree)*. Statements such as “Nobody understands the burden I carry” and “It is hard to find a reliable person to take care of my child” are included. Total scores are achieved by summing the 15 responses for a potential total score ranging from 15 to 60, with higher scores representing higher levels of perceived family burden due to the child’s illness. Internal consistency reliability has ranged from .83 to .89 (Stein & Jessop). The measure has undergone robust construct validity examination, and factor analysis confirmed that all items represent the negative impact on social and family systems (Stein & Jessop).

Data Collection and Management Procedures

After receiving IRB approval from Georgia State University, the student PI began sharing a digital recruitment flyer. Potential parent participants then contacted the student PI by email, text, or phone, according to directions placed on the digital flyer. After each participant was successfully screened for eligibility, a link to the informed consent and Qualtrics survey was emailed to each participant. The student PI’s contact information was provided at multiple stages during the consent process, as well as before and after the survey, for any participant questions or concerns. Consenting participants who wished to participate after reading the entire consent selected the appropriate statement to either proceed with the study or opt out by not completing the questionnaire and measures. The survey assigned identification numbers randomly to each consenting parent participant, and these unique numbers were used to track data and to protect the identification of every participant. After participants gave voluntary consent, they proceeded to the demographics questionnaire and all accompanying instruments, which took no more than

60 minutes to complete. Participants were encouraged to complete the surveys within one week, and upon completion of all instruments, each parent participant was emailed a digital gift card of $20 from Walmart or Target, depending on their preference. Participants were asked about their willingness to be contacted in the future for follow up questions or future studies. The study was suspended temporarily after the first 10 participants to assess for any patterns in missing data and to ensure that the time required for completing all measures was not originally underestimated. After reviewing data from the first 10 participants revealed no concerning patterns, the study was again re-opened. Once the sample size of 106 was reached, the Qualtrics survey was closed to new participants.

Data Analysis

All data was analyzed using IBM SPSS Version 27. Once data was transferred into SPSS, the data was checked for quality, missing values, outliers, and normality. Any outliers and missing values were validated with all study participants via email. Prior to addressing the research questions, assumptions of regression were tested for normality, outliers, and homoscedasticity and lack of multicollinearity. Internal consistency reliability was calculated for each of the instruments used in this study. Descriptive statistics were used to summarize the sample characteristics and responses to questions on the demographics form. Frequencies and percentages were presented for categorical variables, and means and standard deviations were presented for continuous variables. Bivariate correlation and multiple regression were used to examine relationships between the independent and dependent variables including testing for moderation effects. A significance value of .05 was used for all statistical analyses.

Results

A total of 137 potential parent participants initially expressed interest in study participation. Of the 137 potential participants, 31 participants were excluded: 4 lived outside of the United States, 6 had a child who no longer relied on medical technology, 13 had a child whose age excluded them from participating, and 8 participants did not complete all online surveys. This resulted in a final sample of 106 parent participants who completed all questions and measures.

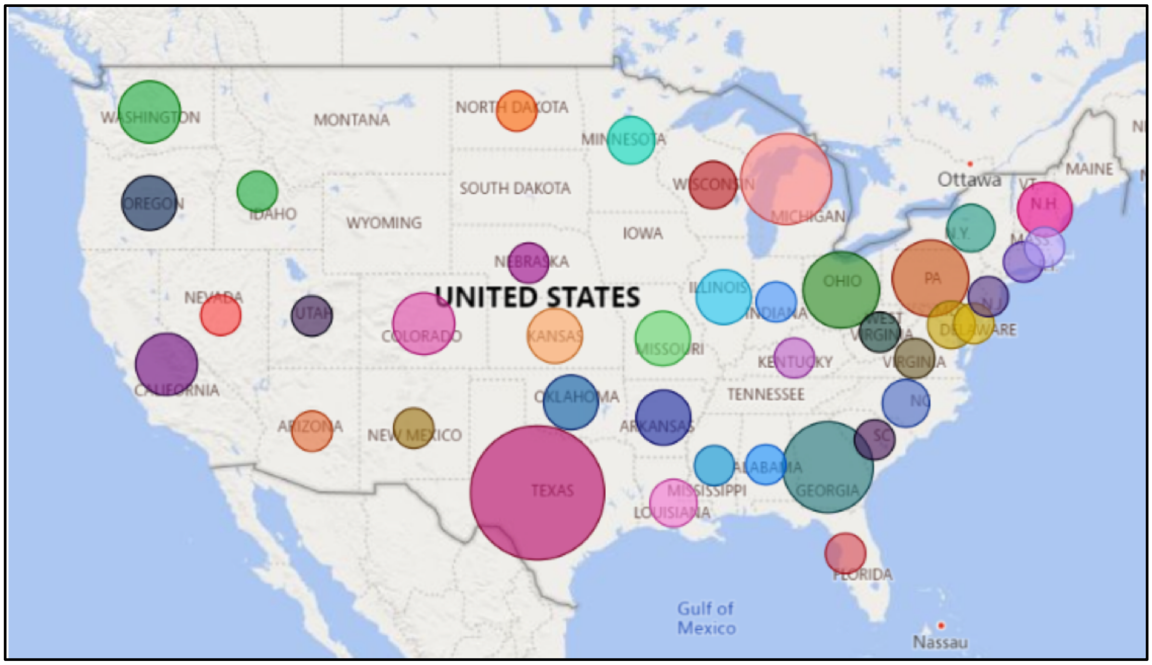
Sample Characteristics

The 106 parent participants of children with medical complexity represented a wide geographical span including 40 of the 50 United States as seen in Figure 3, the majority (13.2%) of which came from Texas. Parent participants' mean age was 32.95 (SD=5.06) and ranged between 23 and 47 years of age. The mean number of dependent children at home, which included their CMC, was 2.56 (SD=1.47). The majority of participants were married/partnered (86.8%) mothers (98.1%) who were the biologic parent (91.5%) of their CMC. The majority (84.9%) of parent participants were white, and just over half (51%) of parents worked outside the home, with 30.2% working full-time.

The majority (37.7%) of parent participants were college graduates with a working car (92.5%) to use for transporting their CMC to health care-related appointments, and with a reported family income of $50,000 or greater (61.3%). The majority of parent participants reported a history of changing their job status to care for their CMC, with 62 (58.5%) taking a leave of absence from their job due to their child's health condition, and 70 (66%) cutting down on work hours because of their child's condition. The majority of participants (61.3%) reported CES-D scores of 16 or higher, indicating these participants have a greater likelihood for clinical depression. Demographic characteristics of the sample are summarized in Table 1.

Figure 3

Representativeness of the Sample (N=106)



Note. Larger bubbles = greater sample size per state

Table 1

Parent Participant Characteristics (N=106)

Characteristics	*M*	*(SD)*	*Observed Range*	*N*	*(%)*
Age (years)	32.95	5.06	23-47	106	
Number of children at home[a]	2.56	1.47	1-8		
Parent Type					
Mother				104	(98.1%)
Father				2	(1.9%)
Biologic				97	(91.5%)

Foster	1	(.9%)
Adoptive	8	(7.5%)
Marital Status		
Married/Partnered	92	(86.8%)
Single/Divorced	14	(13.2%)
Race/Ethnicity		
A. Indian or Alaska Native	4	(3.8%)
Asian	4	(3.8%)
Black or African American	3	(2.8%)
Hispanic	10	(9.4%)
Hawaiian or Pacific Islander	1	(.9%)
White, Non-Hispanic	90	(84.9%)
Two or more races	4	(3.8%)
Employment Status		
Full-time	32	(30.2%)
Part-time	22	(20.8%)
Unemployed	52	(49.1%)
Education Level		
High school graduate	10	(9.4%)
Some college	31	(29.2%)
College graduate	40	(37.7%)
Post-graduate study	25	(23.6%)
Working car for CMC transportation		
No car	4	(3.8%)
Own car	98	(92.5%)
Unreliable car	4	(3.8%)
Household Income		
< $50,000	41	(38.7%)
≥ $50,000	65	(61.3%)
History of Job Status Change		
Left job to care for CMC	39	(36.8%)
Took leave of absence to care for CMC	62	(58.5%)
Cut down hours to care for CMC	70	(66%)
Avoided promotion to care for CMC	28	(26.4%)
Avoided job change to keep insurance	49	(46.2%)
Participants with CES-D scores ≥ 16[b]	65	(61.3%)

Note. [a]Includes CMC. [b]Indicative of greater risk for clinical depression.

Characteristics of CMC

The mean age in months of CMC was 33.78 (SD=18.99) and ranged between 6 and 71 months of age. The majority of CMC were white (71.7%) males (59.4%) with public health insurance (46.2%). All CMC had some type of critical congenital heart disease (CCHD) that required surgical intervention in the first year of life, the three most common of which were ventricular septal defect (39.6%), single ventricle physiology (31.1%) and Tetralogy of Fallot (28.3%). The CMC in this study had a number of other health conditions, with only 15 (14.2%) reporting no other health conditions aside from their CCHD diagnosis. Forty-three (40.6%) CMC had between 1-3 other health conditions, whereas 31 (29.2%) had between 4-6 other health conditions beside their CCHD diagnosis. Seventeen (15.9%) CMC had 7 or more health conditions. The three most common conditions included developmental delay (63.2%), speech/language delay (52.8%), and genetic conditions other than Trisomy 21 (26.4%).

CMC used a mean of 5.67 (SD=4.56) daily medications. Forty (37.7%) children need to be given medication three or more times a day, with the majority (54.7%), needing medication once or twice a day. All CMC in this study used at least one piece of life-sustaining medical equipment, with a mean of 2.53 (SD=2.32) pieces of medical equipment, the most common which was respiratory equipment (49.1%). See Table 2 for the demographic characteristics of CMC.

Table 2

CMC Characteristics (N=106)

Characteristics	*M (SD)*	*Observed Range*	*N*	*(%)*
Age in months	33.77 (19)	6-71		
Gender				
Males			63	(59.4%)
Females			43	(40.6%)
Ethnicity				
A. Indian or Alaska Native			3	(2.8%)
Asian			3	(2.8%)
Black or African American			4	(3.8%)
Hawaiian or Other Pacific Islander			2	(1.9%)
White, non-Hispanic			76	(72.4%)
Hispanic			13	(12.3%)
Two or more races			17	(16.2%)
Insurance Type				
Public			49	(46.2%)
Private			25	(23.6%)
Both			32	(30.2%)
CCHD Type				
VSD			42	(39.6%)
Single Ventricle			33	(31.1%)
TOF			30	(28.3%)
PA			20	(18.9%)
DORV			18	(17%)
TGA			12	(11.3%)
CoA			11	(10.4%)
PVS			9	(8.5%)
CAVC			7	(6.6%)
IAA			5	(4.7%)
TA			3	(2.8%)
TAPVR			2	(1.9%)
Ebstein anomaly			2	(1.9%)
Other Health Conditions				
None			15	(14.2%)
Between 1-3			43	(40.6%)
Between 4-6			31	(29.2%)

More than 6			17	(15.9%)
Number of Daily Medications	5.67 (4.56)	1-25		
Medication Frequency				
No medications			8	(7.5%)
1-2 times a day			58	(54.7%)
Three or more times a day			40	(37.7%)
Number of Medical Equipment	2.53 (2.32)	1-12		
Medical Equipment Type				
Cardiovascular			10	(9.4%)
Respiratory			52	(49.1%)
Digestive			79	(74.5%)
Medication			12	(11.3%)
Mobility			19	(17.9%)
Hearing			3	(2.8%)
Other			7	(6.6%)

Note. VSD=Ventricular septal defect; TOF=Tetralogy of Fallot; PA=Pulmonary atresia; DORV=Double outlet right ventricle; TGA=Transposition of the great arteries; CoA=Coarctation of the aorta; PVS=Pulmonary vein stenosis; CAVC=Complete atrioventricular canal; IAA=Interrupted aortic arch; TA=Truncus arteriosus; TAPVR=Total anomalous pulmonary venous return.

Care Requirements of CMC

Parents reported a mean of 72.25 (SD=49.19) weekly hours providing direct care to manage their CMCs' conditions, and a mean of 8.57 (SD=7.57) hours weekly to coordinate care of their CMC. The majority of parents (37.7%) reported having no one other than themselves to provide care for their CMC, while 32% of the parent sample had just one other person to assist them in unpaid caregiving. The majority of CMC (62.3%) received no paid nursing hours to provide care at home. Of the remaining 37.7% who did receive paid nursing hours, the mean number of weekly hours received was 18.02 (SD=34.79). The CMC in this study required a mean of 7.2 (SD=3) health care specialists to help manage their health care conditions. CMC attended a mean of 5.22 (SD=5.22) well visits in the prior year and a mean of 25.48 (SD=36.24) visits to subspecialists in the prior year, resulting in a mean of 6.44 (SD=7.56) estimated weekly hours traveling to see CMC health care providers. More than half (55.7%) of the parent sample reported paying an estimated $1000 or more annually for out-of-pocket expenses for their CMC, with just over 25% of the entire sample reporting estimated out-of-pocket costs of $5000 or more. Table 3 summarizes characteristics of the care requirements for CMC.

Table 3

Care Requirements of CMC (N=106)

Characteristics	*M*	*(SD)*	*Observed Range*	*N*	*%*
CMC paid weekly nursing hours [a]	18.02	34.79	0-168	41	38.7
Weekly direct care hours	72.25	49.19	1-168		
Weekly care coordination hours	8.57	7.57	1-33		
Number unpaid caregivers	1.09	1.13	1-4		
Number annual well visits	5.22	5.22	0-30		
Number annual specialty visits	25.48	36.24	1-310		
Number of specialists	7.2	3.00	2-19		
Weekly travel hours	6.44	7.56	0-45		
Out-of-pocket Medical Expenses[b]					
$0				9	(8.5%)
$1 to $499				25	(23.6%)
$500 to $999				13	(12.3%)
$1000 to $5000				32	(30.2%)
>$5000				27	(25.5%)

Note. [a]Number and % of CMC who received paid nursing hours. [b]Annual estimates.

Reliability of Instruments

Table 4 summarizes the mean, standard deviation, minimum and maximum scores, and Cronbach's alpha reliability coefficients for all instruments or subscales used to measure the major variables included in this study. All instruments and subscales had acceptable internal reliability consistency defined as a minimum alpha coefficient of >.70.

Personal Resource Questionnaire 2000. The 15-item PRQ 2000 was completed by all 106 participants with a mean total score of 77.89 (SD=17.91), a minimum score of 15 and a maximum score of 105. The Cronbach's alpha reliability coefficient was acceptable at .93.

Revised Family Resource Scale. All 106 parent participants completed the 20-item Revised Family Resource Scale resulting in a mean total score of 72.58 (SD=12.34) with a minimum score of 38 and a maximum score of 75. The Cronbach's alpha reliability coefficient for this scale was acceptable at .91.

Family Management Measure (FaMM) Effort and Ability Subscales. All 106 parent participants completed the 4-item condition management effort subscale of the FaMM to measure perceived workload. The mean subscale score was 16.75 (SD= 2.77), a minimum score of 10, and a maximum score of 20. The Cronbach's alpha for this subscale was .70. In addition, all participants completed the 12-item condition management ability subscale of the FaMM, used to measure perceived capacity. The mean score for this subscale was 43.89 (SD=6.56), the minimum score was 27 and the maximum score was 58. The Cronbach's alpha for this subscale was .71.

Center for Epidemiologic Studies Depression Scale (CES-D). The 20-item CES-D was completed by all 106 participants, resulting in a mean total score of 20.95 (SD=12.01), a minimum score of 0 and a maximum score of 49. The Cronbach's alpha reliability coefficient was acceptable at .93.

Revised Impact on Family Scale (IOF-R). All 106 participants completed the 15-item IOF-R, resulting in a mean total score of 45.21 (SD=8.48). The minimum score for this sample was 18, the maximum score was 60, and the Cronbach's alpha reliability coefficient was acceptable at .89.

Table 4

Descriptive Statistics and Cronbach's Alpha for Major Study Variables (N=106)

Variable	*M*	*(SD)*	*Observed Range*	*Possible Range*	*Cronbach's Alpha*
<u>Perceived Social Support</u> PRQ2000	77.89	17.91	15-105	15-105	.93
<u>Perceived Family Resources</u> FRS-R	72.58	12.34	38-75	20-100	.91
<u>Perceived Workload</u> FaMM Effort Subscale	16.75	2.77	10-20	4-20	.70
<u>Perceived Capacity</u> FaMM Ability Subscale	43.89	6.56	27-58	12-60	.71
<u>Depressive Symptoms</u> CES-D	20.95	12.01	0-49	0-60	.93
<u>Impact on Family</u> IOF-R	45.21	8.48	18-60	15-60	.89

Descriptive Statistics for Major Study Variables

Descriptive statistics were used to check for accuracy, outliers, and missing data. What little missing data existed was obtained by emailing participants who gave permission to be contacted. Boxplots demonstrated that three of the major variables (perceived capacity, perceived social support, and impact on family) had one or two outliers, but had no influence on the results. There was no violation of assumptions when testing assumptions for Pearson's correlations among the major study variables. Prior to addressing the research questions, the assumptions of regression were tested for normality, outliers, lack of multicollinearity, and homoscedasticity, and there were no violations.

Relationships among Major Study Variables

Bivariate correlations were used to examine the relationships among the major study variables, as reported in Table 5. Many significant associations were found between the major variables. Based on bivariate correlations, higher perceived family resources were significantly associated with higher perceived social support, decreased workload, higher perceived capacity, fewer depressive symptoms, and less impact on the family. While higher perceived social support was not significantly associated with perceived workload, it was significantly associated with higher perceived capacity, fewer depressive symptoms, and less impact on the family. Higher workload was associated with higher depressive symptom scores and more impact on the family. Perceived capacity was negatively correlated with perceived workload. Higher perceived capacity was significantly associated with lower depressive symptoms and less impact on the family. Finally, higher depressive symptoms were significantly associated with greater impact on the family.

Table 5

Relationships among Major Study Variables

	FRS-R	PRQ2000	Workload	Capacity	CES-D
1. FRS-R					
2. PRQ2000	.55***				
3. Workload	-0.47***	-0.12			
4. Capacity	.54***	.44***	-0.33***		
5. CES-D	-0.56***	-0.44***	.33***	-0.33***	
6. IOF-R	-0.60***	-0.29**	.69***	-0.44***	.45***

Note. *p<0.05. **p<0.01. ***p<0.001. FRS-R= Revised Family Resource Scale; PRQ2000 = Personal Resource Questionnaire 2000; CES-D= Center for Epidemiologic Studies Depression Scale; IOF-R=Revised Impact on Family Scale.

Data Analyses for Research Questions

Research Question 1: What is the relationship of social support or family resources to parental workload or capacity?

Results from the Pearson product-moment (r) correlation analyses (Table 5) were examined to answer research question 1. There was a moderate negative correlation between family resources and workload (r = -0.47, p <.001) and a strong positive correlation between family resources and capacity (r = .54, p <.001). These statistically significant correlations indicate that increased perceived family resources were associated with a decrease in perceived workload and higher perceived capacity.

There was a weak negative correlation between social support and workload (r = -0.12, p>.05). This relationship was not statistically significant, demonstrating that perceived social support did not influence perceived workload in this study. A moderate positive correlation between social support and capacity (r = .44, p <.001) was found. Thus, this statistically significant association indicates that more perceived social support was associated with higher perceived capacity.

Research Question 2: What is the relationship between parental workload and capacity?

Correlation analyses in Table 5 were examined to answer this research question. There was a moderate negative correlation between workload and capacity (r = - 0.33, p <.001) indicating that lower perceived capacity is significantly associated with an increase in perceived workload.

Research Question 3: What is the relationship of parental workload or capacity to parental depressive symptoms or impact on the family of children with medical complexity?

Correlation analyses in Table 5 were examined to assess the relationships between the predictor variables of workload or capacity to the outcome variables of parental depressive symptoms or impact on the family. There was a moderate positive correlation between workload and depressive symptom scores (r = .33, p < .001) and a strong positive correlation between workload and impact on family scores (r = .69, p <.001).

These statistically significant correlations indicate that depressive symptoms and impact on family scores were higher with higher perceived workload.

When examining the relationship between perceived capacity and the outcome variables of depressive symptoms and impact on family, there was a moderate negative correlation between capacity and depressive symptom scores ($r = -0.33$, $p < .001$), and a moderate negative correlation between capacity and impact on family scores ($r = -0.44$, $p < .001$). These statistically significant correlations indicate that with higher perceived capacity, there are lower depressive symptom scores and impact on family scores.

To better understand the full impact that workload and capacity has on parental depressive symptoms and impact on the family, multiple regression was conducted to test the relationships. Table 6 contains the regression analysis predicting parental depressive symptoms with a three-model series: Model 1 with parental workload, Model 2 with parental capacity, and Model 3 with parental workload and parental capacity. Because the sample was very homogeneous with regard to race, gender, and education, and because this study was exploratory, no covariates were used in any of the regression models.

In Model 1, parental workload has a positive relationship ($\beta = 0.33$, $SE=0.40$, $p<0.001$) with parental depressive symptoms, and parental workload accounts for 10.1% of the variability in parental depressive symptoms (Adj. $R^2= 0.101$). Thus, as parents' perceived workload increases, parental depressive symptoms also increase. Model 2 findings indicate that there is a negative relationship between parental capacity ($\beta = -0.33$, $SE= 0.17$, $p<0.001$) and parental depressive symptoms. Parental capacity accounts for 9.9% of the variability in parental depressive symptoms (Adj. $R^2 = 0.099$). Therefore, parents who have higher perceived capacity are less likely to have depressive symptoms.

Lastly, Model 3 analyses are similar to Models 1 and 2 in that there is a positive relationship between parental workload and depressive symptoms (β = 0.25, SE= 0.41, p=0.010) and there is a negative relationship between parental capacity and depressive symptoms (β = -0.25, SE= 0.17, p=0.011). Taken together, perceived parental workload and parental capacity account for 14.8% of the variation in parental depressive symptoms (Adj. $R^2 = 0.148$). Thus, Models 1 and 3 indicate that parents with higher workload are more likely to have higher levels of depressive symptoms than parents with lower perceived workloads. Additionally, Models 2 and 3 indicate that parents with higher capacity are more likely to have low levels of depressive symptoms when compared to parents with lower levels of perceived capacity.

Table 6

Parental Depressive Symptoms, Regression

	Model 1			Model 2			Model 3		
	β	SE	Sig.	β	SE	Sig.	β	SE	Sig.
Parental									
Workload	.33	.40	<.001***				.25	.41	.010*
Capacity				-.33	.17	<.001***	-0.25	.17	.011*
Constant	-	6.81	.64	-	7.52	<.001***	-	11.94	.063
Adj. R^2		.101			.099			.148	

Note. *p<0.05. **p<0.01. ***p<0.001.

