CHILDREN WITH MEDICAL COMPLEXITY IN THE CANADIAN MARITIMES: A MIXED METHODS STUDY

by

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Submitted in partial fulfilments of the requirements
for the degree of Doctor of Philosophy

at

Dalhousie University
Halifax, Nova Scotia
January 2023

DEDICATION

For my mother, Diane Breneol.
Thank you for your unconditional love and support.
It is because of your tireless advocacy and resiliency, that I am where I am today.

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ABSTRACT

Introduction: Developments in the medical field have improved the survival rates and management of children with complex chronic conditions. To date, policy and program design has primarily relied on findings from either quantitative or qualitative research. However, this may leave gaps in our knowledge about how available resources meet the needs of children with medical complexity. Family-reported unmet care needs, financial challenges, and increased stress levels signal the need for improvements to pediatric complex care. The objective of this research was to use health administrative and family-reported data to gain an in-depth understanding of the prevalence, health resource use, and needs of children with medical complexity and their families in the Canadian Maritimes.

Methods: This study used a three-phased explanatory sequential mixed methods design. In Phase one, a secondary data analysis of health administrative data was used to examine the prevalence and health resource use of children with medical complexity. In Phase two, five case studies were developed from multiple sources of family-reported data to generate a greater understanding of their experiences, health resource use, and needs. In phase three, a joint display table was used to triangulate quantitative and qualitative findings to result in a comprehensive understanding into the population of children with medical complexity.

Results: A total of 3058 children/youth were identified as having medical complexity, representing 0.88% of the Maritime pediatric population. Health data revealed a higher likelihood of outpatient, hospital, and emergency care encounters for children with medical complexity in comparison to children without medical complexity. Case study findings illustrated numerous resources interwoven across the health, social, community, and education sectors that families use to attend to their care needs. Families reported unmet care needs in areas such as care coordination, financial funding supports, and respite care. Triangulation of findings illuminated the extent of resource use that exists in the shadows of the health system.

Conclusions: There is a disconnect between current health systems and the needs of children with medical complexity and their families. By combining health administrative and family-reported data, this study unveiled critical information to inform potential strategic directions to improve pediatric complex care.

LIST OF ABBREVIATIONS AND SYMBOLS USED

CCC	Complex Chronic Conditions
CI	Confidence Interval
CIHI-DAD	Canadian Institute for Health Information Discharge Abstract Database
COM-B	Capability, Opportunity, Motivation – Behaviour Model
CMC	Children with Medical Complexity
EA	Educational Assistant
CPAP	Continuous Positive Airway Pressure
HDNS	Health Data Nova Scotia
ICD	International Classification of Diseases
IRR	Incidence Rate Ratio
IWK	Izaak Walton Killam
MED	MSI Physician Billings
NACRS	National Ambulatory Care Reporting System
NB	New Brunswick
NICU	Neonatal Intensive Care Unit
NP	Nurse Practitioner
NS	Nova Scotia
PEI	Prince Edward Island
PICU	Pediatric Intensive Care Unit
PMCA	Pediatric Medical Complexity Algorithm
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta Analyses
OR	Odds Ratio
VP Shunt	Ventriculoperitoneal Shunt

ACKNOWLEDGMENTS

This work could not have been possible without the tremendous amount of encouragement, inspiration, and collaboration from the incredible people in my life.

To Dr. Fong, the QEH Unit 4 and the IWK 6 Link/North Staff and Families – Thank you for showing me compassion and empathy during my most vulnerable moments. This past year marked 12 years in remission from pediatric Burkitt's Lymphoma. To all families with children with complex needs, your strength, resiliency, and advocacy are what inspired this work.

Thank you to my role models during my undergraduate degree at the University of Prince Edward Island. It was here that I was first introduced to the power of nursing research. Drs. Janet Bryanton and Vicki Foley, thank you for the endless amount of encouragement, letters of support, and beliefs in my potential.

Dr. Janet Curran – My supervisor. Thank you for challenging and supporting me through every success and hurdle. The opportunities you have provided me throughout the years have been instrumental in my development as a future nursing scientist.

Dr. Ruth Martin-Misener, Dr. Marilyn Macdonald, Dr. William Montelpare, Jocelyn Vine, and Dr. Sam Stewart – my committee members. This work could have been possible without each one of you. It is because of your exceptional knowledge, teaching, and encouragement, that this is all possible. Thank you deeply.