Table 7 contains the regression analysis predicting impact on the family with a three-model series: Model 1 with parental workload, Model 2 with parental capacity, and Model 3 with parental workload and parental capacity. In Model 1, parental workload has a positive relationship (β= 0.69, SE=0.22, p<0.001) with impact on family, with parental workload accounting for 46.7% of the variability in impact on the family scores (Adj. R^2= 0.467). Thus, as parents' perceived workload increases, so too does impact on the family. Model 2 findings indicate that there is a negative relationship between parental capacity (β= -0.44, SE= 0.11, p<0.001) and impact on the family. Parental capacity accounts for 18.6% of the variability in impact on family scores (Adj R^2= 0.186). Therefore, parents who have higher perceived capacity are less likely to have an impact on the family.

Lastly, Model 3 analyses are similar to Models 1 and 2 in that there is a positive relationship between parental workload and impact on the family (β= 0.61, SE= 0.22, p=< .001) and there is a negative relationship between parental capacity and impact on the family (β= -0.24, SE= 0.09, p=0.001). Taken together, perceived parental workload and parental capacity account for 51.5% of the variation in impact on family scores (Adj. R^2= 0.515). Thus, Models 1 and 3 indicate that parents with higher workload are more likely to have a greater impact on the family than parents with lower perceived workloads. Additionally, Models 2 and 3 indicate that parents with higher capacity are more likely to have less of an impact on the family when compared to parents of CMC with lower levels of perceived capacity.

Table 7

Impact on Family, Regression

	Model 1			Model 2			Model 3		
	β	SE	Sig.	β	SE	Sig.	β	SE	Sig.
Parental									
Workload	.69	.22	<.001***				.61	.22	<.001***
Capacity				-0.44	.11	<.001***	-.24	.09	.001**
Constant	-	3.71	.008**	-	5.04	<.001***	-	6.36	<.001***
Adj. R^2	.467			.186			.515		

Note. $^{*}p<0.05$. $^{**}p<0.01$. $^{***}p<0.001$.

Part of the theoretical framework used for this study posits that poor health outcomes can occur when illness management workload outweighs the capacity to manage it (Shippee et al., 2012). Therefore, to explore the impact of parental depressive symptoms and impact on the family when workload exceeds capacity, z-scores were first generated for the variables of perceived workload and capacity. These variables required standardization due to the differences in ranges of their scores. A new variable was therefore created to represent workload exceeding capacity by using re-coded variables: workload z-scores greater than capacity z-scores were coded as 1, while workload z-scores that were equal to or less than capacity z-scores were coded as 0.

Results of the simple linear regression analysis is shown in Table 8 using this new variable to predict parental depressive symptoms. Findings indicate that when workload exceeds capacity, it significantly positively predicts parental depressive symptoms (Adj. $R^2 = .203$, $F(1, 104)=27.714$, $p=< .001$). Thus, 20.3% of the variability in parental depressive scores can be explained when workload exceeds capacity.

Table 8

Parental Depressive Symptoms, Regression

	Model		
	β	SE	Sig.
Workload>Capacity	.46	2.09	<.001***
Constant	-	1.53	<.001***
Adj. R^2		.203	

Note. *p<0.05. **p<0.01. ***p<0.001.

Table 9 contains the regression analysis predicting impact on the family when workload exceeds capacity. Findings indicate that when workload exceeds capacity for parents of CMC, it significantly positively predicts impact on family scores ($R^2 = .340$, $F(1, 104) =$

54.996, p= <.001). Thus, 34% of the variability in impact on the family can be explained when workload exceeds capacity.

Table 9

Impact on Family, Regression

	Model		
	β	SE	Sig.
Workload>Capacity	.59	1.34	<.001***
Constant	-	.98	<.001***
Adj. R^2		.340	

Note. *p<0.05. **p<0.01. ***p<0.001.

Research Question 4: What are the relationships among social support, family resources, parental workload and capacity, impact on the family and parental depressive symptoms?

Multiple linear regression was conducted to test the relationships between the predictor variables of social support, family resources, workload, and capacity, and the outcome variables of parental depressive symptoms and impact on the family. Since multiple regression findings examining the impact of workload and capacity on parental depressive symptoms (Table 6) and impact on the family (Table 7) were previously described in Research Question 3, multiple regression to address Research Question 4 first examined the impact of social support and family resources on parental depressive symptoms and impact on the family in the first two models. Then multiple regression was conducted using all four predictor variables in the third regression model.

Table 10 contains the regression analysis predicting parental depressive symptoms with a three-model series: Model 1 with social support, Model 2 with family resources, and Model 3 with all four predictor variables (social support, family resources, workload, and capacity). In Model 1, social support had a negative relationship (β= -0.44, SE=0.06, $p<0.001$) with parental depressive symptoms, and social support accounts for 18.3% of the variability in parental depressive symptoms (Adj. R^2= 0.183). Thus, as parental perceived social support increases, parental depressive symptoms decrease. Model 2 findings indicate that there is a negative relationship between family resources (β= -0.56, SE= 0.12, $p<0.001$) and parental depressive symptoms. Perceived family resources account for 30.8% of the variability in parental depressive symptoms (Adj. R^2= 0.308). This indicates that parents who have higher family resources are less likely to have depressive symptoms. Moreover, when entered individually, family resources had the strongest correlation with depressive symptoms compared with the other three predictor variables of social support, workload, and capacity.

Model 3 findings demonstrate that when all four predictor variables are included in the regression model, only social support (β= -0.21, SE=0.07, p = .04) and family resources (β= -0.39, SE=0.18, $p<0.001$) significantly predict parental depressive symptoms (Adj. R^2= .324, F (4, 101)=13.603, p=<.001). Thus, 32.4% of the variance in parental depressive scores can be explained by the combined effect of social support, family resources, perceived workload, and perceived capacity.

Table 10

Parental Depressive Symptoms, Regression

	Model 1				Model 2				Model 3			
	β	SE	Sig.		β	SE	Sig.		β	SE	Sig.	
Support												
Social Supp.	-0.44	.06	<.001	***					-0.21	.07	.038	*
Family Res.					-0.56	.12	<.001	***	-0.39	.18	<.001	***
Parental												
Workload									.13	.41	.172	
Capacity									.02	.18	.871	
Constant	-	4.73	<.001	***	-	7.41	<.001	***	-	13.67	<.001	***
Adj. R^2	.183				.308				.324			

Note. *p<0.05. **p<0.01. ***p<0.001.

Table 11 contains the regression analysis predicting impact on the family with a three-model series: Model 1 with social support, Model 2 with family resources, and Model 3 with all four predictor variables (social support, family resources, workload, and capacity). In Model 1, social support has a negative relationship (β= -0.29, SE=0.04, p=.002) with impact on the family, and social support accounts for 7.7% of the variability in impact on family scores (Adj. R^2= .077). Thus, as parental perceived social support increases, impact on the family decreases. Model 2 findings indicate that there is a negative relationship between family resources (β= -0.60, SE= 0.08, $p<0.001$) and impact on family scores. Perceived family resources account for 35.4% of the variability in impact on family scores (Adj.R^2= 0.354). This indicates that parents with higher family resources are less likely to impact the family. When entered individually, workload has the strongest correlation with impact on the family when compared with the other three predictor variables.

Model 3 findings demonstrate that when all four predictor variables are included in the regression model, only family resources (β= -0.29, SE=0.10, p = .002) and workload (β=.51, SE=0.23, $p<0.001$) significantly predict impact on the family (Adj. R^2= .565, F (4, 101) =35.077, p=<.001). Thus, 56.5% of the variance in impact on family scores can be explained by the combined effect of social support, family resources, perceived workload, and perceived capacity.

Table 11

Impact on Family, Regression

	Model 1				Model 2				Model 3			
	β	SE	Sig.		β	SE	Sig.		β	SE	Sig.	
Support												
Social Supp.	-0.29	.04	.002	**					-0.03	.04	.760	
Family Res.					-0.60	.08	<0.001	***	-0.29	.10	.002	**
Parental												
Workload									.51	.23	<0.001	***
Capacity										.10	.179	
Constant	-	3.55	<0.001	***	-	5.05	<0.001	***	-	7.75	<0.001	***
Adj. R^2	.077				.354				.565			

Note. *p<0.05. **p<0.01. ***p<0.001.

Research Question 4a: Do social support, family resources or parental capacity moderate the relationship between parental workload and parental depressive symptoms or impact on the family?

Multiple regression was performed with parental depressive symptoms as the outcome variable, and the model included interactions of workload with each of the four moderator variables social support, family resources, and capacity. As seen in Table 12, none of the interaction terms were statistically significant, demonstrating that neither social support, family resources, or parental capacity moderated the relationship between workload and parental depressive symptoms.

Table 12

Depressive Symptoms Regressed on Parental Workload x Support Resource Interactions (Moderation)

Model	B	(SE)	Sig.
1: workload x social support	-.02	.02	.354
2: workload x family resources	-.02	.04	.552
3: workload x capacity	-.03	.06	.543

Note: N=106. Unstandardized coefficient shown with standard errors in (). No interaction terms were statistically significant.

Multiple regression was then conducted with impact on the family as the outcome variable. This model as seen in Table 13, also included the same interaction terms, and again none of the interaction terms were statistically significant. This demonstrated that neither social support, family resources or parental capacity buffered the relationship between workload and impact on the family.

Table 13

Impact on Family Regressed on Parental Workload x Support Resource Interactions (Moderation)

Model	B	(SE)	Sig.
1: workload x social support	-.01	.01	.548
2: workload x family resources	.01	.02	.514
3: workload x capacity	.03	.03	.382

Note: N=106. Unstandardized coefficient shown with standard errors in (). No interaction terms were statistically significant.

Research Question 5: What are the relationships among the number of prescribed CMC medications, number of specialists who care for CMC, number of hours per week of care coordination and number of hours per week providing direct care and parental workload or parental depressive symptoms?

Table 14 contains the regression analysis predicting parental depressive symptoms. Regression results indicated that the model significantly predicted parental depressive symptoms (Adj. $R^2 = .104$, F (4, 101) =4.056, p=.004). The β weights indicated that only one variable, the number of weekly hours parent coordinates care (β = .38, SE = .16, p= <.001), significantly contributed to parental depressive symptoms. Thus, 10.4% of the variance in parental depressive symptom scores can be explained by the combined effect of the number of prescribed CMC medications, number of specialists who care for CMC, number of hours per week of care coordination and number of hours per week providing direct care.

Table 14

Parental Depressive Symptoms, Regression

	Model		
	β	SE	Sig.
Sociodemographics			
Medications[a]	-0.01	.30	.909
Specialists[b]	-0.06	.41	.600
Care coord.[c]	.38	.16	<.001***
Direct care[d]	-0.05	.02	.605
Constant	-	3.3	<.001***
Adj. R^2		.104	

Note. [a]Number of daily medications used by CMC. [b]Number of health care specialists who provide care for CMC. [c]Number of weekly care coordination hours provided by parents. [d]Number of weekly direct care hours parents provide to manage CMC health conditions.

*p<0.05. **p<0.01. ***p<0.001.

Table 15 contains the regression analysis predicting workload. Results indicated that the model significantly predicted workload (Adj. R^2 = .277, F (4, 101) =11.034, p=<.001. The β weights indicated that two of the four predictor variables significantly contributed to workload: the number of weekly hours that parents coordinate their CMC care (β = .22, SE =.03, p= .02) and the total number of health care specialists that care for the CMC (β = .32, SE = .09, p= <.001). Thus, 27.7% of the variance in workload can be explained by the combined effect of the number of prescribed CMC medications, number of specialists who care for the CMC, the number of hours per week that parents provide for their CMC and the number of hours per week providing direct care.

Table 15

Workload, Regression

	Model		
	β	SE	Sig.
Sociodemographics			
Medications[a]	.16	.06	.123
Specialists[b]	.32	.09	<.001***
Care coord.[c]	.22	.03	.017*
Direct care[d]	.10	.01	.239
Constant	-	.68	<.001***
Adj. R^2	.277		

Note. [a] Number of daily medications used by CMC. [b] Number of health care specialists who provide care for CMC. [c] Number of weekly care coordination hours provided by parents. [d] Number of weekly direct care hours parents provide to manage CMC health conditions.

*p<0.05. **p<0.01. ***p<0.001.

Discussion

Findings from this study indicated that adequate family resources ease workload and positively influence capacity, and a lack of family resources is a strong predictor of parental depressive symptoms. While social support does not influence workload, it does positively influence capacity. Workload is the strongest predictor of impact on the family. Moreover, when workload exceeds the capacity required to do the work needed to care for CMC, impact on the family remains significant and it accounts for more variation in parental depressive symptoms. Notably, approximately 61% of parents of CMC with CCHD reported CES-D scores of 16 or higher, consistent with clinical-level depression (Radloff, 1977).

Many support groups exist on Facebook for parents of children with medical complexity and children with CCHD. Therefore, targeted advertising for study participation on social media facilitated recruitment efforts for this study. This recruitment strategy helped overcome the challenges of in-person recruitment due to the COVID-19 pandemic. On the other hand, this strategy led to a very homogeneous sample of mostly white, married/partnered, educated women. This was corroborated by findings from a systematic review showing that recruitment through Facebook, while fast and inexpensive, does limit access to harder-to-reach demographic groups and is limited by its over representation of young white women (Whitaker et al., 2017). In addition, most participants in the current study were mothers of CMC, findings that are consistent with other studies that identify mothers as primary caregivers for children with medical complexity (Rehm, 2013). Future studies will benefit from a more diverse sample that more accurately reflects the United States demographic.

Research has shown that caring for technology dependent children affects social relationships and activities, with some parents finding it easier to stay home (Rehm, 2013). Parents of CMC have reported feeling isolated (Thomson et al., 2016). While some parents report the important role that extended family members play in offloading daily tasks, other parents have limited extended family to assist with physical care (Foster et al., 2022). According to parent reports, family members are simply not trained on the use of their CMC's medical support devices, thus leaving parents to shoulder the workload (Foster et al.). This may explain why there is no statistically significant relationship between social support and workload, in that parents of CMC rely on few others to help them with the day-to-day care, leaving little time for these families to socialize and expand their social network. The current study found that the majority

(37.7%) of parents have no one other than themselves to help provide care to their CMC, and only 32% of parents have just one other person to assist them with unpaid caregiving.

That there is no significant relationship between social support and perceived workload in this sample may be also related to the isolation that occurred due to the COVID-19 pandemic, as described in the literature (Jaspal & Breakwell, 2022). On the other hand, parents who have social support have reported that extended family members who assist with day-to-day tasks allows parents to focus on the care of their CMC (Foster et al., 2022). This may be another explanation for the absence of a significant relationship between social support and workload.

Building on prior research describing the financial challenges that parents of CMC face, this study demonstrates that a lack of adequate family resources has a significant impact on these parents. Specifically, the lack of perceived family resources had the greatest impact on predicting parental depressive symptoms, accounting for 30.8% of the variability in depressive symptoms. In addition, inadequate family resources significantly predicted impact on the family, and accounted for 35.4% of the variability in impact on family scores. These results lend strong support for universal screening of family resources and financial constraints that may further affect the mental health of parents. This is important because parents of CMC are already stressed and overburdened by their daily care responsibilities from which they get little relief. Furthermore, research has identified socioeconomic factors within racial/ethnic groups that influence the health outcomes of children with critical congenital heart disease (Peyvandi et al., 2018). Together, these findings suggest that health disparities exist for *both* parents and their CMC with CCHD when inadequate resources exist.

It is not surprising that higher capacity predicts fewer parental depressive symptoms and less impact on the family, given the frustration that parents of CMC describe when not given the proper training and preparedness for home care. To our knowledge, no studies exist that have quantitatively examined the impact of capacity on parental mental health in this medically complex patient population. This study begins to fill a knowledge gap in our understanding of how lack of preparedness and training for complex home care can influence parental mental health. Although more research is needed, these findings establish a starting point for researchers interested in quantitatively measuring perceived capacity so that interventions aimed at enhancing capacity can be evaluated.

This study found an increase in workload is significantly associated with more parental depressive symptoms but was even more significantly associated with impact on the family. In fact, workload accounted for 46.7% of the variance in impact on family scores. These results are not surprising given the parental burdens that have been described in the CMC literature to date. However, little attention has been given to quantitatively measuring workload or exploring how parental workload impacts mental health and the family. Results of this study provide a foundation from which to build upon to understand the impact of parental workload on the health and well-being of the entire family responsible for caring for medically complex children at home.

The high levels of clinically relevant depressive symptoms were notable in this study with 61.3% of participants having CES-D scores indicative of clinical-level depression (Radloff, 1977). This rate is higher than 19% of mothers with depressive symptoms whose children had chronic health conditions (Brooks et al., 2015), higher than the approximately 40% of parents with technology dependent children (Toly, et al., 2012), and higher than the 25-50% of parents with children with CCHD (Woolf-King et al., 2017). This higher rate also differs from self-reported rates of "poor or fair mental health" rates in 18.6% of parents of CMC in a recent study (Bayer et al., 2021). Nevertheless, these findings underscore the importance of establishing the incidence of depressive symptoms in parents responsible for complex care that requires a high level of attention at home to prevent morbidity and mortality. In addition, findings from this study emphasize the importance for universal screening for parental depressive symptoms so that parents can be connected to mental health supports.

The time devoted to direct caregiving was not associated with depressive symptoms or workload in this study. This is likely due to parents feeling a sense of pride and satisfaction in providing care to their CMC at home, and the gratitude that comes with staying home with their chronically ill children as reported in the literature (Rehm, 2013). Research has demonstrated that parents of CMC experience conflict over the multiple roles they play. These roles include parent, advocate, educator, and case manager (Kirk et al., 2005). However, parents defined themselves, most importantly, as a parent with a vast understanding and commitment to their children's unique needs. This also suggests that parents do not perceive direct caregiving as work per se, which could explain why no relationship was identified between perceived workload and direct caregiving activities in this study.

Care coordination was the greatest predictor of both parental depressive symptoms and workload in this study. Parents of CMC have described care coordination responsibilities as time consuming and frustrating, since it takes away from other responsibilities (Caicedo, 2014; Kuo et al., 2011; Mooney-Doyle & Lindley, 2020). One explanation for this finding is that the task of care coordination is mundane and takes time away from direct caregiving that parents of CMC have described as enjoyable and rewarding. Research has demonstrated higher parent satisfaction levels when care coordination activities are managed by a medical home or care coordination program (Mosquera et al., 2014). The more medically complex the patient, and thus more work for parents, the more time that is required by parents for care coordination tasks (Kuo et al., 2011), a finding which is in keeping with findings from this study.

The current study also found that higher numbers of health care specialists were associated with higher parental workload and depressive symptoms. This is corroborated with qualitative research exploring the demands of parents with CMC. These parents described the task of ensuring that all health care providers are communicating with one another as stressful, frustrating, and burdensome, pointing out that poor communication between health care providers can lead to consequential delays in care (Golden & Nageswaran, 2012). The task of information management is a critical but time-consuming responsibility. So, while a large team of health care specialists for CMC is necessary, it directly impacts both the workload and the mental health of parents in this study.

It is unclear why social support, family resources, and parental capacity did not moderate the relationship between parental workload and parental depressive symptoms or impact on the family. The sample size of 106 may have been one factor influencing these results, in which case larger sample sizes in future studies may be elucidating. Mediation analyses may be a logical next step using available data from this study.

Perceived social support and perceived family resources are contextual influences that can hinder or support parents' management of their CMCs' conditions (Knafl et al., 2012). Both variables were significantly correlated with workload and capacity in this study. The greater the workload, the more likely the impact on the family and for depressive symptoms to occur, while on the other hand as capacity increased, the likelihood of parental depressive symptoms and impact on the family diminished. Collectively, social support, family resources, workload, and capacity explained 32.4% of the variance in depressive symptoms, and 56.5% of the variance in impact on the family. These findings support the usefulness of the theoretical framework used in this study to understand the challenges faced by parents of CMC and their effect on parental mental health and on the family overall.

Limitations

The results of this study must be considered in the context of several limitations. The first is related to the representativeness of the sample. Recruitment efforts were done solely online which can affect the representativeness of the sample. Notably, parent participants were largely homogeneous with regard to marital status, race, education, and gender which limits the generalization of these findings. Although this was a national sample, the non-random sample consisted of a more than 90% white, non-Hispanic, married female participant group . Random sampling would limit biases and have the potential to strengthen future study findings.

A second limitation is that all questionnaires and instruments were self-reported and could not be confirmed by in-person observations or medical records. Self-reporting may have contributed to recall and response bias. Face-to-face interviewing may have provided greater clarity and accuracy of information shared by parent participants.

The use of a cross-sectional study design was another study limitation. The nature of this study design limits our understanding of the temporal order of the variables. Longitudinal studies could evaluate changes in perceived workload and capacity over time, and how experience in managing the care of CMC over time can impact workload, capacity, depressive symptoms, and impact on the family.

Lastly, it is impossible to know how the COVID-19 pandemic has affected parents of CMC, as there are no available measures to differentiate between depressive symptoms due to providing care to CMC versus depressive symptoms due to the pandemic. COVID-19 has contributed to depression and has presented barriers to adequate resources that may have otherwise been in place prior to the pandemic (Dhiman et al., 2020; Racine et al., 2021).

Strengths

Despite the noted limitations of this study, there were several strengths. This study adds to the limited body of literature describing the influence of social support, family resources, workload, and capacity on parental depressive symptoms and impact on the family in children with medical complexity. This study was the first to examine parents' perceived workload and capacity to care for CMC and its impact on the family, as well as its impact on parental depressive symptoms. This study begins to address a gap in the literature that is recognized by parents and providers who care for children with medical complexity, and who have asked for further research to better address the workload and demand required to care for CMC. Moreover, all of the instruments used for this study had adequate internal consistency reliability. In addition, the target sample, while small, was achieved and the study attracted a national participant sample. Parent participants were engaged and eager to participate as noted by their responses to initial postings seeking study participants. There was a notable sense of gratitude by parent participants for the undertaking of the research study. Finally, parents were eager to clarify follow-up questions via email, which led to little or no missing data, further strengthening this study.

Implications for Nursing Practice, Policy, and Future Research

The findings of this study have several implications for nursing practice, policy, and future research. First, nurses must be educated about the potential high rates of depressive symptoms in parents of CMC. Nurses are in a prime position to assess for adequate social supports, family resources and parental depressive symptoms. Such screening is imperative to assist families in obtaining available mental health support and financial resources when they are most needed. Given the links between workload, capacity, depressive symptoms and impact on the family, nurses should focus efforts on developing interventions to improve parents' abilities to meet the demands for delivering home care with a particular emphasis on including parents in all aspects of research. This strategy ensures that targeted interventions are informed by parents' experiences and needs.

The findings of this study highlight the need for the expansion of medical homes, beginning with education around the definition and importance that medical homes play for CMC and their families. Universally providing care coordination services must become the care standard for CMC, and we are heading in the right direction with the passing of recent legislation. The Medicaid Services Investment and Accountability Act of 2019 (introduced as H.R. 1839) was signed into law on April 18, 2019 (govtrack.us, 2019). This law expands a state option for medical homes for CMC and provides additional funding for demonstration projects such as the one described by Mosquera and colleagues (2014). However, participation is not mandatory, and implementation is being left to participating states. Additionally, value-based payment systems that reward clinicians and institutions for providing high quality care must be advocated for. While additional funding by way of the recently passed legislation will help, clinicians and researchers committed to outstanding care for CMC must establish baseline metrics and outcomes beyond readmission and ED visit rates to include caregiver satisfaction, caregiver and patient quality of life, and social integration that can be prospectively examined.

Finally, respite care and work-hour safety standards for parents of CMC must be developed with advocacy and policy support. This is important based on what is known about the unpaid care hours that parents provide for medically fragile children. CMC experts suggest that these parents should not work to care for their children more than 9 consecutive hours based on what is known about the association of adverse events and performance-impairing fatigue in other safety-sensitive occupations such as nursing, transportation, and emergency services (Schall et al., 2020; Williamson et al., 2011). These experts argue that the work provided by parents to care for their CMC should be held to the same safety standards used in other high-risk fields to prevent exhaustion and burn out.

Several directions for future research can be gleaned from this study. In addition to developing and testing interventions to minimize workload and facilitate capacity, further study is needed to understand factors that buffer parental depressive symptoms and impact on the family. Studies exploring resilience and its protective role in mitigating depression are needed, since resilience influences parents' stress response as well as the health of parents and their children (Lisanti, 2018).

Conclusions

The findings from this study add to the limited body of knowledge about the relationship between the perceived workload that parents of CMC experience and their mental health. This study, guided by a derived theoretical model, was the first to explore relationships between social support, family resources, workload, capacity, and how these factors impact parental depressive symptoms and families of CMC. Moreover, this study begins to fill a critical knowledge gap about the prevalence of depressive symptoms in parents of CMC with CCHD, particularly when workload outweighs capacity and when family resources are inadequate. These findings highlight the need for universal screening for social support, family resources, and depressive symptoms. In addition, these findings emphasize the need for policies and care models that reduce workload while building capacity to manage the workload required to care for CMC at home. Medical homes that provide care coordination to these parents are one effective way to minimize workload, yet they are not a widely available resource. Until medical homes universally exist for all CMC, interventions aimed at building capacity and minimizing workload for parents are needed so that the health and well-being of CMC and their parents can be strengthened.

REFERENCES

Allshouse, C., Comeau, M., Rodgers, R., & Wells, N. (2018). Families of children with medical complexity: A view from the front lines. *Pediatrics, 141*(Suppl 3), S195-s201. https://doi.org/10.1542/peds.2017-1284D

Altman, L., Zurynski, Y., Breen, C., Hoffmann, T., & Woolfenden, S. (2018). A qualitative study of health care providers' perceptions and experiences of working together to care for children with medical complexity (CMC). *BMC Health Services Research, 18*(1), 70. https://doi.org/10.1186/s12913-018-2857-8

Avritscher, E. B. C., Mosquera, R. A., Tyson, J. E., Pedroza, C., Samuels, C. L., Harris, T. S., Gomez-Rubio, A., Navarro, F. A., Moody, S. B., & Beyda, R. M. (2019). Post-trial sustainability and scalability of the benefits of a medical home for high-risk children with medical complexity. *Journal of Pediatrics, 206*, 232-239.e233. https://doi.org/10.1016/j.jpeds.2018.10.035

Barnert, E. S., Coller, R. J., Nelson, B. B., Thompson, L. R., Klitzner, T. S., Szilagyi, M., Breck, A. M., & Chung, P. J. (2018). A healthy life for a child with medical complexity: 10 domains for conceptualizing health. *Pediatrics, 142*(3).

Barnert, E. S., Coller, R. J., Nelson, B. B., Thompson, L. R., Tran, J., Chan, V., Padilla, C., Klitzner, T. S., Szilagyi, M., & Chung, P. J. (2019). Key population health outcomes for children with medical complexity: A systematic review. *Maternal and Child Health Journal, 23*(9), 1167-1176. https://doi.org/10.1007/s10995-019-02752-1

Bayer, N. D., Wang, H., Yu, J. A., Kuo, D. Z., Halterman, J. S., & Li, Y. (2021). A national mental health profile of parents of children with medical complexity. *Pediatrics*, *148*(2). https://doi.org/10.1542/peds.2020-023358

Berry, J. G., Agrawal, R. K., Cohen, E., & Kuo, D. Z. (2013). The landscape of medical care for children with medical complexity. *Overland Park, KS: Children's Hospital Association, 7.*

Berry, J. G., Hall, M., Neff, J., Goodman, D., Cohen, E., Agrawal, R., Kuo, D., & Feudtner, C. (2014). Children with medical complexity and medicaid: Spending and cost savings. *Health Affairs, 33*(12), 2199-2206. https://doi.org/10.1377/hlthaff.2014.0828

Brooks, J. L., Beil, H., & Beeber, L. S. (2015). Maternal depressive symptoms and healthcare expenditures for publicly insured children with chronic health conditions. *Maternal and Child Health Journal, 19*(4), 790-797.

Burns, K. H., Casey, P. H., Lyle, R. E., Bird, T. M., Fussell, J. J., & Robbins, J. M. (2010). Increasing prevalence of medically complex children in us hospitals. *Pediatrics, 126*(4), 638-646. https://doi.org/10.1542/peds.2009-1658

Cady, R., & Belew, J. (2017). Parent perspective on care coordination services for their child with medical complexity. *Children, 4*(6), 45.

Caicedo, C. (2014). Families with special needs children: Family health, functioning, and care burden. *Journal of the American Psychiatric Nurses Association, 20*(6), 398-407. https://doi.org/10.1177/1078390314561326

Centers for Disease Control. (2020). *Congenital heart defects*. Retrieved October 1, 2020 from https://www.cdc.gov/ncbddd/heartdefects/cchd-facts.html

Cohen, E., Berry, J. G., Camacho, X., Anderson, G., Wodchis, W., & Guttmann, A. (2012). Patterns and costs of health care use of children with medical complexity. *Pediatrics, 130*(6), e1463-1470. https://doi.org/10.1542/peds.2012-0175

Cohen, E., Berry, J. G., Sanders, L., Schor, E. L., & Wise, P. H. (2018). Status complexicus? The emergence of pediatric complex care. *Pediatrics, 141*(Suppl 3), S202-s211. https://doi.org/10.1542/peds.2017-1284E

Cohen, E., Kuo, D. Z., Agrawal, R., Berry, J. G., Bhagat, S. K., Simon, T. D., & Srivastava, R. (2011). Children with medical complexity: An emerging population for clinical and research initiatives. *Pediatrics, 127*(3), 529-538. https://doi.org/10.1542/peds.2010-0910

Cohn, L. N., Pechlivanoglou, P., Lee, Y., Mahant, S., Orkin, J., Marson, A., & Cohen, E. (2020). Health outcomes of parents of children with chronic illness: A systematic review and meta-analysis. *The Journal of Pediatrics*, 218, 166-177. https://doi.org/10.1016/j.peds.2019.10.068

Cousino, M. K., & Hazen, R. A. (2013). Parenting stress among caregivers of children with chronic illness: A systematic review. *Journal of Pediatric Psychology, 38*(8), 809-828. https://doi.org/10.1093/jpepsy/jst049

Dhiman, S., Sahu, P.K., Reed, W. R., Ganesh, G. S., Goyal, R. K., & Jain, S. (2020). Impact of covid-19 outbreak on mental health and perceived strain among caregivers tending children with special needs. Res*earch in Developmental Disabilities, 107*, 1-15. https://doi.org/10.1016/j.ridd.2020.103790

Faul, F., Erdfelder, E., Lang, A.-G. & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods, 39*, 175-191.

Fayed, N., Guttmann, A., Chiu, A., Gardecki, M., Orkin, J., Hamid, J. S., Major, N., Lim, A., & Cohen, E. (2019). Family-provider consensus outcomes for children with medical complexity. *Developmental Medicine and Child Neurology, 61*(9), 1093-1100. https://doi.org/10.1111/dmcn.14110

Foster, C.C., Shaunfield, S., Black, L.E., Labellarte, P.Z., & Davis, M.M. (2022). Improving support for care at home: Parental needs and preferences when caring for children with medical complexity. *Journal of Pediatric Health Care, 36*(2),154-164. https://doi.org/10.1016/j.pedhc.2020.08.005

Golden, S.L. & Nageswaran, S. Caregiver voices: Coordinating care for children with complex chronic conditions. *Clinical Pediatrics, 51*(8), 723-729. https://doi.10.1177/0009922812445920

GovTrack.us. (2019). H.R. 1226 – 116th Congress: ACE Kids Act of 2019. Retrieved from https://www.govtrack.us/congress/bills/116/hr1226

Gibson-Young, L., Turner-Henson, A., Gerald, L. B., Vance, D. E., & Lozano, D. (2014). The relationships among family management behaviors and asthma morbidity in maternal caregivers of children with asthma. *Journal of Family Nursing, 20*(4), 442-461. https://doi.org/10.1177/1074840714552845

Hammen, C. (2004). Stress and depression. *Annual Review of Clinical Psychology, 1*(1), 293-319. https://doi.org/10.1146/annurev.clinpsy.1.102803.143938

Hofacer, R. D., Panatopoulos, A., Vineyard, J., Tivis, R., Nguyen, E., Jingjing, N., & Lindsay, R. P. (2019). Clinical care coordination in medically complex pediatric cases: Results from the national survey of children with special health care needs. *Global Pediatric Health, 6*, 2333794x19847911. https://doi.org/10.1177/2333794x19847911

Hudson, S. M., Newman, S. D., Hester, W. H., Magwood, G. S., Mueller, M., & Laken, M. A. (2014). Factors influencing hospital admissions and emergency department visits among children with complex chronic conditions: A qualitative study of parents' and providers' perspectives. *Issues in Comprehensive Pediatric Nursing , 37*(1), 61-80. https://doi.org/10.3109/01460862.2013.855844

Jaspal, R. & Breakwell, G.M. (2022). Socio-economic inequalities in social network, loneliness and mental health during the COVID-19 pandemic. *International Journal of Social Psychiatry, 68*(1), 155-165. https://doi.org/10.1177/0020764020976694

Javalkar, K., Rak, E., Phillips, A., Haberman, C., Ferris, M., & Van Tilburg, M. (2017). Predictors of caregiver burden among mothers of children with chronic conditions. *Children, 4*(5), 39.

Khanna, A. K., Prabhakaran, A., Patel, P., Ganjiwale, J. D., & Nimbalkar, S. M. (2015). Social, psychological and financial burden on caregivers of children with chronic illness: A cross-sectional study. *Indian Journal of Pediatrics, 82*(11), 1006-1011. https://doi.org/10.1007/s12098-015-1762-y

Kim, I., Ekas, N. V., & Hock, R. (2016). Associations between child behavior problems, family management, and depressive symptoms for mothers of children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 26*, 80-90.

Kirk, S., Glendinning, C., & Callery, P. (2005). Parent or nurse? The experience of being the parent of a technology-dependent child. *Journal of Advanced Nursing*, *51*(5), 456-464. https://doi.org/10.1111/j.1365-2648.2005.03522.x

Knafl, K. A., & Deatrick, J. A. (2003). Further refinement of the family management style framework. *Journal of Family Nursing, 9*(3), 232-256.

Knafl, K. A., Deatrick, J. A., & Havill, N. L. (2012). Continued development of the family management style framework. *Journal of Family Nursing, 18*(1), 11-34. https://doi.org/10.1177/1074840711427294

Knafl, K. A., Deatrick, J. A., Knafl, G. J., Gallo, A. M., Grey, M., & Dixon, J. (2013). Patterns of family management of childhood chronic conditions and their relationship to child and family functioning. *Journal of Pediatric Nursing, 28*(6), 523-535. https://doi.org/10.1016/j.pedn.2013.03.006

Kuo, D. Z., Cohen, E., Agrawal, R., Berry, J. G., & Casey, P. H. (2011). A national profile of caregiver challenges among more medically complex children with special health care needs. *Archives of Pediatrics and Adolescent Medicine, 165*(11), 1020-1026. https://doi.org/10.1001/archpediatrics.2011.172

Kuo, D. Z., Goudie, A., Cohen, E., Houtrow, A., Agrawal, R., Carle, A. C., & Wells, N. (2014). Inequities in health care needs for children with medical complexity. *Health Affairs, 33*(12), 2190-2198.

Kuo, D.Z. & Houtrow, A.J. (2016). Recognition and management of medical complexity. *Pediatrics, 138*(6), 132-144. https://doi.org/10.1542/peds.2016-3021

Kuo, D. Z., McAllister, J. W., Rossignol, L., Turchi, R. M., & Stille, C. J. (2018). Care coordination for children with medical complexity: Whose care is it, anyway? *Pediatrics, 141*(Suppl 3), S224-s232. https://doi.org/10.1542/peds.2017-1284G

Lassman, D., Hartman, M., Washington, B., Andrews, K., & Catlin, A. (2014). US health spending trends by age and gender: Selected years 2002–10. *Health affairs, 33*(5), 815-822.

Lawoko, S., & Soares, J. J. (2003). Quality of life among parents of children with congenital heart disease, parents of children with other diseases and parents of healthy children. *Quality of Life Research, 12*(6), 655-666.

Leahy-Warren, P., McCarthy, G., & Corcoran, P. (2012). First-time mothers: Social support, maternal parental self-efficacy and postnatal depression. *Journal of Clinical Nursing, 21*(3-4), 388-397.

Lisanti, A. J. (2018). Parental stress and resilience in chd: A new frontier for health disparities research. *Cardiology in the Young, 28*(9), 1142-1150.

Mandic, C. G., Johaningsmeir, S., Corden, T. E., Earle, A., Acevedo-Garcia, D., & Gordon, J. B. (2017). Impact of caring for children with medical complexity on parents' employment and time. *Community, Work & Family, 20*(4), 444-458. https://doi.org/10.1080/13668803.2016.1202195

Mesman, G. R., Kuo, D. Z., Carroll, J. L., & Ward, W. L. (2013). The impact of technology dependence on children and their families. *Journal of Pediatric*

Health Care, 27(6), 451-459. https://doi.org/https://doi.org/10.1016/j.pedhc.2012.05.003

Mooney-Doyle, K., & Lindley, L. C. (2019). The association between poverty and family financial challenges of caring for medically complex children. *Nursing Economics, 37*(4), 198-208.

Mooney-Doyle, K., & Lindley, L. C. (2020). Family and child characteristics associated with caregiver challenges for medically complex children. *Family and Community Health, 43*(1), 74-81.

Mosquera, R. A., Avritscher, E. B., Samuels, C. L., Harris, T. S., Pedroza, C., Evans, P., Navarro, F., Wootton, S. H., Pacheco, S., Clifton, G., Moody, S., Franzini, L., Zupancic, J., & Tyson, J. E. (2014). Effect of an enhanced medical home on serious illness and cost of care among high-risk children with chronic illness: A randomized clinical trial. *JAMA, 312*(24), 2640-2648. https://doi.org/10.1001/jama.2014.16419

Nelson, B. B., Coller, R. J., Saenz, A. A., Chung, P. J., Kaplan, A., Lerner, C. F., & Klitzner, T. S. (2016). How avoidable are hospitalizations for children with medical complexity? Understanding parent perspectives. *Academic Pediatrics, 16*(6), 579-586.

Ni, Z. H., Lv, H. T., Ding, S., & Yao, W. Y. (2019). Home care experience and nursing needs of caregivers of children undergoing congenital heart disease operations: A qualitative descriptive study. *PloS One, 14*(3), e0213154. https://doi.org/10.1371/journal.pone.0213154

Nygard, C., & Clancy, A. (2018). Unsung heroes, flying blind-a metasynthesis of parents' experiences of caring for children with special health-care needs at home. *Journal of Clinical Nursing, 27*(15-16), 3179-3196. https://doi.org/10.1111/jocn.14512

Peyvandi, S., Baer, R.J., Moon-Grady, A.J., Oltman, S.P., Chambers, C.D., Norton, M.E., Rajagopal, S., Ryckman, K.K., Jeliffe-Pawlowski, L.L., & Steurer, M.A. Socioeconomic mediators of racial and ethnic disparities in congenital heart disease outcomes: A population-based study in California. *Journal of the American Heart Association, 7*, e010342. https://doi.10.1161/jaha.118.010342

Pierce, M., Hope, H. F., Kolade, A., Gellatly, J., Osam, C. S., Perchard, R., Kosidou, K., Dalman, C., Morgan, V., Di Prinzio, P., & Abel, K. M. (2019). Effects of parental mental illness on children's physical health: Systematic review and meta-analysis. *British Journal of Psychiatry*, 1-10. https://doi.org/10.1192/bjp.2019.216

Pinquart, M. (2018). Parenting stress in caregivers of children with chronic physical condition-a meta-analysis. *Stress and Health, 34*(2), 197-207. https://doi.org/10.1002/smi.2780

Racine, N., Hetherington, E., McArthur, B., McDonald, S., Edwards, S., Tough, S., & Madigan, S. (2021). Maternal depressive and anxiety symptoms before and during the covid-19 pandemic in canada: A longitudinal analysis. *The Lancet Psychiatry, 8*(5), 405-415. https://doi.org/10.1016/S2215-0366(21)00074-2

Radloff, L.S. (1977). The ces-d scale: A self-report depression scale for research in the general population. *Applied Psychological Measurement, 1*(3), 385-401. https://doi.org/10.1177/014662167700100306

Razzaghi, H., Oster, M., & Reefhuis, J. (2015). Long-term outcomes in children with congenital heart disease: National health interview survey. *The Journal of Pediatrics, 166*(1), 119-124.e111. https://doi.org/https://doi.org/10.1016/j.jpeds.2014.09.006

Rehm, R. (2013). Nursing's contribution to research about parenting children with complex chronic conditions: An integrative reivew, 2002 to 2012. *Nursing Outlook, 61*, 266-290. http://dx.doi.org/10.1016/j.outlook.2013.03.008.

Rempel, G. R., Blythe, C., Rogers, L. G., & Ravindran, V. (2012). The process of family management when a baby is diagnosed with a lethal congenital condition. *Journal of Family Nursing, 18*(1), 35-64.

Romley, J. A., Shah, A. K., Chung, P. J., Elliott, M. N., Vestal, K. D., & Schuster, M. A. (2017). Family-provided health care for children with special health care needs. *Pediatrics, 139*(1). https://doi.org/10.1542/peds.2016-1287

Ruggiero, K., Pratt, P., & Antonelli, R. (2019). Improving outcomes through care coordination: Measuring care coordination of nurse practitioners. *Journal of the American Association of Nurse Practitioners, 31*(8), 476-481. https://doi.org/10.1097/jxx.0000000000000276

Schall, T. E., Foster, C. C., & Feudtner, C. (2020). Safe work-hour standards for parents of children with medical complexity. *JAMA Pediatrics, 174*(1), 7-8. https://doi.org/10.1001/jamapediatrics.2019.4003

Scherer, N., Verhey, I., & Kuper, H. (2019). Depression and anxiety in parents of children with intellectual and developmental disabilities: A systematic review and meta-analysis. *PloS One, 14*(7).

Shippee, N. D., Shah, N. D., May, C. R., Mair, F. S., & Montori, V. M. (2012). Cumulative complexity: A functional, patient-centered model of patient complexity can improve research and practice. *Journal of Clinical Epidemiology, 65*(10), 1041-1051.

Shumskiy, I., Richardson, T., Brar, S., Hall, M., Cox, J., Crofton, C., Peltz, A., Samuels-Kalow, M., Alpern, E. R., Neuman, M. I., & Berry, J. G. (2018). Well-child visits of medicaid-insured children with medical complexity. *Journal of Pediatrics, 199*, 223-230.e222. https://doi.org/10.1016/j.jpeds.2018.04.003

Simon, T. D., Berry, J., Feudtner, C., Stone, B. L., Sheng, X., Bratton, S. L., Dean, J. M., & Srivastava, R. (2010). Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics, 126*(4), 647-655. https://doi.org/10.1542/peds.2009-3266

Spratling, R. (2017). Understanding the health care utilization of children who require medical technology: A descriptive study of children who require tracheostomies. *Applied Nursing Research, 34*, 62-65.

Stein, R. E. K., & Jessop, D. J. (2003). The impact on family scale revisited: Further psychometric data. *Developmental and Behavioral Pediatrics, 24*, 1, 9-16.

Tak, Y. R., & McCubbin, M. (2002). Family stress, perceived social support and coping following the diagnosis of a child's congenital heart disease. *Journal of Advanced Nursing, 39*(2), 190-198.

Thomson, J., Shah, S. S., Simmons, J. M., Sauers-Ford, H. S., Brunswick, S., Hall, D., Kahn, R. S., & Beck, A. F. (2016). Financial and social hardships in families of children with medical complexity. *Journal of Pediatrics, 172*, 187-193.e181. https://doi.org/10.1016/j.jpeds.2016.01.049

Toledano-Toledano, F., & Domínguez-Guedea, M. T. (2019). Psychosocial factors related with caregiver burden among families of children with chronic conditions. *Biopsychosocial Medicine, 13*(1), 6.

Toly, V.B., Musil, C.M., & Carl, J.C. (2012). A longitudinal study of families with technology-dependent children. *Research in Nursing & Health, 35*(1), 40-54. https://doi.org/10.1002/nur.21454

Van Der Linde, D., Konings, E. E., Slager, M. A., Witsenburg, M., Helbing, W. A., Takkenberg, J. J., & Roos-Hesselink, J. W. (2011). Birth prevalence of congenital heart disease worldwide: A systematic review and meta-analysis. *Journal of the American College of Cardiology, 58*(21), 2241-2247.

Van Horn, M. L., Bellis, J. M. & Snyder, S. W. (2001). Family resource scale-revised: Psychometrics and validation of a measure of family resources in a sample of low-income families. *Journal of Psychoeducational Assessment, 19,* 54-68.

Vessey, J. A., DiFazio, R. L., Strout, T. D., & Snyder, B. D. (2017). Impact of non-medical out-of-pocket expenses on families of children with cerebral palsy following orthopaedic surgery. *Journal of Pediatric Nursing, 37*, 101-107. https://doi.org/10.1016/j.pedn.2017.07.006

Walker, L. O., & Avant, K. C. (2019). *Strategies for theory construction in nursing* (6th ed.). Pearson.

Weinert, C. (2003). Measuring social support: PRQ2000. *Measurement of Nursing Outcomes, 3,* 161-172.

Whitaker, C., Stevelink, S., & Fear, N. (2017). The use of facebook in recruiting participants for health research purposes: A systematic review. Journal of Medical Internet Research,19(8), 1-11. https://doi:10.2196/jmir.org/2017/8/e290

White-Traut, R., Rankin, K., Fabiyi, C., Liu, L., Cheung, I., & Norr, K. (2017). Maternal characteristics associated with social support in at-risk mothers of premature infants. *Journal of Obstetric, Gynecologic & Neonatal Nursing, 46*(6), 824-833.