Dr. Heather Rose, Dr. Marion Brown, Dr. Andrea Bishop, NP Carolyn Mitchell, and Dr. Pantelis Andreou. The knowledge that I gained from working with each one of you has helped shape my perspectives as a researcher and clinician. Dr. Andreou – thank you for taking the time to show me every step of analyzing a complex dataset – this was such an incredible learning experience.

Sarah King, Shauna Best, Stacy Burgess, Angela Arra and Jackie Pidduck. Thank you all so much for your guidance in shaping this work. It has been a true pleasure to learn from incredible advocates like yourselves.

Drs. Andrea Bishop and Christine Cassidy – mentors and friends. Thank you for always being there. Thank you for being a safe place to ask questions (I know there have been many over the years) and for bringing me into your networks.

To all the STIPC crew – Helen, Julie, Lauren, Ben, Daniel, Matt, Holly, Sharon, Catie, Allyson, and Mari. I am so thankful to have worked with each one of you and to have developed lifelong friendships.

Brianna (Richardson) Hughes, Dr. Keshia Jefferies, and Dr. Martha Paynter – my PhD cohort. I am not sure that I can find the right words to describe how thankful I am for you all. You are an incredible group of women to have taken this journey with.

To my beautiful friends – Samantha, Robyn, Vuk, Claire, and Meghan. You are the epitome of friendship. Thank you for never letting me walk a journey alone. It's time for a Deep and Delicious cake and 6 forks.

To my partner – Dardan. Just before the world shut down, you came into my life, and now I cannot imagine it without you. Thank you for celebrating every success and listening with open ears at every challenge. I cannot wait to see what our next journey will bring.

To my big siblings - Charla, Tarra, and Brandon. Thank you for being such incredible role models and showing me that the sky is the limit.

To my second family – the McMurrers. You have been cheering me on at every graduation since kindergarten. Lennie – I know you will be with me while I cross that stage this one last time.

And to conclude – thank you to my mother, Diane, for being my everything. I simply would not be who I am today without your endless love and support. My success is your success. I love you to the moon and back.

Chapter 1: INTRODUCTION

Significant progress and advancements in the medical field have contributed to a notable change in the health of children over the past several decades (Wise, 2004). These ongoing innovations in medical treatments and technologies have contributed to the improved survival rates and long-term management of children with complex chronic health conditions (Burke & Alverson, 2010; Cohen et al., 2011). Current research suggests that approximately 15% of children in the United States have a chronic condition impacting their daily life (Strickland et al., 2015; Wise, 2004). This is an estimated 400% increase in children living with a chronic condition over the last 60 years (Perrin et al., 2014). This epidemiologic trend in the pediatric population has brought forth the importance of ensuring health policies, programs, and practices are being developed to improve the health service delivery and experiences for children with chronic conditions and their families.

Recognizing the need for a comprehensive conceptualization of this pediatric cohort, The Maternal and Child Health Bureau developed the now widely adopted definition used across research and clinical initiatives (McPherson et al., 1998). Denoted as 'children with special health care needs', these children and youth are characterized as those "who have or are at increased risk of chronic physical, developmental, behavioral, or emotional condition and require health care and related services of a type or amount beyond that required by children generally" (Mcpherson et al., 1998, p.138). Since the development of this definition, the body of literature exploring the needs of this population and their use of health resources has been steadily increasing. However, it is within this population that presents an emerging subgroup of children requiring the most

extensive health care resources due to the complexity of their condition and needs (Burns et al., 2010; Cohen et al., 2018; Cohen et al., 2011) .

While there has been a wide variety of terms used to describe this subpopulation of children and youth with exceptional health care needs (i.e. children with complex medical conditions), many researchers, clinicians, and policy makers have adopted the term 'children with medical complexity' while discussing this smaller pediatric cohort (Berry et al., 2014; Breneol et al., 2017; Cohen et al., 2018; Cohen et al., 2011). Children with medical complexity are primarily understood to be those with life-long complex chronic conditions requiring high levels of specialty care (Breneol et al., 2017; Brenner & DeLamater, 2014; Cohen et al., 2011; Feudtner, 2000). For illustrative purposes, a child with medical complexity might have a diagnosis of cerebral palsy requiring a gastrostomy tube and wheelchair and necessitates careful monitoring by a large team of professions to manage their care needs and prevent potential complications. While this terminology still varies across initiatives, 'children with medical complexity' is said to adopt 'person-first' language and will be used for such reasons (Cohen et al., 2011). However, developing a comprehensive definition for this cohort has proven challenging due to the difficultly in operationalizing 'medical complexity' at both a population and individualized level (Berry, Hall, Cohen, O'Neill, & Feudtner, 2015; Cohen et al., 2018, 2011). For example, what one family may perceive to be medically complex may differ from the perception of a health care provider (Berry et al., 2015). Further, contrary to the adult population where a select number chronic conditions represents the large majority of individuals with medical complexity, this pediatric cohort is made up of a wide variety of rare and distinctive diagnoses (Berry et al., 2015).