Whiting, M. (2013). Impact, meaning and need for help and support: The experience of parents caring for children with disabilities, life-limiting/life-threatening illness or technology dependence. *Journal of Child Health Care, 17*(1), 92-108.

Whiting, M. (2014). Support requirements of parents caring for a child with disability and complex health needs. *Nursing Children and Young People, 26*(4).

Williamson, A., Lombardi, D. A., Folkard, S., Stutts, J., Courtney, T. K., & Connor, J. L. (2011). The link between fatigue and safety. *Accident Analysis & Prevention, 43*(2), 498-515. https://doi.org/https://doi.org/10.1016/j.aap.2009.11.011

Woodgate, R. L., Edwards, M., Ripat, J. D., Borton, B., & Rempel, G. (2015). Intense parenting: A qualitative study detailing the experiences of parenting children with complex care needs. *BMC Pediatrics, 15*(1), 197.

Woolf-King, S. E., Anger, A., Arnold, E. A., Weiss, S. J., & Teitel, D. (2017). Mental health among parents of children with critical congenital heart defects: A systematic review. *Journal of the American Heart Association, 6*(2). https://doi.org/10.1161/jaha.116.004862

Zhang, Y., Wei, M., Shen, N., & Zhang, Y. (2015). Identifying factors related to family management during the coping process of families with childhood chronic conditions: A multi-site study. *Journal of Pediatric Nursing, 30*(1), 160-173.

APPENDIX A

A Theory for Understanding Parental Workload and Capacity to Care for Children with Medical Complexity[1]

Published March 2022 in *Research and Theory for Nursing Practice*

[1] Lawrence, P.R. & Spratling, R. (2022). A theory for understanding parental workload and capacity to care for children with medical complexity. *Research and Theory for Nursing Practice, 36*(1), 34-46. http://dx.doi.org/10.1891/RTNP-2021-0026

Abstract

Background and Purpose: Children with medical complexity (CMC) experience poor health outcomes despite the high cost of care, and their parents face challenges in providing complex care. Poor health outcomes may be related to an imbalance between parental demands to manage care and their ability to meet the demands needed to provide complex care. However, this phenomenon has not been explored. In addition, much of the existing research focused on CMC lacks an overarching theoretical framework. The purpose of this article is to outline factors that impact families of CMC described in the literature. This article proposes a modified framework using theory derivation, which highlights the concepts of parental workload and capacity and demonstrates how they are related to CMC health. **Methods:** A revised theoretical framework using theory derivation by Walker and Avant is presented using findings from the CMC literature that most affect the parents of these children. **Results:** Applying content from two existing theories using concepts of relevance results in a framework that provides richer insight into the relationship between parental workload and parental capacity, particularly when parental workload outweighs parental capacity. This framework allows for the examination of how an imbalance between workload and capacity impacts CMC health outcomes. **Implications for Practice:** Although further study is needed to test the proposed theory, the framework can be used to examine these relationships with hopes of developing interventions to decrease parental workload and enhance parental ability.

Keywords: capacity, children with medical complexity (CMC), parent, theory derivation, workload

A Theory for Understanding Parental Workload and Capacity to Care for Children with Medical Complexity

Introduction

Children with medical complexity (CMC) have one or more chronic, lifelong health conditions that can be severe and have substantial health service needs. In addition, CMC have functional limitations and high health care use, placing these children at risk for poor health and family outcomes (Cohen et al., 2011). Much of the extant literature on CMC and their families has focused on describing the drivers of CMC care costs (Berry et al., 2014), poor CMC health outcomes (Feinstein et al., 2014; Shumskiy et al., 2018), and the challenges that parents face in providing necessary care for their children (Allshouse et al., 2018; Mooney-Doyle & Lindley, 2020). Parental demands combined with challenges in parents' ability to manage the care of their child's complex needs may contribute to these poor health outcomes. However, there is a paucity of research examining these relationships. Notably, CMC enrolled in comprehensive care programs that assist parents in coordinating care have dramatically better health outcomes (Avritscher et al., 2019; Mosquera et al., 2014), suggesting that parental workload and ability to manage complex needs of these children are relevant concepts for improving CMC health. Moreover, theoretical frameworks are rarely included in studies focused on this patient population, underscoring the need for exploring frameworks to guide researchers invested in improving the health and well-being of CMC and their families.

Using a theoretical framework to guide research provides a roadmap that can be used to describe phenomena, predict care outcomes, and explain research findings

(Walker & Avant, 2019). The Family Management Style Framework (FMSF) provides insight into how families manage care for chronically ill children (Knafl et al., 1996; Knafl et al., 2012). This theory contains dimensions of ability to manage illness and how easy or challenging the illness is to manage. The Cumulative Complexity Model (CCM) directly addresses the concepts of workload and capacity in adults with chronic illness. The CCM posits that when an imbalance exists between demand and ability (where workload exceeds capacity), health outcomes worsen (Shippee et al., 2012). These two middle-range theories were identified as potentially meaningful in understanding how parents' demands (workload) and abilities (capacity) in caring for children with medical complexity impact health outcomes of these children. The concepts of parental workload and capacity from the CMC literature were used to propose a revised theory using theory derivation by Walker and Avant (2019). This revised theory helps researchers and health care providers understand how an imbalance between parental workload and capacity can affect CMC health outcomes.

Children with Medical Complexity: Definition and Prevalence

While representing just 0.4% of all children, CMC health care expenses total 40% of all pediatric hospital costs (Cohen et al., 2012). In addition, the number of CMC has grown as a result of improved survival rates of conditions that were once fatal (Kuo et al., 2011). The term CMC is often used interchangeably with other terms including "medically complex", "chronically ill", and "children with special health care needs" to signify the chronicity of illness as well as the *time, resources and expertise* needed by parents to manage the care of these children (Cohen et al., 2018). The disproportionate care costs of CMC are partially due to the complexity of day-to-day care. A number of

CMC characteristics influence the complexity of care. For instance, CMC require inpatient and outpatient care from a variety of health care specialists and rely on a number of medications and therapies. CMC require surgeries, procedures, and life-sustaining technologies, and experience acute illnesses and exacerbations of chronic illnesses that require unique health care management (Berry et al., 2013). A concept analysis of the term "children with medical complexity" confirms that these children were previously considered so complex that they required long-term inpatient care. CMC now live at home with their families who provide care (Rogers et al., 2021). While studies have described the demands and burdens that parents encounter when caring for CMC, no research studies examine the relationships between parental workload, parental capacity, and CMC health outcomes. Identifying patterns of a family's ability to manage care for a chronically ill child will lead to a better understanding of family adaptation in the face of adversity and to interventions that improve health outcomes for chronically ill children (Knafl et al., 2013).

Relevant Concepts Affecting Parents of CMC

Workload to Manage CMC Care

Workload is defined as the demands placed on parents to manage the day-to-day care of CMC, which has a significant, daily impact on families (Ward et al., 2015). Much of the CMC literature describes parents as burdened by this time-intensive and exhausting role despite the many joys that the role brings (Ward et al.). Moreover, CMC parents describe the medical system as complex and difficult to navigate (Cady & Belew, 2017), compounding the workload parents experience.

Workload Due to Direct Care

Parents provide the majority of direct care at home for CMC who require specialized attention. Daily care can include procedures, medical equipment use, and complex medication schedules (Rempel et al., 2012; Spratling, 2017). Approximately 5.6 million special health care needs children receive an annual 1.5 billion hours of family-provided care at home, with parents providing the majority of direct care to CMC, sometimes for 20 or more hours a week (Romley et al., 2017). High amounts of caregiving time are associated with decreased quality of life for parents (Lawoko & Soares, 2003). Moreover, caregiving demands are associated with more stress-related illness in parents (Cousino & Hazen, 2013), underscoring the detrimental effects that this workload has on parents of CMC.

Workload Due to Care Coordination

In addition to the time parents spend providing direct care, they also spend between 3.9 and 6.6 hours per week coordinating care (Caicedo, 2014; Romley et al., 2017). Parental care coordination has the potential to mitigate crisis-driven overtreatment of CMC, especially when seamless parental communication occurs. However, this degree of parental effort takes *time and skills* that are typically provided by nurses and other health care providers in program-based medical homes (Ruggiero et al., 2019). More than 66% of parents of children with special health care needs receive no clinic-based support to coordinate care (Hofacer et al., 2019). This leaves parents to manage the workload of coordinating care by themselves.

Workload Due to Health Care Access and Utilization

Parental workload consists of spending considerable time taking CMC to appointments, attending a median of 11 to 15 specialty clinics for CMC each year (Kuo et al., 2011). Some parents need to travel frequently to health care specialists. These appointments are sometimes hundreds of miles away because specialists are often part of an urban, tertiary care center (Cady & Belew, 2017). Appointments far from home place further strain and workload on parents due to lost work hours because of the time needed to attend medical appointments with their CMC. Health care specialists prescribe disease-managing medications for CMC, resulting in multiple medications and complex medical regimens (Aboneh & Chui, 2017; Cohen et al., 2011). This puts CMC at higher risk for home medication errors, which in turn leads to emergency room visits and hospitalizations (Feinstein et al., 2014). Children with sickle cell disease and seizure disorders with complex medication schedules were found to experience a 22% home medication error rate, and at least one medication error was noted in 95% of cases when a medication reminder system such as calendar or pill box, wasn't used (Walsh et al., 2011).

Workload Due to Financial Hardship

Some researchers have included financial strains experienced by parents of CMC as a significant challenge (Kuo et al., 2011; Mooney-Doyle et al., 2019; Thomson et al., 2016). These financial demands are inextricably linked to the time required to care for their CMC. Researchers conducting a concept analysis on caregiver burden note that financial hardship is an antecedent (Liu et al., 2020). Kuo et al. (2011) finds that 46.3% of parents paid more than $1,000 in the prior year toward out-of-pocket health care-related costs, and 56.8% of parents reported financial difficulties due to the costs of unreimbursed CMC care. More than 50% of families needed a family member to stop working in order to provide direct care to their CMC. Nearly 49% of parents reported not having enough income to cover CMC medical expenses. CMC family members were more likely to report not filling a prescription or not seeing a dentist or physician due to financial difficulties (Thomson et al., 2016). Similarly, Mooney-Doyle and Lindley (2019) find that poverty is significantly associated with both the financial challenges experienced by families as well as with a family member stopping work outside the home to care for their medically complex child. It is clear from these studies, some of which use large, nationally representative samples, that financial burdens experienced by the parents of CMC are substantial and directly related to the care that CMC require. Parental workload to manage their CMC condition consists of the time, skills, and resources necessary to manage a health condition, including financial resources that when in short supply, can add further burden.

Capacity to Manage CMC Care

Capacity is defined as the ability, knowledge, readiness, or competency needed to manage a health condition (Shippee et al., 2012). Regardless of parents' ability to do so, they are required to manage complex treatments of their CMC at home. Care management can include sophisticated medication regimens, preparing complex diets, or tracheostomy and feeding tube care, often with little training or ongoing testing to ensure competency over time (Page et al., 2020; Spratling, 2017; Ward et al., 2015). Parents are expected to master these demands (Knafl et al., 2013). However, few studies have examined the concepts of parental ability, competence, and readiness to manage the complex care that CMC require. Studies that have examined these concepts are largely descriptive and qualitative in nature. Nearly 32% of parents of children with congenital heart disease lack the ability and knowledge to care for their child (Ni et al., 2019).

Parents of children with single ventricle heart disease describe themselves as similar to health professionals, needing to quickly learn how to provide specific care to their child (Rempel et al., 2012). A meta-synthesis of qualitative research examining parental experiences caring for children with special health care needs describes parents as feeling overwhelmed and unprepared to care for a chronically ill child (Nygard & Clancy, 2018). Parents learn the skills necessary but with little guidance.

Few studies were identified in the literature that examine the relationship between ability (capacity) and the health outcomes of CMC or those with chronic illness. Gibson-Young et al. (2014) examined maternal behaviors in managing their children's chronic asthma. When mothers had a lower perceived ability to manage chronic asthma, the number of child hospitalizations significantly increased. This finding suggests that some health care utilization patterns and child health outcomes are related to parents' knowledge and confidence to manage the disease. In another study of variables that might influence CMC hospitalizations, Nelson et al. (2016) explores modifiable factors that influenced parents' decisions to bring their CMC in for acute care. Through interviews, these researchers found that parents seek help when they surpass a comfort threshold when caring for their CMC at home. In this study, parents expressed a desire for learning more about their CMC's condition to help them respond more effectively to concerning symptoms with the hope of preventing unnecessary acute care visits. Parents believe that having expanded access to health care providers who know their CMC will help them make decisions about when to seek acute care more appropriately. This suggests that improving family capacity by increasing parental knowledge and provider access may decrease hospitalizations and associated hospital costs.

Zhang et al. (2015) studied aspects of family management with 399 Chinese caregivers whose children have a chronic health condition. They conclude that geography, family income, and maternal education are the greatest predictors of caregiver ability. Urban location, higher income, and maternal education predict more confident management ability. It is important to highlight these findings since it is widely reported that families of CMC experience significant financial strain which may influence their ability to care for CMC. The paucity of research examining the concept of parental capacity to meet the demands necessary to manage CMC care is notable, underscoring the need to further examine these relationships and their impact on CMC health outcomes.

Knafl et al. (2013) used the Family Management Measure (FaMM) to identify patterns of family response to childhood chronic illness. Measurement scores range from those that reflect family-focused to condition-focused patterns. Most families (57%) demonstrate a family-focused or somewhat family-focused pattern that was associated with significantly better family and child functioning. The remaining 43% of families are somewhat condition-focused (35%) or condition-focused (8%) which is associated with considerable effort needed to manage their child’s chronic condition. Maternal scores especially reflected that the work required to manage the chronic condition was difficult and that mothers did not feel competent to manage it. These findings suggest that a high degree of parental workload can lead to a caregiver’s feelings of incompetence and have an impact on overall family functioning.

Several target areas have been identified as important to families and health care providers when evaluating initiatives to improve the lives of parents and their CMC

(Fayed et al., 2019). One of the target areas developed in conjunction with parents of CMC includes outcomes of "parent ability to manage their child's health and parent ability to keep up with the demand" (Fayed et al., 2019, p. 1095), supporting the idea that parental demand (workload) and parental ability (capacity) are significant concepts of interest to CMC parents, health care providers, and researchers.

Theory Derivation Using Workload and Capacity

The process of theory derivation uses and modifies existing theories to help better explain or predict a phenomenon of interest. Theory derivation is a creative and useful process when new insights about a phenomenon are needed to inspire further research or when there is a lack of available theories that include concepts of interest (Walker & Avant, 2019). Walker and Avant present theory derivation in a series of steps: 1) becoming familiar with the phenomenon of interest and proceeding with theory derivation if suitability of existing theories is lacking, 2) looking for analogies in other fields, 3) choosing a parent theory that offers a way of explaining or making predictions about the phenomenon of interest, 4) identifying the parent theory concepts to be used for clarification, and 5) using the parent theory concepts to develop a new statement or statements that provide a richer meaning for the phenomenon of interest.

Existing Frameworks Using Workload and Capacity Concepts

The philosophical underpinnings that led to an understanding of a family's response to child chronic illness initially came from literature examining normalization and dissociation in response to child polio in the 1960's (Davis, 1963). A concept analysis on normalization served as the basis for theory development on family management of a family member's chronic illness (Knafl & Deatrick, 1986). This work supported the view that family adaptation to chronic illness was poorly understood. Identifying this theoretical gap led to qualitative research by Knafl et al. (1996) that described parental responses to child chronic illness. Responses ranged from floundering to thriving styles to understand how families manage and incorporate a child's chronic illness into their lives. These researchers found that a floundering style was associated with parents feeling inadequate, burdened, and exhausted when managing child chronic illness. Additional qualitative work led to the development of the Family Management Style Framework (FMSF) for understanding how families incorporate a child's chronic illness into everyday family life (Knafl et al., 2012).

The concepts of parental workload and capacity were chosen as the basis for theory derivation because of the notable challenges described in the scientific literature that these parents experience in day-to-day CMC care management. In addition, concept analyses on children with medical complexity (Rogers et al., 2021) and on caregiver burden (Liu et al., 2020) describe CMC as complex and note that caregiver strain is multifaceted. Antecedents of caregiver burden include financial stress, extensive time necessary to provide direct care, and lack of social activities that prevent additional support that may alleviate burden. These burdens are consistent with findings in the CMC literature that point to workload and capacity as being relevant concepts for parents.

The Cumulative Complexity Model (CCM) posits that poor health outcomes can occur when illness management workload outweighs the capacity to manage it (Shippee et al., 2012). This imbalance was not identified as a concept of relevance in a search of the CMC literature or in the FMSF literature. However, the concept of an imbalance between workload and capacity has the potential to explain why CMC health outcomes remain poor. Theory derivation is useful when a set of concepts may be related to one another but lacks the structure necessary to fit the needs of a theory required to expand understanding of a patient population (Walker & Avant, 2019). Therefore, theory derivation was used to modify the FMSF. The theory derivation process enhances the dimension of management mindset in the FMSF, which includes parental effort and ability, by instead using the CCM concepts of workload and capacity. The modified framework considers how the health outcomes of CMC are affected when workload outweighs capacity.

Family Management Style Framework

The Family Management Style Framework (FMSF) is a middle-range theory for understanding how families respond to, manage, and incorporate a child's chronic illness into everyday family life (Knafl et al., 1996). According to the FMSF, patterns range from a thriving management style with a high degree of confidence and management ability, to a floundering style with difficulty in managing the illness (Knafl et al.). The FMSF is comprised of three main components within which are eight dimensions. These dimensions assist in understanding how families integrate the child's chronic condition management into their lives. One of the eight dimensions, known as "management mindset", refers to a parent's view of their ability to manage the illness, as well as whether the disease is easy or difficult to manage. The model has been used extensively in the pediatric chronic disease literature and has been applied to other family situations, from breastfeeding low birthweight infants (Krouse, 2002) to understanding family perceptions of nursing home placement of an elderly family member (Koplow et al., 2015).

Cumulative Complexity Model

Shippee et al. (2012) proposed the Cumulative Complexity Model (CCM) to explain how patient complexity is driven by an imbalance between patient workload and patient capacity to self-manage one’s chronic illness. Complexity is defined as a dynamic state using the patient’s clinical condition, which includes personal and social factors that make up a patient’s workload. An imbalance between a patient’s workload and capacity to manage that workload can lead to poor health outcomes. An increase in illness burden further diminishes capacity. Workload refers to the everyday life demands in addition to the tasks and responsibilities of having an illness or chronic condition, such as taking medications, scheduling, and attending medical appointments, undergoing laboratory and other diagnostic tests, and completing healthcare-related paperwork. Capacity refers to the patient’s ability to handle the workload, defined by a patient’s literacy, social support, socioeconomic resources, and physical and mental functioning. The model has been used to guide several adult research studies including one aimed at understanding effective interventions for reducing hospital readmissions (Leppin et al., 2014). The model has also been used as a framework for guiding a systematic review focused on multimorbid patient treatment burden (Rosbach & Andersen, 2017).

These two frameworks have strengths in their potential application to understanding the demands (workload) and ability (capacity) of parents to care for their children with medical complexity. For example, both frameworks are focused on patients with chronic illness, with the FMSF specific to the pediatric population. Moreover, both frameworks acknowledge that patient complexity is dynamic and constantly changes and evolves over time. The CCM emphasizes the functional definitions of workload and capacity as pragmatic concepts that influence health outcomes. The FMSF has added to the extensive body of knowledge regarding family management styles in a variety of populations beyond children with chronic illness.

However, there are limitations with each of the models. The CCM is adult focused and has been used sparsely in the literature. The FMSF is complex, containing three components and eight dimensions, and the concepts are not intuitively linked to their operational definitions. The concepts of parental workload and capacity are hidden in the FMSF within the dimension of management mindset, which is found in the first of three components of the framework.

While the strengths of the FMSF and CMC significantly outweigh their limitations, a revised framework was needed to define the concepts of parental workload and parental capacity and explain how these concepts influence health outcomes of CMC, particularly when workload exceeds parental capacity more clearly. The FMSF dimensions of effort and ability are similar to the constructs of workload and capacity from the CCM. However, the CCM goes further to suggest that an imbalance between workload and capacity can lead to poor health outcomes (Shippee et al., 2012) Therefore, workload and capacity from the CCM were incorporated into the FMSF, enhancing the FMSF by offering a new and insightful way of exploring the effects of parental workload and capacity on CMC health outcomes.

Integrated Theoretical Framework

The derived theoretical framework incorporates the concepts of workload and capacity from the CCM to enhance these concepts within the FMSF. This process results in a revised framework that provides richer meaning for understanding the relationship between parental workload and parental capacity for parents of CMC. In the FMSF, the concepts of effort and ability to manage CMC care are separate dimensions, with no specified relationship between them. The derived framework now illuminates how these two concepts interact with one another and influence the health outcomes of CMC, particularly when parental workload outweighs parental capacity. Moreover, this framework fills a gap in the CMC literature since most studies involving this patient population lack an overarching framework for understanding how CMC health can be improved.

New Statement Development Using Workload and Capacity

In the theoretical framework developed for use with children with medical complexity and their families, a relationship between workload and capacity exists, and is proposed to affect the health outcomes of CMC based on evidence in the literature. A relationship exists between parental workload and capacity. For example, demanding and complex home care regimens for CMC reduce parent capacity due to parental fatigue and stress. This phenomenon has been described in studies involving CMC (Caicedo, 2014; Cousino & Hazen, 2013). Knafl et al. (2013) found that mothers who regarded their child's chronic condition management as difficult also felt less competent to manage the chronic condition. A direct relationship exists between parental workload and CMC health outcomes, as evidenced by improved CMC health outcomes when parental workload is decreased through the support of a medical home or coordinated care program (Avritscher et al., 2019; Mosquera et al., 2014; Ruggiero et al., 2019). A direct relationship exists between parental workload, capacity, and health outcomes based on research involving mothers of children with chronic asthma. Children had worse asthma outcomes when their mothers had lower perceived ability and higher perceived effort to manage their child's asthma (Gibson-Young et al., 2014).

The derived framework can be used to expand researchers' and health care providers' knowledge and understanding of this growing patient population and their parents. The framework can be used to examine other factors that may affect CMC parents, including differences between mothers and fathers, as well as the workload of single CMC parents. In addition, it can be used to explore and understand how parental workload and capacity influence parental and sibling health and well-being. Furthermore, the framework can be used to investigate how contextual factors such as financial resources and social support impact parental workload and capacity.

Conclusion

Parents of children with medical complexity carry enormous burdens over the life course of their children. As the number of CMC has grown, so too has their medical complexity, compounding the demands on parents. Parental workload to care for CMC includes a remarkable amount of time providing direct care, time attending a variety of health care-related appointments, and time and attention coordinating complex aspects of CMC care. Moreover, parents experience financial burdens due to the required time away from paid work to manage all aspects of CMC care, adding further to the challenges they already experience. Randomized clinical trials of parents who receive substantial support from the use of a medical home have demonstrated improved health outcomes of CMC. These research findings suggest that parental workload and capacity are concepts that influence CMC health outcomes. In addition, CMC parents and health care providers identified these concepts as common priorities that require further study. However, knowledge gaps about the relationships of these concepts and how they impact the health of CMC remain. A derived theoretical framework using concepts that directly impact the lives of parents and families of CMC will add to researchers' and health care providers' understanding of parental workload and their capacity to meet the demands of caring for medically complex children. This is especially important since medical homes and coordinated care programs are not currently available to each and every CMC. Further study is needed to test this framework in parents of CMC with the hope of developing future interventions to attenuate parental workload and enhance parental capacity to meet the complex needs of their children.

References

Aboneh, E. A., & Chui, M. A. (2017). Care coordination, medical complexity, and unmet need for prescription medications among children with special health care needs. *Research in Social & Administrative Pharmacy, 13*(3), 524-529. https://doi.org/10.1016/j.sapharm.2016.05.043

Allshouse, C., Comeau, M., Rodgers, R., & Wells, N. (2018). Families of children with medical complexity: A view from the front lines. *Pediatrics, 141*(Suppl 3), S195-s201. https://doi.org/10.1542/peds.2017-1284D

Avritscher, E. B. C., Mosquera, R. A., Tyson, J. E., Pedroza, C., Samuels, C. L., Harris, T. S., Gomez-Rubio, A., Navarro, F. A., Moody, S. B., & Beyda, R. M. (2019). Post-trial sustainability and scalability of the benefits of a medical home for high-risk children with medical complexity. *Journal of Pediatrics, 206*, 232-239.e233. https://doi.org/10.1016/j.jpeds.2018.10.035

Berry, J. G., Agrawal, R. K., Cohen, E., & Kuo, D. Z. (2013). The landscape of medical care for children with medical complexity. *Overland Park, KS: Children's Hospital Association, 7*.

Berry, J. G., Hall, M., Neff, J., Goodman, D., Cohen, E., Agrawal, R., Kuo, D., & Feudtner, C. (2014). Children with medical complexity and medicaid: Spending and cost savings. *Health Affairs, 33*(12), 2199-2206. https://doi.org/10.1377/hlthaff.2014.0828

Cady, R., & Belew, J. (2017). Parent perspective on care coordination services for their child with medical complexity. *Children, 4*(6), 45.

Caicedo, C. (2014). Families with special needs children: Family health, functioning, and care burden. *Journal of the American Psychiatric Nurses Association, 20*(6), 398-407. https://doi.org/10.1177/1078390314561326

Cohen, E., Berry, J. G., Camacho, X., Anderson, G., Wodchis, W., & Guttmann, A. (2012). Patterns and costs of health care use of children with medical complexity. *Pediatrics, 130*(6), e1463-1470. https://doi.org/10.1542/peds.2012-0175

Cohen, E., Berry, J. G., Sanders, L., Schor, E. L., & Wise, P. H. (2018). Status complexicus? The emergence of pediatric complex care. *Pediatrics, 141*(Suppl 3), S202-s211. https://doi.org/10.1542/peds.2017-1284E

Cohen, E., Kuo, D. Z., Agrawal, R., Berry, J. G., Bhagat, S. K., Simon, T. D., & Srivastava, R. (2011). Children with medical complexity: An emerging population for clinical and research initiatives. *Pediatrics, 127*(3), 529-538. https://doi.org/10.1542/peds.2010-0910

Cousino, M. K., & Hazen, R. A. (2013). Parenting stress among caregivers of children with chronic illness: A systematic review. *Journal of Pediatric Psychology, 38*(8), 809-828. https://doi.org/10.1093/jpepsy/jst049

Davis, F. (1963). *Passage through crisis: Polio victims and their families*. Indianapolis: Bobbs-Merrill.

Fayed, N., Guttmann, A., Chiu, A., Gardecki, M., Orkin, J., Hamid, J. S., Major, N., Lim, A., & Cohen, E. (2019). Family-provider consensus outcomes for children with medical complexity. *Developmental Medicine and Child Neurology, 61*(9), 1093-1100. https://doi.org/10.1111/dmcn.14110

Feinstein, J. A., Feudtner, C., & Kempe, A. (2014). Adverse drug event-related emergency department visits associated with complex chronic conditions. *Pediatrics, 133*(6), e1575-1585. https://doi.org/10.1542/peds.2013-3060

Gibson-Young, L., Turner-Henson, A., Gerald, L. B., Vance, D. E., & Lozano, D. (2014). The relationships among family management behaviors and asthma morbidity in maternal caregivers of children with asthma. *Journal of Family Nursing, 20*(4), 442-461. https://doi.org/10.1177/1074840714552845

Hofacer, R. D., Panatopoulos, A., Vineyard, J., Tivis, R., Nguyen, E., Jingjing, N., & Lindsay, R. P. (2019). Clinical care coordination in medically complex pediatric cases: Results from the national survey of children with special health care needs. *Global Pediatric Health, 6*, 2333794x19847911. https://doi.org/10.1177/2333794x19847911

Kaushal, R., Goldmann, D. A., Keohane, C. A., Christino, M., Honour, M., Hale, A. S., Zigmont, K., Lehmann, L. S., Perrin, J., & Bates, D. W. (2007). Adverse drug events in pediatric outpatients. *Ambulatory Pediatrics, 7*(5), 383-389.

Keim-Malpass, J., Letzkus, L. C., & Kennedy, C. (2015). Parent/caregiver health literacy among children with special health care needs: A systematic review of the literature. *BMC Pediatrics, 15*, 92. https://doi.org/10.1186/s12887-015-0412-x

Knafl, K., Breitmayer, B., Gallo, A., & Zoeller, L. (1996). Family response to childhood chronic illness: Description of management styles. *Journal of Pediatric Nursing, 11*(5), 315-326.

Knafl, K., & Deatrick, J. A. (1986). How families manage chronic conditions: An analysis of the concept of normalization. *Research in Nursing & Health, 9*(3), 215-222.

Knafl, K. A., Deatrick, J. A., & Havill, N. L. (2012). Continued development of the family management style framework. *Journal of Family Nursing, 18*(1), 11-34. https://doi.org/10.1177/1074840711427294

Knafl, K. A., Deatrick, J. A., Knafl, G. J., Gallo, A. M., Grey, M., & Dixon, J. (2013). Patterns of family management of childhood chronic conditions and their relationship to child and family functioning. *Journal of Pediatric Nursing, 28*(6), 523-535. https://doi.org/10.1016/j.pedn.2013.03.006

Koplow, S. M., Gallo, A. M., Knafl, K. A., Vincent, C., Paun, O., & Gruss, V. (2015). Family caregivers define and manage the nursing home placement process. *Journal of Family Nursing, 21*(3), 469-493.

Krouse, A. M. (2002). The family management of breastfeeding low birth weight infants. *Journal of Human Lactation, 18*(2), 155-165.

Kuo, D. Z., Cohen, E., Agrawal, R., Berry, J. G., & Casey, P. H. (2011). A national profile of caregiver challenges among more medically complex children with special health care needs. *Archives of Pediatrics and Adolescent Medicine, 165*(11), 1020-1026. https://doi.org/10.1001/archpediatrics.2011.172

Lawoko, S., & Soares, J. J. (2003). Quality of life among parents of children with congential heart disese, parents of children with other diseases, and parents of healthy children. *Quality of Life Research, 12*(6), 655-666.

Leppin, A. L., Gionfriddo, M. R., Kessler, M., Brito, J. P., Mair, F. S., Gallacher, K., Wang, Z., Erwin, P. J., Sylvester, T., & Boehmer, K. (2014). Preventing 30-day hospital readmissions: A systematic review and meta-analysis of randomized trials. *JAMA Internal Medicine, 174*(7), 1095-1107.

Liu, Z., Heffernan, C., & Tan, J. (2020). Caregiver burden: A concept analysis. *International Journal of Nursing Sciences, 7*, 438-445. https://doi.org/10.1016/j.ijnss.2020.07.012

Mooney-Doyle, K., & Lindley, L. C. (2019). The association between poverty and family financial challenges of caring for medically complex children. *Nursing Economics, 37*(4), 198-208.

Mooney-Doyle, K., & Lindley, L. C. (2020). Family and child characteristics associated with caregiver challenges for medically complex children. *Family and Community Health, 43*(1), 74-81.

Mosquera, R. A., Avritscher, E. B., Samuels, C. L., Harris, T. S., Pedroza, C., Evans, P., Navarro, F., Wootton, S. H., Pacheco, S., Clifton, G., Moody, S., Franzini, L., Zupancic, J., & Tyson, J. E. (2014). Effect of an enhanced medical home on serious illness and cost of care among high-risk children with chronic illness: A randomized clinical trial. *JAMA, 312*(24), 2640-2648. https://doi.org/10.1001/jama.2014.16419

Ni, Z. H., Lv, H. T., Ding, S., & Yao, W. Y. (2019). Home care experience and nursing needs of caregivers of children undergoing congenital heart disease operations: A qualitative descriptive study. *PloS One, 14*(3), e021315 https://doi.org/10.1371/journal.pone.0213154

Nygard, C., & Clancy, A. (2018). Unsung heros, flying blind – A metasynthesis of parents' experiences of caring for chldren with special health care needs at home. *Journal of Clinical Nursing, 27*, 3179-3196. https://doi.org/10.1111/jocn.14512

Page, B. F., Hinton, L., Harrop, E., & Vincent, C. (2020). The challenges of caring for children who require complex medical care at home: 'The go between for everyone is the parent and as the parent that's an awful lot of responsibility'. *Health Expectations, 1-11*. https://doi:10.1111/hex.13092

Pierce, M., Hope, H. F., Kolade, A., Gellatly, J., Osam, C. S., Perchard, R., Kosidou, K., Dalman, C., Morgan, V., Di Prinzio, P., & Abel, K. M. (2019). Effects of parental mental illness on children's physical health: Systematic review and meta-analysis. *British Journal of Psychiatry*, 1-10. https://doi.org/10.1192/bjp.2019.216

Rempel, G. R., Blythe, C., Rogers, L. G., & Ravindran, V. (2012). The process of family management when a baby is diagnosed with a lethal congenital condition. *Journal of Family Nursing, 18*(1), 35-64.

Rogers, J., Reed, M.P., Blaine, K., & Manning, H. (2021). Children with medical complexity: A concept analysis. *Nursing Forum*, 1-8. https://doi:10.1111/nuf.12559

Romley, J. A., Shah, A. K., Chung, P. J., Elliott, M. N., Vestal, K. D., & Schuster, M. A. (2017). Family-provided health care for children with special health care needs. *Pediatrics, 139*(1). https://doi.org/10.1542/peds.2016-1287

Rosbach, M., & Andersen, J. S. (2017). Patient-experienced burden of treatment in patients with multimorbidity–A systematic review of qualitative data. *PloS One, 12*(6), e0179916.

Ruggiero, K., Pratt, P., & Antonelli, R. (2019). Improving outcomes through care coordination: Measuring care coordination of nurse practitioners. *Journal of the American Association of Nurse Practitioners, 31*(8), 476-481. https://doi.org/10.1097/jxx.0000000000000276

Shippee, N. D., Shah, N. D., May, C. R., Mair, F. S., & Montori, V. M. (2012). Cumulative complexity: A functional, patient-centered model of patient complexity can improve research and practice. *Journal of Clinical Epidemiology, 65*(10), 1041-1051.

Shumskiy, I., Richardson, T., Brar, S., Hall, M., Cox, J., Crofton, C., Peltz, A., Samuels-Kalow, M., Alpern, E. R., Neuman, M. I., & Berry, J. G. (2018). Well-child visits of Medicaid-insured children with medical complexity. *Journal of Pediatrics, 199*, 223-230.e222. https://doi.org/10.1016/j.jpeds.2018.04.003

Spratling, R. (2017). Understanding the health care utilization of children who require medical technology: A descriptive study of children who require tracheostomies. *Applied Nursing Research, 34*, 62-65.

Walker, L. O., & Avant, K. C. (2019). *Strategies for theory construction in nursing*. (6th ed.). Pearson.

Walsh, K. E., Mazor, K. M., Stille, C. J., Torres, I., Wagner, J. L., Moretti, J., Chysna, K., Stine, C. D., Usmani, G. N., & Gurwitz, J. H. (2011). Medication errors in the homes of children with chronic conditions. *Archives of Disease in Childhood, 96*(6), 581-586.

Ward, C., Glass, N., & Ford, R. (2015). Care in the home for seriously ill children with complex needs: A narrative literature review. *Journal of Child Health Care, 19*(4), 524-531. https://doi.org/10.1177/1367493514538327

Zhang, Y., Wei, M., Shen, N., & Zhang, Y. (2015). Identifying factors related to family management during the coping process of families with childhood chronic conditions: A multi-site study. *Journal of Pediatric Nursing, 30*(1), 160-173. https://doi.org/10.1016/j.pedn.2014.10.

APPENDIX B

Parental Workload in Caring for Children with Medical Complexity: A Review of Socioeconomic Status Impact on Parental Depressive Symptoms

Manuscript submitted to *Journal of Pediatric Nursing* on April 9, 2022

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Abstract

Theoretical Principles: Children with medical complexity (CMC) experience poor health outcomes despite their high care costs. Their parents face a tremendous workload and extraordinary challenges, including economic hardship, to provide care to their children. Guided by a theoretical framework, this review discusses what is known about this workload and how it impacts the mental health of these parents over time.

Phenomena Addressed: The workload required by parents of CMC is presented, including workload related to direct care, care coordination, and health care access. Parents of CMC also experience financial strain that is directly related to the care their medically complex children require at home. The relationships between workload, socioeconomic status, and parental mental health are explored.

Research Linkages: Research has demonstrated that as socioeconomic status worsens, depression increases. Due to the financial strains that families of CMC experience, the impact of parental depression may add further to the demands that parents endure and to the health disparities that CMC already experience. Medical homes, safe work hour standards, respite care, and routine screening for parental stress, depressive symptoms, and family resource needs are critical to supporting parents of CMC to alleviate their workload and improve their mental health.

Keywords: children with medical complexity, depression, parent, socioeconomic status, stressors

Parental Workload in Caring for Children with Medical Complexity: A Review of Socioeconomic Status Impact on Parental Depressive Symptoms

Introduction

Children with medical complexity (CMC) represent patients with high health care utilization due to their chronic, sometimes life-limiting health conditions. Their medical diagnoses vary in number and type, and they require a host of health care services (Cohen et al., 2011; Kuo et al., 2011). Considerable research has focused on their disproportionately high healthcare costs (Simon et al., 2010) and poor health outcomes (Cohen et al., 2012; Kuo et al., 2018; Shumskiy et al., 2018) despite the intensive care they receive. Moreover, a large proportion of families with CMC report financial hardship (Mooney-Doyle & Lindley, 2019). While studies examining CMC outcomes are increasing, research examining the broader impact on parents has been limited, particularly the impact on parental mental health. Understanding factors associated with parental depressive symptoms is important given evidence for worse health outcomes in children whose parents have poor mental health (Pierce et al., 2019), and evidence for higher pediatric health care use in depressed mothers (Brooks et al., 2015). Poor mental health may limit parents' ability to care for children, particularly for those children who are more medically complex and who require complex care at home such as CMC. However, these relationships have not been examined in this patient population.

It has been well established that as socioeconomic status (SES) decreases, depression increases (Lorant et al., 2007). Due to the financial strains that the families of CMC are known to sustain, the impact of parental depression may add further to the disparities that CMC already experience, and therefore requires further exploration. To understand how parental mental health is impacted, this review outlines the workload associated with raising children with medical complexity including their associated health care service needs and costs. Given what is known about the relationship between SES and depression (Freeman et al., 2016; Lorant et al., 2007) and between SES and social stress (Szanton et al., 2005), this review supports the idea that depression may be more marked in parents of CMC, underscoring the need to further explore and understand the relationships between workload, SES, and depression in this population. This paper concludes with several recommendations to reduce the workload experienced by parents of CMC.

Theoretical Framework

This review is guided by a framework that integrates components of two theories to provide a deeper understanding of the challenges that parents of CMC experience (Lawrence & Spratling, 2022). This integrated framework was derived from the Family Management Style Framework (Knafl et al., 2012) and the Cumulative Complexity Model (Shippee et al., 2012) to examine relationships between relevant concepts identified by parents of CMC as important in their lives (Fayed et al., 2019). Specifically, the framework explores the relationship between the workload that parents of CMC encounter and parents' ability to do the work necessary to care for their CMC (Lawrence & Spratling). The framework posits that poor CMC and family outcomes occur when workload exceeds the capacity to care for the complex medical needs of these children. Family resources, which includes socioeconomic status and specifically financial resources, may moderate (buffer) the relationship between the workload that parents of

CMC experience and parental depressive symptoms (see Figure 1). However, these relationships have not been explored in this patient population.

Review

CMC Definition

Children with medical complexity (CMC) are referred to as “the sickest of the sick” yet are often confused with other terms such as children with special health care needs (CSHCN) (Cohen et al., 2011). While CSHCN do indeed have special health care-related needs due to a chronic illness, there is considerable difference between CSHCN and those who are more medically complex. Specifically, CMC are defined by four main features. They have substantial health service needs, have one or more chronic and typically lifelong medical conditions such as complex congenital heart disease (CCHD), have functional limitations, and have high health care utilization (Cohen et al., 2011; Kuo et al., 2011). Clarifying the definition of CMC is essential because several studies described in this review include pediatric chronic diseases such as asthma and epilepsy that are considered serious but don’t always fit the definition for medical complexity. Rather, those are considered conditions under the umbrella of CSHCN, especially when they are categorical diagnoses in isolation and not in combination with other medical problems (Berry et al., 2015).

Workload Experienced by Parents of CMC

Workload is defined as the day-to-day demands and care activities that parents must provide for their CMC and is a main component of the derived theory used to understand how workload impacts parent and family outcomes (Lawrence & Spratling, 2021). These demands occur on a routine basis and have a significant impact on families, since CMC receive their majority of care at home (Javalkar et al., 2017; Page et al., 2020). Care activities might include giving enteral tube feedings, performing tracheostomy care, and administering complex medication regimens. This care can be intensive, requiring physical labor and high levels of attention to monitor for and prevent clinical decompensation (Schall et al., 2020). Care coordination programs, often referred to as “medical homes”, decrease parental stressors by reducing serious CMC illness, reducing hospital costs and hospital lengths of stay (Avritscher et al., 2019; Mosquera et al., 2014; Ruggiero et al., 2019a). Medical homes also improve care access and parent satisfaction by seamlessly communicating and caring holistically for CMC. However, these programs are uncommon and not a widely available resource to parents (Allshouse et al., 2018; Hofacer et al., 2019). Therefore, most parents are left to navigate a complex health care system on their own without much guidance, resulting in shortcomings that “hit the CMC population first and hardest” (Allshouse et al., 2018, p. S196).

Parents of CMC are at increased risk for experiencing stress and depressive symptoms directly associated with the complex care that CMC require, in addition to the social and economic stressors that these parents are already known to experience (Pinquart, 2018; Ratliffe et al., 2002; Rehm, 2013). Also referred to as “challenges, demands or burdens” by parents (Javalkar et al., 2017; Nygard & Clancy, 2018; Page et al., 2020), these stressors are directly related to the care that CMC require at home (Javalkar et al., 2017; Page et al., 2020). The workload and demands placed on parents to manage the daily home care of CMC are enormous. Parents are sometimes sent home with insufficient resources to manage the care that health care providers expect (Schall et al., 2020). Moreover, parents also feel unsupported when navigating what they describe as a complex health care system that is fragmented, siloed, and confusing (Allshouse et al., 2018; R. Cady & J. Belew, 2017), amplifying the workload they experience.

Workload Related to Direct Care

Parents provide most of the direct home care for their child. Often, this care requires complex skills (Mooney-Doyle & Lindley, 2020) for which parents receive little training (Barnert et al., 2019; Spratling, 2017). Parents are responsible for specialized skills at home including managing complex medication schedules and life-sustaining medical technology such as feeding tubes, tracheostomies and ventilators (Caicedo, 2014; Mooney-Doyle & Lindley, 2020) Nearly half of all children with complex medical needs only receive unpaid care at home by one or more family members, and nearly 12% of these children receive 21 or more hours of direct, unpaid care each week at home by a family member (Romley et al., 2017). Experts now emphasize that these unpaid parent work hours *exceed* what is considered safe in other industries such as nursing, medicine, and the aviation industry (Schall et al., 2020).

Workload Related to Care Coordination

In addition to providing direct care, the time parents spend coordinating care is considerable, with some parents describing it as a full-time job (R. G. Cady & J. L. Belew, 2017). Parents spend a median of between 2 hours per week (Kuo et al., 2011) and 6.6 hours each week to coordinate CMC care (Caicedo, 2014; Mooney-Doyle & Lindley, 2020; Romley et al., 2017). Effective care coordination requires time and skills that are usually provided by medical professionals in a program-based medical home (Antonelli et al., 2008; Ruggiero et al., 2019b). However, more than 65% of CMC parents receive no care coordination support, magnifying the workload required by parents to manage the complexities involved in the care of their CMC when professional care coordination support is not available (Hofacer et al., 2019). Many parents need to quickly adopt these skills as best they can and without much guidance (R. G. Cady & J. L. Belew, 2017; Mooney-Doyle & Lindley, 2020). Without resources, adequate training and preparedness, poorer CMC health outcomes can occur (Boroughs & Dougherty, 2016).

Workload Related to Health Care Access

Parents spend a considerable amount of time taking their CMC to health care appointments. It has been reported that parents and their CMC attend a median of between 11 and 15 medical appointments each year (Kuo et al., 2011). Others report a median of 13 different outpatient physicians providing necessary care from 6 distinct medical specialties to manage the medical needs of CMC (Cohen et al., 2012). Parents describe traveling frequently to their CMC's health care specialists, sometimes a significant distance from home (Kuo et al., 2016) . This is largely due to many specialists being part of urban, tertiary-care centers (R. Cady & J. Belew, 2017). As one can imagine, parents must often take time away from paid work to attend these appointments. The more medically complex the child, the more specialty providers that are required to manage the myriad of conditions, requiring even more appointments (Kuo et al., 2015).

Despite the degree to which parents and their CMC interact with the health care system, CMC are twice as likely to have one or more unmet need when compared to children without a complex medical condition despite family income, primary language, or having Medicaid (Kuo et al., 2014). This finding suggests that medical complexity is a primary determinant of the health inequities seen among CMC, rather than other typical social determinants of health. Unmet needs include prescriptions and preventive medical services such as dental and vision care. More than 5% of more medically complex children have 5 or more unmet medical service needs (Kuo et al., 2011), which is in keeping with findings from later studies (Kuo et al., 2014). Families also have difficulty accessing non-medical services such as early intervention and childcare when compared to parents of children who are less medically complex, which again points to complexity as being a dominant reason for health inequities experienced by CMC (Kuo et al.). The frequency of health care visits does not necessarily ensure that all needs are met for this fragile patient population.