Children with medical complexity are a vulnerable and growing population within our health care system (Berry, Hall, et al., 2013; Burns et al., 2010; Cohen et al., 2018). Since the early 2000s, there has been a substantial increase in the published literature exploring pediatric complex care (Cohen et al., 2018). This emerging evidence has revealed a number of concerning findings regarding high rates of health care utilization and unmet care needs (Berry et al., 2014, 2018; Coller et al., 2017; Dewan & Cohen, 2013; O'Mahony et al., 2013). As many as 13 care providers may be involved in the care of one child at any given time, creating the potential for numerous communication and care coordination gaps (Cohen, Berry, et al., 2012). Additionally, the unique medical characteristics of these children place them at greater risk for adverse health outcomes, frequent hospital readmissions, fragmented coordination of care, and multiple unmet health care needs (Berry et al., 2014, 2018; Coller et al., 2017; Dewan & Cohen, 2013; Kuo et al., 2014; O'Mahony et al., 2013). In response to this growing body of evidence, many national associations have released guidelines to inform care management (Canadian Association of Pediatric Health Centres, 2016, 2018; Elias et al., 2012). However, while evidence is beginning to accumulate to define this population of children with medical complexity, understanding their health resource use and needs is also critical to inform the design of comprehensive programs and policies.

1.1. Health Care Needs

There has been an increasing emphasis on supporting the delivery of care for individuals with chronic conditions in their homes and home communities (Gordon et al., 2007; Kirk, 1999). As many as 89% of hospitalized children with complex chronic conditions are discharged home (Berry, Hall, et al., 2013). However, the challenges

associated with the provision of care for these children have important implications for both families and care providers in their home community (Berry, Hall, et al., 2013). Unfortunately, the health care needs of this population are infrequently met by the current health care system, leaving a substantial burden on families to provide expert medical care and facilitate care coordination activities for their child (Cohen et al., 2018; Dewan & Cohen, 2013; Kuo, Cohen, Agrawal, Berry, & Casey, 2011). Further, the challenges of caring for children with medical complexity have enduring effects on families, which include significant financial problems, increased stress levels and increased rates of depression (Charlton et al., 2017; Kuo et al., 2011; Kuo et al., 2014; Looman et al., 2009; Noyes, 2000; Toly et al., 2012). Despite these troubling reports, our understanding of the experiences and care needs of this vulnerable population outside of the hospital context is limited.

1.2. Health Resource Use

Children with medical complexity are often characterized based on their high rates of resource use across health sectors and settings (Cohen et al., 2011). Findings from observational studies suggest that health resource use and expenditure is disproportionate across the population, with individuals with complex medical needs representing a small proportion of the population, but a large percentage of health care resources (Cohen et al., 2012; Cohen & William, 2012; Wodchis, Austin, & Henry, 2016; Zook & Moore, 1980). Most of these studies, however, have focused on adult populations resulting in a limited understanding about high-users in the pediatric population (Cohen & William, 2012). Studies emerging from the United States (US) have shown an increased use of acute care services for children with complex chronic conditions (Burns

et al., 2010; Simon et al., 2010); yet little is known in regards to their health resource use after their initial diagnosis and discharge. Further studies conducted in the US, which examined specific illness presentations within the pediatric population, found similar results (Buescher et al., 2006; Neff et al., 2004; Neff et al., 2006). However, many of these studies have limitations in their prevalence estimates, as populations of interest were often too narrow (focusing on children with specific clinical conditions) or too broad (examining children with non-complex chronic conditions such as asthma) and examined only children enrolled in specific US health care plans (Buescher et al., 2006; Neff et al., 2004; Neff et al., 2006).