Workload Related to Financial Strain

In addition to the workload associated with providing day to day care for CMC, many parents face economic stressors that compound the workload they experience. High CMC health care costs are due in large part to their complexity of health conditions. These complex conditions require unique inpatient and outpatient medical and nursing management from one or more health care specialists, in addition to expensive medications, therapies, procedures and life-sustaining technologies (Allshouse et al., 2018; Berry et al., 2013). Approximately 5.6 million special health care needs children receive an annual 1.5 billion hours of unpaid, family-provided health care, underscoring the substantial economic burden these families carry (Romley et al., 2017). The need for extensive traveling for specialty care also directly impacts the financial health of many families. This is due primarily to lost wages from parents' interrupted work hours to attend CMC appointments and to be present for extended hospital stays (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019, 2020). Moreover, CMC condition complexity appears to be increasing over time (Burns et al., 2010). With increasing medical complexity and the expenditures associated with it, these out-of-pocket costs are passed on to parents.

Many parents experience personal financial insecurity and economic hardship due to out-of-pocket, non-reimbursed expenses and lost wages from an inability to work to care for their medically complex children (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019, 2020; Vessey et al., 2017). Kuo et al. (2011) reported 46.3% of parents paid more than $1,000 in the prior year toward out-of-pocket medical expenses, and 48.7% of parents reported a lack of income to cover the medical expenses of their CMC. This same study also found that 56.8% of parents reported financial difficulties directly related to the costs of unreimbursed CMC care and 54.1% of families required that one family member stop working to provide direct care at home for their CMC, consistent with later studies (Thomson et al., 2016).

Families of children with severe cerebral palsy spent between $193 and $7,192.71 *per hospitalization* for hip or spine surgery when transportation, lodging, food, dependent care, housekeeping, lost work hours and incidental expenses as non-medical out-of-pocket expenses were included in the analysis (Vessey et al., 2017). In addition to reporting financial strain, more than 75% of families in this study needed additional funds to cover their child's expenses and the costs associated with missed work hours. While health insurance covers most hospital-related medical costs, it is well known that it doesn't cover all expenses that are associated with travel requirements to more distant health care centers for expert, regional care.

Another indirect cause of financial hardship that affects families of CMC are negative career impacts such as the loss of a promotion or the need to take a leave of absence to care for their CMC (Mandic et al., 2017). Although rarely reported in the literature, one study found that 46% of primary caregivers of CMC took a leave of absence, compared to 31% of spouses (Mandic et al.). This same study notes that 16% of primary caregivers and 15% of their spouses left jobs involuntarily to care for their CMC, and 9% of primary caregivers and 12% of their spouses turned down job promotions to provide direct home care to their CMC. These employment changes and losses have a significant impact on the financial well-being of these families.

Not surprisingly, parents report a reduced quality of life partially due to the financial difficulties associated with caring for their chronically ill child (Lawoko & Soares, 2003). Ultimately, the financial costs to CMC parents are inextricably linked to the unavoidable time and resources required to independently care for their medically complex children at home. It is clear from a number of studies, some of which use large, nationally representative samples, that the workload of parents of CMC is substantial, affecting families' time, financial status, and CMC health and well-being (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019, 2020).

Relationship between Workload and Parental Depression

According to the theoretical framework used to guide this review (Lawrence & Spratling, 2021) CMC and parent health outcomes can be negatively impacted when workload outweighs capacity. While the challenges and demands that parents face in providing direct care for their CMC have been well described, less is known about the mental health consequences that this level of care at home has on parents of CMC. Different aspects of health in parents of CMC are beginning to emerge in the literature, yet less is known about the mental health, particularly depression, of CMC parents (Barnert et al., 2019).

Several studies have examined depression among parents whose children have categorical chronic conditions such as congenital heart disease (Woolf-King et al., 2017), Type 1 diabetes, renal disease, or epilepsy (Khanna et al., 2015) and intellectual and developmental disabilities (Scherer et al., 2019). A systematic review and meta-analysis assessing health outcomes of parents caring for children with chronic illnesses included 23 studies on depression in their analysis, 11 of which used the Center for Epidemiological Studies-Depression (CES-D), which measures for depressive symptoms (Cohn et al., 2020). Findings from these studies are similar, with depressive symptoms for parents of children with one or more chronic diseases ranging between 31% (Scherer et al.) and 38% (Khanna et al.) and as high as 50% (Woolf-King et al.), compared to just 19% in parents of healthy children (Cohn et al.).

Although studies have examined depressive symptoms in parents whose children have a variety of categorical chronic health problems, less attention has been paid to exploring parental depressive symptoms in parents of CMC, highlighting an important knowledge gap for this growing, more medically complex patient population. Given the well-established link between stress and depression (Hammen, 2004), it is reasonable to anticipate that the stress associated with the workload required to care for CMC places their parents at increased risk for depression.

Relationship between Socioeconomic Status and Depression

Socioeconomic status (SES) includes economic and educational resources that are important to caring for CMC. SES has been linked to both stress exposure (Szanton et al.,

2005) and depression in the general population (Freeman et al., 2016; Lorant et al., 2007; Missinne & Bracke, 2012). SES may serve as a moderating factor between the workload and stressors that parents experience in caring for their CMC and parental mental health. For example, depressive symptoms may be lower for parents with higher SES who have more favorable economic and educational means to better cope with the stressors associated with caring for CMC. Woolf-King et al. (2017) emphasized the need to further study the moderating factors that most influence the mental health of parents with children with critical congenital heart disease (CCHD) based on a lack of consistent findings across 30 studies used to understand the impact CCHD has on parental mental health.

Large studies in the general adult population have examined the relationship between socioeconomic status (SES) and depression, finding more depression associated with lower SES, both cross-sectionally (Freeman et al., 2016) and over time (Lorant et al., 2007). Studies have looked at the relationship between SES and parental depression among different populations of children with a variety of chronic health conditions such as asthma (Zhou et al., 2014) and intellectual and developmental disabilities (Scherer et al., 2019), and demonstrated similar findings, although none of these studies specifically included the CMC population. Driscoll et al. (2010) found comparable levels of caregiver depressive symptoms in parents of both type 1 diabetes (T1D) and cystic fibrosis (CF) children, with 33.3% of T1D parents and 32.2% of CF parents reporting clinically relevant depressive symptoms. Regression analyses demonstrated lower caregiver education as the best predictor for caregiver depressive symptoms in the T1D group, whereas lack of employment was the best predictor for caregiver depressive symptoms in the CF group, suggesting that depression in parents of children with different chronic diseases are impacted by different SES variables in a variety of ways.

From these studies that examine the relationship between different SES variables and depression in parents of children with a variety of chronic conditions, research has established that more depressive symptoms are correlated with lower household income, lower caregiver education, or lack of employment. However, research examining these relationships in parents of the CMC population is lacking.

Discussion

Evidence from this review highlights parents of CMC experience a number of stressors as a direct result of the workload in providing day to day care to their children. This workload includes the time required to provide direct care for their CMC, time required to coordinate complex care, and time away from work and other daily responsibilities to attend a variety of health care appointments or to be present for lengthy hospitalizations. The financial strains that parents of CMC experience from lost wages and out-of-pocket expenses are enormous and directly impact the SES of these families. Expenses such as health care visit and medication copays, non-reimbursed equipment costs, and travel expenses to health care specialists add further to the financial stressors that these parents already experience.

The links between stress and depression (Hammen, 2004) and between SES and depression (Freeman et al., 2016; Lorant et al., 2007) have been well established. Researchers must focus their efforts on the mental health of CMC parents. This is important, based on what is known about depression in parents of children with categorical chronic health conditions, in addition to what is known about the financial burdens parents of CMC experience. However, there is a gap in our understanding of the degree to which these parents experience depression. Moreover, a knowledge gap exists in the moderating effects that SES may have on the relationship between workload and depression for parents of CMC who already experience significant financial strain. Finally, research is needed to examine other variables that may ease the workload of these parents and potentially minimize depression, but studies looking first at the prevalence of depression is a reasonable starting point.

Implications for Practice, Nursing Research, Policy, and Advocacy

Routine screening and assessment of family financial health and parental mental well-being is critical, particularly since research demonstrates that as SES worsens, depression increases. Such screening is imperative to assist families in obtaining available mental health support and financial resources when they are most needed.

Research examining the relationship between social support and the workload that parents of CMC experience is also important, since parents are often isolated and have few social supports (Thomson et al., 2016). In addition, studies that explore resilience and its protective role in mitigating depression are needed, since resilience influences parents' stress response as well as the health of parents and their children (Lisanti, 2018).

Importantly, respite care and work-hour safety standards for parents of CMC must be developed with proper advocacy and policy support. This is important based on what is known about the unpaid, direct care hours that parents provide to care for these medically fragile children. CMC experts suggest that parents of CMC should not work to care for their children more than 9 consecutive hours based on what is known about the association of adverse events and performance-impairing fatigue in other safety-sensitive occupations such as nursing, transportation, and emergency services (Schall et al., 2020; Williamson et al., 2011). These experts argue that the work provided by parents to care for their CMC should be held to the same safety standards used in other high-risk fields to prevent exhaustion and burn out.

Conclusion

Parents of children with medical complexity carry enormous burdens over the course of their children's' lives. Time-intensive responsibilities that require a high level of attention include providing direct care and coordination for their CMC and attending a variety of health care-related appointments. These responsibilities often require parents to cut down on, stop paid work, or forgo work promotions. These circumstances result in financial strain for many families, and poverty for some. The financial impact experienced by parents is compounded by non-reimbursed, out of pocket expenses that can quickly accumulate with medical complexity. The more medically complex these children are, the more time constraints parents experience, thus creating a viscous cycle that leads to further financial stress. Experts suspect that parental depression is another burden that parents of CMC experience. However, further research is needed to examine its prevalence, and to see if SES attenuates parental depression in a growing patient population who is disproportionately impacted by financial hardship.

References

Allshouse, C., Comeau, M., Rodgers, R., & Wells, N. (2018). Families of children with medical complexity: A view from the front lines. *Pediatrics, 141*(Suppl 3), S195-s201. https://doi.org/10.1542/peds.2017-1284D

Antonelli, R. C., Stille, C. J., & Antonelli, D. M. (2008). Care coordination for children and youth with special health care needs: A descriptive, multisite study of activities, personnel costs, and outcomes. *Pediatrics, 122*(1), e209-216. https://doi.org/10.1542/peds.2007-2254

Avritscher, E. B. C., Mosquera, R. A., Tyson, J. E., Pedroza, C., Samuels, C. L., Harris, T. S., Gomez-Rubio, A., Navarro, F. A., Moody, S. B., & Beyda, R. M. (2019). Post-trial sustainability and scalability of the benefits of a medical home for high-risk children with medical complexity. *Journal of Pediatrics, 206*, 232-239.e233. https://doi.org/10.1016/j.jpeds.2018.10.035

Barnert, E. S., Coller, R. J., Nelson, B. B., Thompson, L. R., Tran, J., Chan, V., Padilla, C., Klitzner, T. S., Szilagyi, M., & Chung, P. J. (2019). Key population health outcomes for children with medical complexity: A systematic review. *Maternal and Child Health Journal, 23*(9), 1167-1176. https://doi.org/10.1007/s10995-019-02752-1

Berry, J. G., Agrawal, R. K., Cohen, E., & Kuo, D. Z. (2013). The landscape of medical care for children with medical complexity. *Overland Park, KS: Children's Hospital Association, 7*.

Berry, J. G., Hall, M., Cohen, E., O'Neill, M., & Feudtner, C. (2015). Ways to identify children with medical complexity and the importance of why. *Journal of Pediatrics, 167*(2), 229-237. https://doi.org/10.1016/j.jpeds.2015.04.068

Boroughs, D. S., & Dougherty, J. (2016). In-home care of the child on chronic mechanical ventilation. In *Caring for the Ventilator Dependent Child* (pp. 165-183). Springer.

Brooks, J. L., Beil, H., & Beeber, L. S. (2015). Maternal depressive symptoms and healthcare expenditures for publicly insured children with chronic health conditions. *Maternal and Child Health Journal, 19*(4), 790-797.

Burns, K. H., Casey, P. H., Lyle, R. E., Bird, T. M., Fussell, J. J., & Robbins, J. M. (2010). Increasing prevalence of medically complex children in us hospitals. *Pediatrics, 126*(4), 638-646. https://doi.org/10.1542/peds.2009-1658

Cady, R., & Belew, J. (2017). Parent perspective on care coordination services for their child with medical complexity. *Children, 4*(6), 45.

Cady, R. G., & Belew, J. L. (2017). Parent perspective on care coordination services for their child with medical complexity. *Children (Basel), 4*(6). https://doi.org/10.3390/children4060045

Caicedo, C. (2014). Families with special needs children: Family health, functioning, and care burden. *Journal of the American Psychiatric Nurses Association, 20*(6), 398-407. https://doi.org/10.1177/1078390314561326

Cohen, E., Berry, J. G., Camacho, X., Anderson, G., Wodchis, W., & Guttmann, A. (2012). Patterns and costs of health care use of children with medical complexity. *Pediatrics, 130*(6), e1463-1470. https://doi.org/10.1542/peds.2012-0175

Cohen, E., Kuo, D. Z., Agrawal, R., Berry, J. G., Bhagat, S. K., Simon, T. D., & Srivastava, R. (2011). Children with medical complexity: An emerging population for clinical and research initiatives. *Pediatrics, 127*(3), 529-538. https://doi.org/10.1542/peds.2010-0910

Cohn, L. N., Pechlivanoglou, P., Lee, Y., Mahant, S., Orkin, J., Marson, A., & Cohen, E. (2020). Health outcomes of parents of children with chronic illness: A systematic review and meta-analysis. *The Journal of Pediatrics*, 218, 166-177.

Driscoll, K. A., Johnson, S. B., Barker, D., Quittner, A. L., Deeb, L. C., Geller, D. E., Gondor, M., & Silverstein, J. H. (2010). Risk factors associated with depressive symptoms in caregivers of children with type 1 diabetes or cystic fibrosis. *Journal of Pediatric Psychology, 35*(8), 814-822.

Fayed, N., Guttmann, A., Chiu, A., Gardecki, M., Orkin, J., Hamid, J. S., Major, N., Lim, A., & Cohen, E. (2019). Family-provider consensus outcomes for children with medical complexity. *Developmental Medicine and Child Neurology, 61*(9), 1093-1100. https://doi.org/10.1111/dmcn.14110

Freeman, A., Tyrovolas, S., Koyanagi, A., Chatterji, S., Leonardi, M., Ayuso-Mateos, J. L., Tobiasz-Adamczyk, B., Koskinen, S., Rummel-Kluge, C., & Haro, J. M. (2016). The role of socio-economic status in depression: Results from the courage (aging survey in europe). *BMC Public Health, 16*(1), 1098.

Hammen, C. (2004). Stress and depression. *Annual Review of Clinical Psychology, 1*(1), 293-319. https://doi.org/10.1146/annurev.clinpsy.1.102803.143938

Hofacer, R. D., Panatopoulos, A., Vineyard, J., Tivis, R., Nguyen, E., Jingjing, N., & Lindsay, R. P. (2019). Clinical care coordination in medically complex pediatric cases: Results from the national survey of children with special health care needs. *Global Pediatric Health, 6*, 2333794x19847911. https://doi.org/10.1177/2333794x19847911

Javalkar, K., Rak, E., Phillips, A., Haberman, C., Ferris, M., & Van Tilburg, M. (2017). Predictors of caregiver burden among mothers of children with chronic conditions. *Children, 4*(5), 39.

Khanna, A. K., Prabhakaran, A., Patel, P., Ganjiwale, J. D., & Nimbalkar, S. M. (2015). Social, psychological and financial burden on caregivers of children with chronic illness: A cross-sectional study. *The Indian Journal of Pediatrics, 82*(11), 1006-1011.

Knafl, K. A., Deatrick, J. A., & Havill, N. L. (2012, Feb). Continued development of the family management style framework. *Journal of Family Nursing, 18*(1), 11-34. https://doi.org/10.1177/1074840711427294

Kuo, D. Z., Berry, J. G., Glader, L., Morin, M. J., Johaningsmeir, S., & Gordon, J. (2016). Health services and health care needs fulfilled by structured clinical programs for children with medical complexity. *Journal of Pediatrics, 169*, 291-296.e291. https://doi.org/10.1016/j.jpeds.2015.10.010

Kuo, D. Z., Cohen, E., Agrawal, R., Berry, J. G., & Casey, P. H. (2011). A national profile of caregiver challenges among more medically complex children with special health care needs. *Archives of Pediatrics and Adolescent Medicine, 165*(11), 1020-1026. https://doi.org/10.1001/archpediatrics.2011.172

Kuo, D. Z., Goudie, A., Cohen, E., Houtrow, A., Agrawal, R., Carle, A. C., & Wells, N. (2014). Inequities in health care needs for children with medical complexity. *Health Affairs, 33*(12), 2190-2198.

Kuo, D. Z., McAllister, J. W., Rossignol, L., Turchi, R. M., & Stille, C. J. (2018). Care coordination for children with medical complexity: Whose care is it, anyway? *Pediatrics, 141*(Suppl 3), S224-s232. https://doi.org/10.1542/peds.2017-1284G

Kuo, D. Z., Melguizo-Castro, M., Goudie, A., Nick, T. G., Robbins, J. M., & Casey, P. H. (2015). Variation in child health care utilization by medical complexity. *Maternal and Child Health Journal, 19*(1), 40-48.

Lawoko, S., & Soares, J. J. (2003). Quality of life among parents of children with congenital heart disease, parents of children with other diseases and parents of healthy children. *Quality of Life Research, 12*(6), 655-666.

Lawrence, P. R., & Spratling, R. (2022). A theory for understanding parental workload and capacity to care for children with medical complexity. *Research and Theory for Nursing Practice, 36*(1), 34-46. https://doi.org/10.1891/rtnp-2022-0026

Lisanti, A. J. (2018). Parental stress and resilience in chd: A new frontier for health disparities research. *Cardiology in the Young, 28*(9), 1142-1150.

Lorant, V., Croux, C., Weich, S., Deliège, D., Mackenbach, J., & Ansseau, M. (2007). Depression and socio-economic risk factors: 7-year longitudinal population study. *The British jJournal of Psychiatry, 190*(4), 293-298.

Mandic, C. G., Johaningsmeir, S., Corden, T. E., Earle, A., Acevedo-Garcia, D., & Gordon, J. B. (2017). Impact of caring for children with medical complexity on parents’ employment and time. *Community, Work & Family, 20*(4), 444-458. https://doi.org/10.1080/13668803.2016.1202195

Missinne, S., & Bracke, P. (2012). Depressive symptoms among immigrants and ethnic minorities: A population based study in 23 european countries. *Social Psychiatry and Psychiatric Epidemiology, 47*(1), 97-109.

Mooney-Doyle, K., & Lindley, L. C. (2019). The association between poverty and family financial challenges of caring for medically complex children. *Nursing Economics, 37*(4), 198-208.

Mooney-Doyle, K., & Lindley, L. C. (2020). Family and child characteristics associated with caregiver challenges for medically complex children. *Family and Community Health, 43*(1), 74-81.

Mosquera, R. A., Avritscher, E. B., Samuels, C. L., Harris, T. S., Pedroza, C., Evans, P., Navarro, F., Wootton, S. H., Pacheco, S., Clifton, G., Moody, S., Franzini, L., Zupancic, J., & Tyson, J. E. (2014). Effect of an enhanced medical home on serious illness and cost of care among high-risk children with chronic illness: A randomized clinical trial. *JAMA, 312*(24), 2640-2648. https://doi.org/10.1001/jama.2014.16419

Nygard, C., & Clancy, A. (2018). Unsung heroes, flying blind-a metasynthesis of parents' experiences of caring for children with special health-care needs at home. *Journal of Clinical Nursing, 27*(15-16), 3179-3196. https://doi.org/10.1111/jocn.14512

Page, B. F., Hinton, L., Harrop, E., & Vincent, C. (2020). The challenges of caring for children who require complex medical care at home:‘The go between for everyone is the parent and as the parent that’s an awful lot of responsibility’. *Health Expectations*, 23, 1144-1154. https://doi.org/10.1111/hex.13092

Pierce, M., Hope, H. F., Kolade, A., Gellatly, J., Osam, C. S., Perchard, R., Kosidou, K., Dalman, C., Morgan, V., Di Prinzio, P., & Abel, K. M. (2019). Effects of parental mental illness on children's physical health: Systematic review and meta-analysis. *British Journal of Psychiatry*, 1-10. https://doi.org/10.1192/bjp.2019.216

Pinquart, M. (2018). Parenting stress in caregivers of children with chronic physical condition-a meta-analysis. *Stress and Health, 34*(2), 197-207. https://doi.org/10.1002/smi.2780

Ratliffe, C. E., Harrigan, R. C., Haley, J., Tse, A., & Olson, T. (2002). Stress in families with medically fragile children. *Issues in Comprehensive Pediatric Nursing, 25*(3), 167-188.

Rehm, R. S. (2013). Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012. *Nursing Outlook, 61*(5), 266-290.

Romley, J. A., Shah, A. K., Chung, P. J., Elliott, M. N., Vestal, K. D., & Schuster, M. A. (2017). Family-provided health care for children with special health care needs. *Pediatrics, 139*(1). https://doi.org/10.1542/peds.2016-1287

Ruggiero, K., Pratt, P., & Antonelli, R. (2019). Improving outcomes through care coordination: Measuring care coordination of nurse practitioners. *Journal of the American Association of Nurse Practitioners, 31*(8), 476-481. https://doi.org/10.1097/jxx.0000000000000276

Schall, T. E., Foster, C. C., & Feudtner, C. (2020). Safe work-hour standards for parents of children with medical complexity. *JAMA Pediatrics, 174*(1), 7-8. https://doi.org/10.1001/jamapediatrics.2019.4003

Scherer, N., Verhey, I., & Kuper, H. (2019). Depression and anxiety in parents of children with intellectual and developmental disabilities: A systematic review and meta-analysis. *PloS One, 14*(7).

Shippee, N. D., Shah, N. D., May, C. R., Mair, F. S., & Montori, V. M. (2012). Cumulative complexity: A functional, patient-centered model of patient complexity can improve research and practice. *Journal of Clinical Epidemiology, 65*(10), 1041-1051.

Shumskiy, I., Richardson, T., Brar, S., Hall, M., Cox, J., Crofton, C., Peltz, A., Samuels-Kalow, M., Alpern, E. R., Neuman, M. I., & Berry, J. G. (2018). Well-child visits of medicaid-insured children with medical complexity. *Journal of Pediatrics, 199*, 223-230.e222. https://doi.org/10.1016/j.jpeds.2018.04.003

Simon, T. D., Berry, J., Feudtner, C., Stone, B. L., Sheng, X., Bratton, S. L., Dean, J. M., & Srivastava, R. (2010). Children with complex chronic conditions in inpatient hospital settings in the United States. *Pediatrics, 126*(4), 647-655. https://doi.org/10.1542/peds.2009-3266

Spratling, R. (2017). Understanding the health care utilization of children who require medical technology: A descriptive study of children who require tracheostomies. *Applied Nursing Research, 34*, 62-65.