In the Canadian context, one study conducted in Ontario revealed that children with medical complexity represented an estimated 0.67% of their pediatric population, but accounted for approximately one-third of provincial pediatric health care spending (Cohen, Berry, et al., 2012). The first pan-Canadian report on the population of children with medical complexity released in 2020 found similar findings, suggesting an overall rate of 948 per 100,000 children and youth (Canadian Institute for Health Information, 2020). To our knowledge, these are the only two studies of its kind in Canada, leaving a paucity of research examining the prevalence and health resource use for this vulnerable population in additional and specific Canadian contexts. Further, all previously mentioned studies estimated health resource use using health administrative data and often lacked family-reported resource use. Given that not all health resources are captured within health administrative data (e.g., care coordination), a significant gap remains in our understanding of the resources required and accessed by children with medical complexity and their families.

1.3. Mixed Methodology

Gaining an understanding into the patterns of health resource use and care needs of children with medical complexity and their families can be a challenge. However, without this knowledge, health system policies and programs will lack in their ability to fully address the care needs of this population. Research is needed to explore this population through a variety of perspectives to fully capture and understand the current state of care within our health care structures. Leveraging the strengths of mixed methods research is one strategy to gain a more comprehensive understanding into the health resource use and needs of children with medical complexity and their families. There are a variety of benefits to applying a mixed methods approach when exploring complex phenomena such as the issue presented. By collecting, analyzing, and triangulating both quantitative and qualitative data, researchers can harness the strengths of each approach while simultaneously compensating for their respective limitations (Creswell & Plano Clark, 2018). Further, mixed methods research can be used when qualitative and quantitative data alone do not sufficiently address the identified problem (Creswell & Plano Clark, 2018). Despite these benefits, much of the current research in the field of pediatric complex care uses qualitative or quantitative data independently, creating a partial picture of this complex problem.

Health administrative data are one type of population level data that are being used in the study of children with medical complexity and their families (Berry et al., 2015). Health administrative data can be defined as the routinely collected data for administrative and billing purposes created with every health care encounter (Cadarette & Wong, 2015). While not primarily collected for research purposes, health researchers can

harness its potential to uncover critical information about the health system and those it serves. While this data source has a number of strengths, such as having access to large population samples across various timeframes, there are important limitations to the use of health data to consider. Specific to children with medical complexity, health data is limited in its ability to fully capture family-identified needs and a child's functional limitations (Berry et al., 2015). Further, families may utilize a range of health resources that are not captured by health administrative data alone (i.e. private respite care services, local community-run health programs, acupuncture, medical transportation resources, etc). This leads to a significant gap in our understanding of the health resource use and care needs of children with medical complexity and their families. Qualitative methods can address this gap by speaking directly to children and families about their lived experience. As such, combining health administrative data with richly descriptive qualitative reports from families is one strategy to fully explore their health resource use and care needs. Further, placing the emphasis and weight on the qualitative approach can amplify the voices of families at the forefront of pediatric complex care research (quan → QUAL). Employing a mixed methods approach would provide researchers, clinicians, and decision makers with a detailed and comprehensive understanding into the prevalence, clinical characteristics, health resource use, and health needs of children with medical complexity and their families. With this, decision makers can make evidence-informed recommendations to support the development of comprehensive family-oriented health programs and policies.

1.4. Research Problem, Purpose and Significance

To develop a child and family centered strategy to meet the needs of this population we must first understand their care needs and current health resource use. Previous studies have suggested that while this population may be small in numbers, they consume high rates of health resources (Buescher et al., 2006; Cohen, Berry, et al., 2012; Neff et al., 2004; Neff et al., 2006). Further, families report multiple unmet care needs and disturbed family functioning (Kirk et al., 2005; Kuo et al., 2011; Kuo et al., 2014). These findings highlight the urgent need for the development of strategies and interventions that are tailored to support the health of children with medical complexity and their families. However, we still lack a clear and comprehensive understanding into the patterns of health resource use and health needs of children with medical complexity and their families. This is particularly apparent in the Canadian context and even more so within the Canadian Maritimes. Developing a greater understanding of the population of children with medical complexity in the Canadian Maritimes, including their prevalence, clinical characteristics, health resource use, and health needs, is urgently needed as the first step in identifying and addressing the needs of this vulnerable population. Without this evidence, health system policies, programs, and practice changes will lack relevancy and sustainability.