Szanton, S. L., Gill, J. M., & Allen, J. K. (2005). Allostatic load: A mechanism of socioeconomic health disparities? *Biological Research for Nursing, 7*(1), 7-15. https://doi.org/10.1177/1099800405278216

Thomson, J., Shah, S. S., Simmons, J. M., Sauers-Ford, H. S., Brunswick, S., Hall, D., Kahn, R. S., & Beck, A. F. (2016). Financial and social hardships in families of children with medical complexity. *Journal of Pediatrics, 172*, 187-193.e181. https://doi.org/10.1016/j.jpeds.2016.01.049

Vessey, J. A., DiFazio, R. L., Strout, T. D., & Snyder, B. D. (2017). Impact of non-medical out-of-pocket expenses on families of children with cerebral palsy following orthopaedic surgery. *Journal of Pediatric Nursing, 37*, 101-107. https://doi.org/10.1016/j.pedn.2017.07.006

Williamson, A., Lombardi, D. A., Folkard, S., Stutts, J., Courtney, T. K., & Connor, J. L. (2011). The link between fatigue and safety. *Accident Analysis & Prevention, 43*(2), 498-515. https://doi.org/https://doi.org/10.1016/j.aap.2009.11.011

Woolf-King, S. E., Anger, A., Arnold, E. A., Weiss, S. J., & Teitel, D. (2017). Mental health among parents of children with critical congenital heart defects: A systematic review. *J Am Heart Assoc, 6*(2). https://doi.org/10.1161/jaha.116.004862

Zhou, T., Yi, C., Zhang, X., & Wang, Y. (2014). Factors impacting the mental health of the caregivers of children with asthma in china: Effects of family socioeconomic status, symptoms control, proneness to shame, and family functioning. *Family Process, 53*(4), 717-730. https://onlinelibrary.wiley.com/doi/abs/10.1111/famp.12099

APPENDIX C

Social Support and Family Resources Influence Workload and Capacity in Parents of Children with Medical Complexity

Target Journal: *Nursing Research*

ABSTRACT

Background: Children with medical complexity (CMC) represent a small patient population with chronic, sometimes life-limiting conditions, functional limitations, and high health care use. Research examining factors that impact the lives of their families has been limited.

Objectives: This study examined relationships among family resources and social support, and how these variables impact parents' workload and their capacity to care for their CMC.

Methods: A non-experimental, cross-sectional, correlational design was conducted in a national sample of 106 parents of CMC diagnosed with critical congenital heart disease (CCHD). Three instruments were used to measure perceived family resources, perceived social support, and perceived workload and capacity to care for this medically complex population.

Results: Parent participants were largely white (84.9%), married/partnered (86.8%), biological (91.5%) mothers (98.1%) ranging in age from 23-47 years of age with incomes of $50,000 or more (61.3%). The majority of CMC (40.6%) had between 1-3 other health conditions in addition to CCHD that required an average of 5 daily medications, and 2 pieces of medical technology, and 7 health care providers to provide specialty care. Parents reported a mean of 72.25 weekly hours providing direct care to manage their CMC's health conditions, and a mean of 8.57 weekly hours to coordinate care. Higher family resources were associated with less workload ($r = -0.47$, $p < .001$) and increased capacity ($r = .54$, $p < .001$), while more social support was associated with higher capacity ($r = .44$, $p < .001$). Decreased capacity was associated with an increase in workload (r = -

0.33, p <.001). Social support and family resources accounted for 24.5% of the variance in workload and accounted for 30.1% of the variance in capacity.

Discussion: The more optimal parents' social support and family resources are, the less likely their workload is high, and the more likely they feel able to meet workload demands. Parents more capable of meeting workload demands do indeed perceive less workload. Results emphasize the importance of universal screening for social support and family resources. Pre-discharge training and simulation programs, in addition to expanding the availability of medical homes responsible for care coordination can alleviate parents' workload and enhance parents' capacity to meet workload demands.

Key words: child; heart defects, congenital; resources; social support; workload.

INTRODUCTION

Children with medical complexity (CMC), whose medical diagnoses vary in number and type, represent patients with high health care use due to their chronic health conditions that require a number of health care services (Cohen et al., 2011; Kuo et al., 2011). Conditions such as critical congenital heart disease (CCHD) are examples of diagnoses that can be life-long and severe. Although CMC represent less than one percent of all children in the United States (Berry et al., 2014), their care costs are disproportionally high, accounting for health care spending between $50-$110 billion annually, with up to 80 percent of this cost due to hospitalizations (Cohen et al., 2012; Lassman et al., 2014). While studies focused on CMC health outcomes are increasing, research examining the broader impact on the family is limited. Understanding factors that alleviate parental workload to care for the complex needs of CMC and that enhance parents' capacity to manage the care that is required is particularly important for parents who have consistently described this care as demanding and stressful (Javalkar et al., 2017) . Moreover, parents of CMC, together with health care providers who care for CMC, have identified "parents' ability to keep up with the demand" as an important issue that warrants further study when examining research priorities (Fayed et al., 2019, p. 1097).

Parents are responsible for managing complex medication schedules and medical equipment such as feeding tubes, tracheostomies, oxygen, and/or ventilators (Mooney-Doyle & Lindley, 2020). Parents take time away from paid work to attend numerous appointments with health care specialists which are not always conveniently located nearby (Cady & Belew, 2017; Mooney-Doyle & Lindley, 2019). Despite the time parents take to provide care to their CMC, these patients are more likely to have several unmet medical needs met (Kuo et al., 2011). This workload, sometimes referred to in the scientific literature as “demands” or “burdens”, is directly related to the daily care that parents provide to their CMC (Javalkar et al., 2017; Mooney-Doyle & Lindley, 2020). This pile-up of demands affects families financially and socially (Thomson et al., 2016). The workload related to caregiving is *in addition* to other routine daily responsibilities such as caring for other children, laundry, and meal preparation, while sometimes working outside the home.

Parents of CMC are negatively impacted economically. Studies describing the experiences of parents of CMC consistently report financial hardships that go hand-in-hand with the time required to care for CMC (Kuo et al., 2014). Parents caring for CMC report costly, out-of-pocket expenditures (Vessey et al., 2017), and more than half of parents stop working outside the home to care for their CMC (Kuo et al., 2011; Mooney-Doyle & Lindley, 2019). More than half of families experience financial problems which was found to be significantly associated with poverty in one study (Mooney-Doyle & Lindley).

In addition, parents of CMC experience social hardships. Many parents describe negative impacts when social support is lacking, which includes feeling isolated and poorly understood by others (Caicedo, 2014; Mesman et al., 2013). Families of CMC are more likely to report little expectation of receiving any help from family or friends when assistance is needed (Thomson et al., 2016). Understaffed and underfunded community and home care services make the scarcity of social support that much more difficult for parents of CMC (Berry et al., 2014; Kuo et al., 2011). Parents of CMC describe the loss of free time to socialize or make friends, and the inability to get breaks from caring for their CMC (Whiting, 2013). Indeed, the complicated nature of caring for CMC places parents at increased risk for experiencing stressors associated with the workload that CMC require at home, in addition to the social and economic stressors that these parents are already known to experience (Pinquart, 2018; Thomson et al., 2016).

Social support improves coping and parenting self-efficacy and deters caregiver burden and depressive symptoms in some pediatric patient populations (Leahy-Warren et al., 2012; Tak & McCubbin, 2002). Social support and family resources, which includes financial resources, may play a role in easing the burdens experienced by these families. However, these relationships have rarely been explored in the CMC population. Therefore, the purpose of this study was to examine relationships among family resources, social support, workload, and capacity in parents of children with medical complexity. Eligible participants were 18 years or older, English-speaking, and the biological, adoptive, or foster parent (single or partnered) who considered themselves to be the primary caregiver of a child 6 months through 5 years of age who fits the definition of CMC by Cohen et al. (2011). To provide a thread of consistency and homogeneity in a group whose medical diagnoses are often heterogeneous, this study included CMC diagnosed CCHD as defined by the Centers for Disease Control (Centers for Disease Control, 2020).

Theoretical Framework

The theoretical framework used to guide this study was an integrated model derived from the Family Management Style Framework (Knafl et al., 2012) and the Cumulative Complexity Model (Shippee et al., 2012). The model (Lawrence & Spratling,

2022) incorporates the concepts of workload and capacity from the Cumulative Complexity Model (CCM) to enhance the dimensions of effort and ability within the Family Management Style Framework (FMSF) as seen in Figure 1. The FMSF provides a framework for understanding how families respond to, manage, and incorporate a child's chronic illness into everyday family life. According to the FMSF, a family's social network and family resources contribute to the ease or difficulty in how a family manages a child's chronic illness (Knafl et al., 2012). The dimensions of ability and effort from the FMSF are similar to the constructs of workload and capacity from the CCM. However, the CCM goes further to posit that when workload outweighs capacity, poor health outcomes can occur (Shippee et al., 2012). Given theoretical frameworks have rarely been included in studies focused on parents and their CMC, this framework fills an important gap in the CMC literature.

MATERIALS AND METHODS

A non-experimental, cross-sectional, correlational design was used to examine relationships among family resources, social support, workload, and capacity in parents of children with medical complexity. Institutional review board approval was obtained from Georgia State University prior to participant recruitment and data collection. The target population for this study were parents of CMC diagnosed with CCHD between the ages of 6 months and 5 years of age. The definition of CMC by Cohen et al. (2011) was used. Selection of CMC between the age of 6 months through 5 years of age provided an acceptable age range to allow for an adequate sample size, and during which timeframe a large proportion of CCHD surgical procedures take place. At the same time, this age range excluded the newborn period during which time is known be very stressful for parents of a new child with medical complexities or CCHD (Woolf-King et al., 2018).

A power analysis using G*Power (Faul et al., 2009), was used to determine the sample size for this study based on the specific aims and research questions of this study. Power analysis was conducted using bivariate correlation and multiple regression. Using a moderate effect size, a power of .80, and an alpha level of .05, the sample size was determined by selecting the largest calculated sample size which is 92. After accounting for a 15% incomplete participant survey rate, the final target sample size was 106.

All aspects of this study were conducted remotely. A non-random (convenience and snowball) sample of 106 parents who met the inclusion criteria were recruited through the use of a digital flyer shared through local and national social media platforms and CMC parent support groups which allowed for a potentially more diverse sample of CMC parents. Data collected in Qualtrics included demographic data, participants' responses to three instruments, and the email addresses for participants who wished to receive participation renumeration in the form of a $20 e-gift card.

Measures and Instruments

Demographic Variables

A demographic survey developed by the researchers was used to assess a number of parent and CMC characteristics and had a Flesch-Kincaid grade level of 6.2.

Family Resources

Family resources was measured using the Revised Family Resource Scale (FRS-R) (Van Horn et al., 2001). This 20-item self-report scale measures the perception of available family resources including money, time for self and family, and basic needs such as food, housing, and clothing. Using a 5-point response scale ranging from 1 (*never adequate*) to 5 (*always adequate*), total scores are obtained by summing responses with a potential total score of 20 to 100, with higher scores representing more perceived family resources. Internal consistency reliability for this instrument has ranged from .72 - .84, and validity has been established by exploratory and confirmatory factor analysis that resulted in the revised instrument that was stable using four factors (Van Horn et al.).

Social Support

Social support was measured using the The Personal Resource Questionnaire 2000 (PRQ2000) (Weinert, 2003). This instrument uses15 positively worded items to measure the overall level of perceived social support. Using a 7-point Likert scale ranging from 1 (*strongly disagree*) to 7 (*strongly agree*), total scores are achieved by summing the 15 responses for a potential total score ranging from 15 to 105, with higher scores representing higher levels of perceived social support. Internal consistency reliability for this instrument has ranged from .87 to .93 from different adult samples and has undergone robust construct validity examination (Weinert).

Parental Workload and Capacity

The Family Management Measure (FaMM) measures parents’ perceptions of a chronically ill child’s treatment regimen and its incorporation into day-to-day life (Knafl et al., 2011). Two of the six subscales of the FaMM, used to measure perceived effort (workload) and perceived ability (capacity) on a 5-point Likert scale, were used for this study. The condition management effort (workload) subscale measures parents’ perceptions of the workload needed to manage their children’s chronic conditions using 4 items, with higher values signifying greater effort needed to manage the condition.

Internal consistency reliability ranged between .74 for mothers and .78 for fathers (Knafl et al). The condition management ability (capacity) subscale measures parents' perceptions of their competence to care for children's' chronic illnesses using 12 items, and higher values are associated with parents' view of themselves as more capable of managing the chronic condition. Internal consistency reliability for this subscale ranged between .72 for mothers and .73 for fathers (Knafl et al.). Construct validity has been supported by significant correlations with other family functioning measures (Knafl et al.).

Statistical Analysis

All data was analyzed using IBM SPSS Version 27. The data was checked for quality, missing values, outliers, and normality. Any outliers and missing values were validated with all study participants via email. Prior to addressing the research questions, assumptions of regression were tested for normality, outliers, and homoscedasticity and lack of multicollinearity. Internal consistency reliability was calculated for each of the instruments used in this study. Descriptive statistics were used to summarize the sample characteristics and responses to questions on the demographics form. Frequencies and percentages were presented for categorical variables and means and standard deviations were presented for continuous variables. Bivariate correlation and multiple regression was used to examine relationships between the independent and dependent variables including testing for moderation effects. A significance value of .05 was used for all statistical analyses.

RESULTS

Sample Characteristics

The 106 parent participants of children with medical complexity represented 40 of the 50 United States, the majority (13.2%) of which came from Texas. Parent participants' mean age was 32.95 (SD=5.06) and ranged between 23 and 47 years of age. The mean number of dependent children at home, which included their CMC, was 2.56 (SD=1.47). The majority of participants were married/partnered (86.8%) mothers (98.1%) who were the biologic parent (91.5%) of their CMC. The majority (84.9%) of parent participants were white, and just over half (51%) of parents worked outside the home, with 30.2% working full-time.

The majority (37.7%) of parent participants were college graduates with a working car (92.5%) to use for transporting their CMC to health care-related appointments, and with a reported family income of $50,000 or greater (61.3%). The majority of parent participants reported a history of changing their job status to care for their CMC, with 62 (58.5%) taking a leave of absence from their job due to their child's health condition, and 70 (66%) cutting down on work hours because of their child's condition.

Characteristics of CMC

The mean age in months of CMC was 33.78 (SD=18.99) and ranged between 6 and 71 months of age. The majority of CMC were white (71.7%) males (59.4%) with public health insurance (46.2%). All CMC had some type of critical congenital heart disease (CCHD) that required surgical intervention in the first year of life, the three most common of which were ventricular septal defect (39.6%), single ventricle physiology

(31.1%) and Tetralogy of Fallot (28.3%). The CMC in this study had a number of other health conditions, with only 15 (14.2%) reporting no other health conditions aside from their CCHD diagnosis. Forty-three (40.6%) CMC had between 1-3 other health conditions, whereas 31 (29.2%) had between 4-6 other health conditions beside their CCHD diagnosis. Seventeen (15.9%) CMC had 7 or more health conditions. The three most common conditions included developmental delay (63.2%), speech/language delay (52.8%), and genetic conditions other than Trisomy 21 (26.4%).

CMC used a mean of 5.67 (SD=4.56) daily medications. Forty (37.7%) children need to be given medication three or more times a day, with the majority (54.7%), needing medication once or twice a day. All CMC in this study used at least one piece of life-sustaining medical equipment, with a mean of 2.53 (SD=2.32) pieces of medical equipment, the most common which was respiratory equipment (49.1%). See Table 1 for the demographic characteristics of CMC.

Table 1

Parent Participant (N=106)

Characteristics	*M*	*(SD)*	*Observed Range*	*N*	*(%)*
Parent					
Age (years)	32.95	5.06	23-47	106	
Number of children at home[a]	2.56	1.47	1-8		
Parent Type				104	
Mother				2	(98.1%)
Father				97	(1.9%)
Biologic				1	(91.5%)
Foster				8	(.9%)
Adoptive					(7.5%)
Marital Status					
Married/Partnered				92	(86.8%)
Single/Divorced				14	(13.2%)

Race/Ethnicity		
A. Indian or Alaska Native	4	(3.8%)
Asian	4	(3.8%)
Black or African American	3	(2.8%)
Hispanic	10	(9.4%)
Hawaiian or Pacific Islander	1	(.9%)
White, Non-Hispanic	90	(84.9%)
Two or more races	4	(3.8%)
Employment Status		
Full-time	32	(30.2%)
Part-time	22	(20.8%)
Unemployed	52	(49.1%)
Education Level		
High school graduate	10	(9.4%)
Some college	31	(29.2%)
College graduate	40	(37.7%)
Post-graduate study	25	(23.6%)
Working car for CMC		
transportation	4	(3.8%)
No car	98	(92.5%)
Own car	4	(3.8%)
Unreliable car		
Household Income		
< $50,000	41	(38.7%)
≥ $50,000	65	(61.3%)
History of Job Status Change		
Left job to care for CMC	39	(36.8%)
Took leave to care for CMC	62	(58.5%)
Cut down hours to care for	70	(66%)
CMC	28	(26.4%)
Avoided promotion CMC	49	(46.2%)
Avoided job change to keep insurance		

Table 2

CMC Characteristics (N=106)

Characteristics	*M (SD)*	*Observed Range*	*N*	*(%)*
Age in months	33.77 (19)	6-71		
Gender				
Males			63	(59.4%)
Females			43	(40.6%)
Ethnicity				
A. Indian or Alaska Native			3	(2.8%)
Asian			3	(2.8%)
Black or African American			4	(3.8%)
Hawaiian or Other Pacific Islander			2	(1.9%)
White, non-Hispanic			76	(72.4%)
Hispanic			13	(12.3%)
Two or more races			17	(16.2%)
Insurance Type				
Public			49	(46.2%)
Private			25	(23.6%)
Both			32	(30.2%)
CCHD Type				
VSD			42	(39.6%)
Single Ventricle			33	(31.1%)
TOF			30	(28.3%)
PA			20	(18.9%)
DORV			18	(17%)
TGA			12	(11.3%)
CoA			11	(10.4%)
PVS			9	(8.5%)
CAVC			7	(6.6%)
IAA			5	(4.7%)
TA			3	(2.8%)
TAPVR			2	(1.9%)
Ebstein anomaly			2	(1.9%)
Other Health Conditions				
None			15	(14.2%)
Between 1-3			43	(40.6%)
Between 4-6			31	(29.2%)

More than 6			17	(15.9%)
Number of Daily Medications	5.67 (4.56)	1-25		
Medication Frequency				
No medications			8	(7.5%)
1-2 times a day			58	(54.7%)
Three or more times a day			40	(37.7%)
Number of Medical Equipment	2.53 (2.32)	1-12		
Medical Equipment Type				
Cardiovascular			10	(9.4%)
Respiratory			52	(49.1%)
Digestive			79	(74.5%)
Medication			12	(11.3%)
Mobility			19	(17.9%)
Hearing			3	(2.8%)
Other			7	(6.6%)

Note. VSD=Ventricular septal defect; TOF=Tetralogy of Fallot; PA=Pulmonary atresia; DORV=Double outlet right ventricle; TGA=Transposition of the great arteries; CoA=Coarctation of the aorta; PVS=Pulmonary vein stenosis; CAVC=Complete atrioventricular canal; IAA=Interrupted aortic arch; TA=Truncus arteriosus; TAPVR=Total anomalous pulmonary venous return.

Care Requirements of CMC

Parents reported a mean of 72.25 (SD=49.19) weekly hours providing direct care to manage their CMCs' conditions, and a mean of 8.57 (SD=7.57) hours weekly to coordinate care of their CMC. The majority of parents (37.7%) reported having no one other than themselves to provide care for their CMC, while 32% of the parent sample had just one other person to assist them in unpaid caregiving. The majority of CMC (62.3%) received no weekly paid nursing hours to provide care at home. Of the remaining 37.7%

who did receive paid nursing hours, the mean number of weekly hours received was 18.02 (SD=34.79). The CMC in this study required a mean of 7.2 (SD=3) health care specialists to help manage their health care conditions. CMC attended a mean of 5.22 (SD=5.22) well visits in the prior year and a mean of 25.48 (SD=36.24) visits to subspecialists in the prior year, resulting in a mean of 6.44 (SD=7.56) estimated weekly hours traveling to see CMC health care providers. More than half (55.7%) of the parent sample reported paying an estimated $1000 or more annually for out-of-pocket expenses for their CMC, with just over 25% reporting estimated out-of-pocket costs of $5000 or more. See Table 2 for characteristics of the care requirements for CMC.

Table 3

Care Requirements of CMC (N=106)

Characteristics	*M*	*(SD)*	*Observed Range*	*N*	*%*
CMC paid weekly nursing hours [a]	18.02	34.79	0-168	41	38.7
Weekly direct care hours	72.25	49.19	1-168		
Weekly CC hours	8.57	7.57	1-33		
Number unpaid caregivers	1.09	1.13	1-4		
Number annual well visits	5.22	5.22	0-30		
Number annual specialty visits	25.48	36.24	1-310		
Number of specialists	7.2	3.00	2-19		
Weekly travel hours	6.44	7.56	0-45		
Out-of-pocket Medical Expenses[b]					
$0				9	(8.5%)
$1 to $499				25	(23.6%)
$500 to $999				13	(12.3%)
$1000 to $5000				32	(30.2%)
>$5000				27	(25.5%)

Note. [a]Number and % of CMC who receive paid nursing hours. [b]Annual estimates.

Reliability of Instruments

All instruments and subscales had acceptable internal reliability consistency defined as a minimum alpha coefficient of >.70.

Personal Resource Questionnaire 2000. The 15-item PRQ 2000 was completed by all 106 participants with a mean total score of 77.89 (SD=17.91), a minimum score of 15 and a maximum score of 105. The Cronbach's alpha reliability coefficient was acceptable at .93.

Revised Family Resource Scale. All 106 parent participants completed the 20-item Revised Family Resource Scale resulting in a mean total score of 72.58 (SD=12.34) with a minimum score of 38 and a maximum score of 75. The Cronbach's alpha reliability coefficient for this scale was acceptable at .91.

Family Management Measure (FaMM). All 106 parent participants completed the 4-item condition management effort subscale of the FaMM to measure perceived workload. The mean subscale score was 16.75 (SD= 2.77), a minimum score of 10, and a maximum score of 20. The Cronbach's alpha for this subscale was .70. In addition, all participants completed the 12-item condition management ability subscale of the FaMM, used to measure perceived capacity. The mean score for this subscale was 43.89 (SD=6.56), the minimum score was 27 and the maximum score was 58. The Cronbach's alpha for this subscale was .71.

Relationships among Major Study Variables

Correlation analyses were used to assess relationships between the major variables. Cohen's standard was used to evaluate the strength of the relationships. Based on bivariate correlations, higher perceived family resources were significantly associated with higher perceived social support (r = .55, p < .001), decreased workload (r = -0.47, p < .001) and higher perceived capacity (r = .54, p < .001). While higher perceived social support was not significantly associated with perceived workload, it was significantly associated with higher perceived capacity (r = .44, p < .001). Perceived capacity was negatively correlated with perceived workload (r = - 0.33, p < .001), indicating that as perceived capacity decreased, workload increased.

Impact of Social Support and Family Resources on Workload

To assess the impact of social support and family resources on parents' perceived workload, multiple regression was conducted to test the relationships as seen in Table 3. Because the participant sample was very homogeneous with regard to race, gender, and education, and because this study was exploratory, no covariates were used in any of the regression models. In Model 1, there was no statistically significant association found between perceived social support and workload (β = -0.14, SE = .02, p = .140) demonstrating that perceived social support did not influence perceived workload in this study. Model 2 findings indicated that there is a negative relationship between family resources and workload (β = -0.49, SE = .03, p < .001) and family resources accounted for 23% of the variability in workload (Adj. R^2 = .230). Therefore, perceived workload decreases as perceived family resources increase. Model 3 analyses are similar to Models 1 and 2 in that there was no significant relationship between social support and workload, but there was a statistically significant relationship between family resources and workload (β = -0.59, SE = .04, p < .001). Taken together, social support and family resources account for 24.5% of the variation in workload (Adj. R^2 = .245).

Table 4

Workload, Regression

	Model 1			Model 2			Model 3		
	β	SE	Sig.	β	SE	Sig.	β	SE	Sig.
Support									
Social support	-.14	.02	.140				.18	.02	.081
Family resources				-0.49	.03	<.001***	-0.59	.04	<.001***
Constant	-	1.25	<.001***	-	1.82	<.001***	-	1.80	<.001***
Adj. R^2		.011			.230			.245	

$p<0.05$ *, $p<0.01$ **, $p<0.001$ *

Impact of Social Support and Family Resources on Capacity

Table 4 contains the regression analysis used to test the relationships between social support, family resources, and perceived capacity. In Model 1, social support had a significant positive relationship to capacity ($\beta = .44$, SE .03, $p < .001$) with social support accounting for 18.2% of the variability in perceived capacity. Thus, as social support increases, so too does perceived capacity. Model 2 findings indicate a significant positive relationship between family resources and capacity ($\beta = .54$, SE .07, $p < .001$), with family resources accounting for 27.9% of the variability in perceived capacity. This indicates that with more adequate family resources, perceived capacity increases. Model 3 findings demonstrate that when both social support and family resources are entered simultaneously in the regression model, they together account for 30.1% of the variability in perceived capacity (Adj. $R^2 = .301$). The β weights indicated that both social support ($\beta = .20$, SE = .04, $p < .05$) and family resources ($\beta = .42$, SE= .08, $p < .001$) significantly predict capacity, with family resources having a greater effect on capacity than social support.

Table 5

Capacity, Regression

	Model 1			Model 2			Model 3		
	β	SE	Sig.	β	SE	Sig.	β	SE	Sig.
Support									
Social support	.44	.03	<.001***				.20	.04	.042*
Family resources				.54	.07	<.001***	.42	.08	<.001***
Constant	-	2.59	<.001***	-	4.13	<.001***	-	4.07	<.001***
Adj. R^2		.182			.279			.301	

p<0.05 *, p<0.01 **, p<0.001 ***

Relationship between Workload and Capacity

Simple linear regression was used to examine the relationship between workload and capacity. As seen in Table 5, workload significantly negatively predicts capacity ($\beta = -0.33$, $p < .001$), and 9.9% of the variance in workload can be explained by capacity (Adj. $R^2 = .099$). Thus, as capacity diminishes, workload increases.

Table 6

Workload, Regression

	β	Model SE	Sig.
Capacity	-0.33	.04	<.001 ***
Constant	-	1.73	<.001 ***
Adj. R^2		.099	

p<0.05 *, p<0.01 **, p<0.001 ***

DISCUSSION

Findings from this study indicate that adequate family resources ease the perceived workload of parents with CMC and is associated with more capacity to manage the work required of parents. While social support does not influence workload, it is associated with higher perceived capacity. Moreover, when capacity is diminished, perceived workload is higher for these parents. Results of this study add to the body of research reporting the challenges experienced by parents of CMC. Specifically, this study begins to fill a knowledge gap about the quantitatively measurable relationship between workload and capacity, and how social support and family resources impact parents' workload and capacity.

To our knowledge, this study is the first to explore relationships between social support, family resources, workload, and capacity in parents of CMC with CCHD. Given the challenges in caring for CMC with CCHD at home, it is not surprising that when perceived family resources were adequate, workload was less prevalent, and capacity was higher. Nor was it surprising that decreased capacity is associated with more workload. Taken together, these findings highlight the need to address factors for supporting families in ways that enhance their capacity and ease workload.

Research has demonstrated that caring for children with medical complexity affects the relationships and social lives of the families that care for them. Parents of technology dependent children have reported that it is often easier to remain at home (Rehm, 2013). This may explain why there is no statistically significant relationship between social support and workload, in that parents of CMC rely on few others to help them with the day-to-day care, leaving little time for these families to socialize. On the other hand, parents who have a strong social support network have reported that extended family members who assist with day-to-day tasks allows parents to focus on the care of their CMC (Foster et al., 2022). This may be another explanation for the absence of a significant relationship between social support and workload.

Building on prior research describing the financial challenges that parents of CMC face, this study demonstrates that a lack of adequate family resources has a significant impact on these parents. Specifically, a lack of perceived family resources significantly predicted workload. This finding lends strong support for universal screening of family resources so that workload can potentially be decreased. Screening all parents is important because parents of CMC are already stressed and overburdened by their daily care responsibilities from which they get little relief.

Perceived social support and perceived family resources are contextual influences that can hinder or support parents' management of their CMCs' conditions (Knafl et al., 2012). Both variables were significantly correlated with workload and capacity in this study. These findings support the usefulness of the theoretical framework used in this study to understand the challenges faced by parents of CMC.

The results of this study must be considered in the context of several noted limitations. Recruitment efforts were done solely online which can affect the representativeness of the sample. Notably, participants in this study were largely homogeneous with regard to marital status, race, education, and gender, which limits the generalization of our findings. Although this was a national sample, the non-random sample consisted of a more than 90% white, non-Hispanic, married female participant group. Random sampling would limit biases and strengthen future study results. A second limitation is that all study questionnaires and instruments were self-reported and therefore could not be confirmed by in-person observations or medical records. Self-reporting may have contributed to recall and response bias. Face-to-face interviewing may have provided greater accuracy of the information shared by participants. Lastly, it is difficult to know how the COVID-19 pandemic affected participants' lives. No measures exist that differentiate between workload due to providing care to CMC versus workload due to challenges from the pandemic.

Nurses are in a prime position to assess for adequate social supports, family resources and parental depressive symptoms. Such screening is imperative to assist families in obtaining the supports necessary. Respite care and work-hour safety standards for parents of CMC must be developed with advocacy and policy support. This is important based on what is known about the unpaid care hours that parents provide for medically fragile children. CMC experts suggest that these parents should not work to care for their children more than 9 consecutive hours based on what is known about the association of adverse events and performance-impairing fatigue in other safety-sensitive occupations such as nursing, transportation, and emergency services (Schall et al., 2020; Williamson et al., 2011). These experts argue that the work provided by parents to care for their CMC should be held to the same safety standards used in other high-risk fields to prevent exhaustion and burn out. Further nursing research is needed to develop and test interventions aimed at minimizing workload and facilitating capacity in parents of CMC.

Conclusion

The findings from this study add to the limited body of knowledge about the relationship between social and family supports and perceived workload for parents of CMC. This study, guided by an integrated theoretical framework, was the first to explore relationships between social support and family resources, and how they impact workload and capacity. These findings highlight the need for universal screening for social support and family resources. In addition, these findings emphasize the need for policies and care models that reduce workload while building capacity to manage the workload required to care for CMC at home. Medical homes that provide care coordination to these parents are one effective way to minimize workload, yet they are not a widely available resource. Until medical homes universally exist for all CMC, interventions aimed at building capacity and minimizing workload for parents are needed so that the health and well-being of CMC and their parents can be strengthened.

References

Berry, J. G., Hall, M., Neff, J., Goodman, D., Cohen, E., Agrawal, R., Kuo, D., & Feudtner, C. (2014, Dec). Children with medical complexity and medicaid: Spending and cost savings. *Health Affairs, 33*(12), 2199-2206. https://doi.org/10.1377/hlthaff.2014.0828

Cady, R., & Belew, J. (2017). Parent perspective on care coordination services for their child with medical complexity. *Children, 4*(6), 45.

Caicedo, C. (2014, Nov-Dec). Families with special needs children: Family health, functioning, and care burden. *Journal of the American Psychiatric Nurses Association, 20*(6), 398-407. https://doi.org/10.1177/1078390314561326

Centers for Disease Control. (2020). *Congenital heart defects*. Retrieved October 1, 2020 from https://www.cdc.gov/ncbddd/heartdefects/cchd-facts.html

Cohen, E., Berry, J. G., Camacho, X., Anderson, G., Wodchis, W., & Guttmann, A. (2012, Dec). Patterns and costs of health care use of children with medical complexity. *Pediatrics, 130*(6), e1463-1470. https://doi.org/10.1542/peds.2012-0175

Cohen, E., Kuo, D. Z., Agrawal, R., Berry, J. G., Bhagat, S. K., Simon, T. D., & Srivastava, R. (2011, Mar). Children with medical complexity: An emerging population for clinical and research initiatives. *Pediatrics, 127*(3), 529-538. https://doi.org/10.1542/peds.2010-0910

Faul, F., Erdfelder, E., Buchner, A., & Lang, A. G. (2009, Nov). Statistical power analyses using g*power 3.1: Tests for correlation and regression analyses.

Behavior Research Methods, 41(4), 1149-1160. https://doi.org/10.3758/brm.41.4.1149

Fayed, N., Guttmann, A., Chiu, A., Gardecki, M., Orkin, J., Hamid, J. S., Major, N., Lim, A., & Cohen, E. (2019, Sep). Family-provider consensus outcomes for children with medical complexity. *Developmental Medicine and Child Neurology, 61*(9), 1093-1100. https://doi.org/10.1111/dmcn.14110

Foster, C. C., Shaunfield, S., Black, L. E., Labellarte, P. Z., & Davis, M. M. (2022). Improving support for care at home: Parental needs and preferences when caring for children with medical complexity. *Journal of Pediatric Health Care, 36*(2), 154-164.

Javalkar, K., Rak, E., Phillips, A., Haberman, C., Ferris, M., & Van Tilburg, M. (2017). Predictors of caregiver burden among mothers of children with chronic conditions. *Children, 4*(5), 39.

Knafl, K., Deatrick, J. A., Gallo, A., Dixon, J., Grey, M., Knafl, G., & O'Malley, J. (2011, Jun). Assessment of the psychometric properties of the family management measure. *J Pediatr Psychol, 36*(5), 494-505. https://doi.org/10.1093/jpepsy/jsp034

Knafl, K. A., Deatrick, J. A., & Havill, N. L. (2012, Feb). Continued development of the family management style framework. *Journal of Family Nursing, 18*(1), 11-34. https://doi.org/10.1177/1074840711427294

Kuo, D. Z., Cohen, E., Agrawal, R., Berry, J. G., & Casey, P. H. (2011, Nov). A national profile of caregiver challenges among more medically complex children with special health care needs. *Archives of Pediatrics and Adolescent Medicine, 165*(11), 1020-1026. https://doi.org/10.1001/archpediatrics.2011.172

Kuo, D. Z., Goudie, A., Cohen, E., Houtrow, A., Agrawal, R., Carle, A. C., & Wells, N. (2014). Inequities in health care needs for children with medical complexity. *Health Affairs, 33*(12), 2190-2198.

Lassman, D., Hartman, M., Washington, B., Andrews, K., & Catlin, A. (2014). Us health spending trends by age and gender: Selected years 2002–10. *Health affairs, 33*(5), 815-822.

Lawrence, P. R., & Spratling, R. (2022, Feb 1). A theory for understanding parental workload and capacity to care for children with medical complexity. *Res Theory Nurs Pract, 36*(1), 34-46. https://doi.org/10.1891/rtnp-2022-0026

Leahy-Warren, P., McCarthy, G., & Corcoran, P. (2012). First-time mothers: Social support, maternal parental self-efficacy and postnatal depression. *Journal of clinical nursing, 21*(3-4), 388-397.

Mesman, G. R., Kuo, D. Z., Carroll, J. L., & Ward, W. L. (2013, 2013/11/01/). The impact of technology dependence on children and their families. *Journal of Pediatric Health Care, 27*(6), 451-459. https://doi.org/https://doi.org/10.1016/j.pedhc.2012.05.003

Mooney-Doyle, K., & Lindley, L. C. (2019). The association between poverty and family financial challenges of caring for medically complex children. *Nursing Economics, 37*(4), 198-208.

Mooney-Doyle, K., & Lindley, L. C. (2020). Family and child characteristics associated with caregiver challenges for medically complex children. *Family and Community Health, 43*(1), 74-81.

Pinquart, M. (2018, Apr). Parenting stress in caregivers of children with chronic physical condition-a meta-analysis. *Stress and Health, 34*(2), 197-207. https://doi.org/10.1002/smi.2780

Rehm, R. S. (2013). Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012. *Nursing outlook, 61*(5), 266-290.

Schall, T. E., Foster, C. C., & Feudtner, C. (2020). Safe work-hour standards for parents of children with medical complexity. *JAMA Pediatrics, 174*(1), 7-8. https://doi.org/10.1001/jamapediatrics.2019.4003

Shippee, N. D., Shah, N. D., May, C. R., Mair, F. S., & Montori, V. M. (2012). Cumulative complexity: A functional, patient-centered model of patient complexity can improve research and practice. *Journal of Clinical Epidemiology, 65*(10), 1041-1051.

Tak, Y. R., & McCubbin, M. (2002). Family stress, perceived social support and coping following the diagnosis of a child's congenital heart disease. *Journal of advanced nursing, 39*(2), 190-198.

Thomson, J., Shah, S. S., Simmons, J. M., Sauers-Ford, H. S., Brunswick, S., Hall, D., Kahn, R. S., & Beck, A. F. (2016, May). Financial and social hardships in families of children with medical complexity. *Journal of Pediatrics, 172*, 187-193.e181. https://doi.org/10.1016/j.jpeds.2016.01.049

Van Horn, M. L., Bellis, J. M., & Snyder, S. W. (2001). *Family resource scale-revised: Psychometrics and validation of a measure of family resources in a sample of* *low-income families* [doi:10.1177/073428290101900104]. Clinical Psychology Publishing Co.

Vessey, J. A., DiFazio, R. L., Strout, T. D., & Snyder, B. D. (2017, Nov - Dec). Impact of non-medical out-of-pocket expenses on families of children with cerebral palsy following orthopaedic surgery. *Journal of Pediatric Nursing, 37*, 101-107. https://doi.org/10.1016/j.pedn.2017.07.006

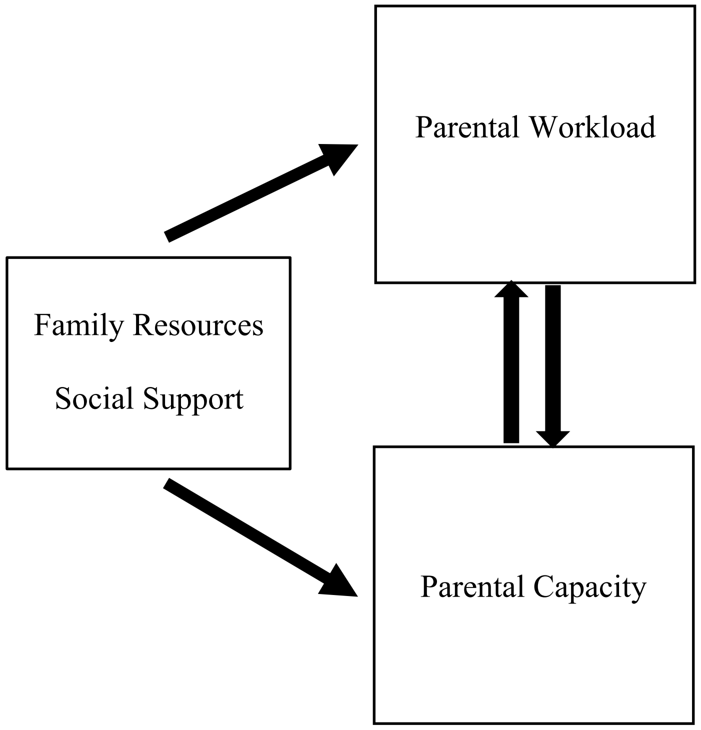
Weinert, C. (2003). Measuring social support: Prq2000. *Measurement of nursing outcomes, 3*, 161-172.

Whiting, M. (2013). Impact, meaning and need for help and support: The experience of parents caring for children with disabilities, life-limiting/life-threatening illness or technology dependence. *Journal of Child Health Care, 17*(1), 92-108.

Woolf-King, S. E., Arnold, E., Weiss, S., & Teitel, D. (2018, Jul). "There's no acknowledgement of what this does to people": A qualitative exploration of mental health among parents of children with critical congenital heart defects. *J Clin Nurs, 27*(13-14), 2785-2794. https://doi.org/10.1111/jocn.14275

Figure 1

Derived Theory for Understanding the Influence of Family Resources and Social Support on Parental Workload and Capacity to Care for Children with Medical Complexity



APPENDIX D

Supplemental Material – Recruitment Flyer

PARENT VOLUNTEERS NEEDED

for a study involving parents of children with medical complexity who also have certain types of heart defects

You may qualify if:

*** You are 18 years of age or older**
*** You have a child with medical complexity who is between 6 months and 5 years of age**
*** Your child uses medical technology (like a feeding tube)**

If you qualify, you will receive a $20 gift card for completing on-line questionnaires that may take up to 1 hour to complete.

If you are interested in participating, email, call, or text.

For questions, contact Patricia R. Lawrence, PhD (c), RN, MSN
Phone number: 774-292-9234
Email: plawrence4@student.gsu.edu

APPENDIX D

Supplemental Material – Demographics Questionnaire

Parent information

1. Parent age in years: _____
2. Are you (check one): Biologic parent_____ Foster parent _____ Adoptive parent _____
3. Gender: Male _____ Female _____
4. Marital Status: Single __ Married __ Separated __Divorced __Widowed __ Not married but living with a partner ___
5. Education (Check one):
    a. Less than 9th grade _____
    b. Some high school _____
    c. High school graduate _____
    d. Some college _____
    e. College graduate _____
    f. Post-graduate study _____
6. Do you have a working car that you can depend on to take your child to health care visits? Yes _____ No _____
7. Your residence zip code _____
8. Race:
    a. Asian _____
    b. African American/Black _____
    c. American Indian or Alaska Native _____
    d. Caucasian _____
    e. Native Hawaiian or Other Pacific Islander _____
    f. Two or more races _____
9. Ethnicity: Are you Hispanic or Latino of any race? Yes _____ No _____
10. Annual Household Income (check one):
    a. Less than $10,000 _____
    b. $10,001-$20,000 _____
    c. $20,001-40,000 _____
    d. $40,001-$50,000 _____
    e. Between $50,001 and $75,000 _____
    f. Between $75,001 and $100,000 _____
    g. Greater than $100,000
11. Work (check one): Full time _____ Part-time _____ I do not work _____
12. In the last 12 months, have your or another family member:
    a. Left a job or taken a leave of absence because of your child’s health or condition? Yes _____ No _____

    b. Cut down on the hours you work because of your child's health or condition?
       Yes _____ No _____
    c. Avoided changing jobs because of concerns about maintaining health insurance for your child? Yes _____ No _____
    d. Avoided taking a promotion because of your child's health or condition?
       Yes _____ No _____

13. How many children live in your home? _____
14. Number of hours you spend each week (on average) providing direct care for your child? _______
15. Number of hours you spend each week (on average) coordinating your child's care (includes making phone calls, appointments, filling out paperwork for your child) _______
16. Number of hours per week you spend traveling (by public transportation or personal car) to meet your child's health care needs? _____
17. How much money did you pay for your child's medical, health, dental, and vision care in the last 12 months that your insurance did **not** cover?
    a. $0
    b. $1-$249
    c. $250-$499
    d. $500-$999
    e. $1000-$5000
    f. More than $5000

<u>Child Information</u>

1. Child's age in months ______ years _____
2. Child's gender: Male _____ Female _____
3. Child's race:
    a. Asian _____
    b. African American/Black _____
    c. American Indian or Alaska Native _____
    d. Caucasian _____
    e. Native Hawaiian or Other Pacific Islander _____
    f. Two or more races _____
4. Child's ethnicity: Is child Hispanic or Latino of any race? Yes _____ No _____
5. Health insurance: Public_____ Private _____ Both _____
6. Child's medical diagnoses: Select all that apply (Drop down menu will include arthritis, asthma, autism or autistic spectrum disorder, cerebral palsy, diabetes, seizure disorder, headaches/migraines, Down syndrome, Tourette syndrome, anxiety problem, depression, blood disorder (such as sickle cell disease or hemophilia), cystic fibrosis, genetic condition beside Down Syndrome, Behavior problem, developmental delay, Intellectual disability, speech/language problem, learning disability, other)

7. Child’s critical congenital heart disease diagnosis (Drop down menu to include all diagnoses of CCHD from CDC)

8. What medical needs does your child have? (check all that apply)
    a. Tracheostomy
    b. Ventilator
    c. Oxygen only
    d. BiPAP/CPAP
    e. Gastrostomy tube “G-tube”
    f. Nasogastric tube “NG tube”
    g. Central line for IV medications or access (PICC line, CVL or Port-a-Cath)
    h. Subcutaneous infusion
    i. Nebulizer
    j. Vest / Cough-assist device
    k. Other: _______________
9. Number of trained caregivers at home _____
10. Professional nursing hours your child receives per week _____
11. Rehabilitation therapies (Check all that apply)
    a. Physical Therapy (PT) How many times per week? ______
    b. Occupational Therapy (OT) How many times per week? ______
    c. Speech Therapy (ST) How many times per week? ______
    d. Other therapy _________ How many times per week? ______
12. How many visits in the last year has your child seen the pediatrician for well child care?
13. Specialists involved in your child’s care (check all that apply)
    a. Cardiologist How many visits in the last year? _____
    b. Pulmonologist How many visits in the last year? _____
    c. Neurologist How many visits in the last year? _____
    d. Orthopedist How many visits in the last year? _____
    e. Gastroenterologist How many visits in the last year? _____
    f. Nephrologist How many visits in the last year? _____
    g. Endocrinologist How many visits in the last year? _____
    h. Other How many visits in the last year? _____
14. Number of medications your child takes every day _____
15. How many times per day do you give your child any medication? _____