The purpose of this research was to use both health administrative and family reported data to gain an in-depth understanding into the prevalence, health resource use, and care needs of children with medical complexity and their families in the Canadian Maritimes [(Prince Edward Island, (PEI), Nova Scotia (NS), and New Brunswick (NB)]. This research considered the following components: (1) prevalence and clinical

characteristics of children with medical complexity discharged from the Maritimes' pediatric tertiary care centre between 2004-2014; (2) rates of health resource use over a 5-year time frame; and (3) family-identified experiences, health resource use, and care needs. An explanatory sequential mixed methods design was chosen to address these components and achieve the following outlined research objectives:

1. Phase One (A) /Quantitative Phase: Describe the prevalence and clinical characteristics of children with medical complexity in the three Canadian Provinces (PEI, NS, and NB) using 2004 -2014 health administrative data.
2. Phase One (B) /Quantitative Phase: Describe the health resource use of children with medical complexity in Nova Scotia using five years of health administrative data.
3. Phase Two / Qualitative Phase: Describe the experiences, health resource use, and health needs of children with medical complexity and their families in each of the three Canadian Maritime Provinces (PEI, NS, and NB)
4. Phase Three / Data Triangulation: Explore in what ways do family-reported experiences, care needs, and use of health resources converge and diverge with the characteristics and health resource use identified within the health administrative data.

This research addressed the overarching question: How can health administrative and family-reported data be used to gain a comprehensive understanding into the patterns of health resource use and health needs of children with medical complexity and their families in the Canadian Maritimes?

To attend to the overarching research question outlined above, the following sub-questions were explored:

1) What is the prevalence and clinical characteristics of children with medical complexity in the Canadian Maritime Provinces?
2) What is the health resource use as described by health administrative data for children with medical complexity living in Nova Scotia?
3) What are the family-reported experiences, health resource use, and health needs of children with medical complexity and their families in the Canadian Maritime Provinces living in their home communities?
4) In what ways do the family-reported experiences, care needs, and health resource use converge and diverge with the characteristics and health resource use as reported by health administrative data among children with medical complexity and their families in the Canadian Maritime Provinces?

By pursuing these research objectives and questions, this study unveiled critical information about children with medical complexity and their families to health researchers, clinicians, policy makers, administrators, and families themselves. Mixed methods research has been underutilized in the current literature surrounding pediatric complex care. As such, this research is proposing a novel approach in the study of children with medical complexity and their health resource use, contributing to the advancement of this body of research. While the development of an intervention is beyond the scope of this research, it is anticipated that results can be used in future work to inform the design of health policy and programs in the Canadian Maritimes to improve

the health resource use, experiences, and outcomes for children with medical complexity and their families.

The following chapters consist of a literature review (Chapter 2) on the prevalence, health resource use, and care needs of children with medical complexity. This literature review is further supplemented by a scoping review exploring how health administrative data are informing health policy, practice, and research for children with medical complexity (*Manuscript 1*; Chapter 2). *Manuscript 2* (Chapter 3) provides the protocol guiding this mixed methods research. *Manuscript 3* (Chapter 4) addresses research questions #1 and #2 by detailing the prevalence rates of children with medical complexity, their clinical characteristics, and resource use in the Maritimes, as indicated by health administrative data. Chapter 5 provides further methodological detail into the specific health administrative datasets and details how phase 1 results informed recruitment and data collection in phase 2. *Manuscript 4* (Chapter 6) addresses research question #3, by building multiple case studies illustrating family-reported experiences, health resource use, and needs. *Manuscript 5* (Chapter 7) attends to research question #4, by triangulating phase 1 and phase 2 data to create a comprehensive understanding into the prevalence, resource use, and health needs of children with medical complexity and their families. Chapter 8, the final chapter, summarizes the implications to health education, practice, and policy and provides further direction for future research to improve pediatric complex care in the Maritimes.

Chapter 2: LITERATURE REVIEW

Children with medical complexity are a small, yet vulnerable population within the Canadian health care system. This pediatric cohort are frequently characterized based on the presence of complex chronic conditions affecting one or more organ systems requiring high levels of specialty care (Cohen, Berry, et al., 2012; Feudtner, 2000). Recent literature has suggested that while these children may be small in numbers, they consume a disproportionate amount of resources across health settings and report high rates of unmet health care needs (Berry, Hall, et al., 2013; Cohen, Berry, et al., 2012; Kuo et al., 2011, 2011; Neff et al., 2004; O'Mahony et al., 2013). There is a disconnect between the care needs of these children and their families and our current health care structures (Berry, Agrawal, et al., 2013; Cohen et al., 2018). Despite the increasing attention and focus on children with medical complexity and their families over the last two decades (Cohen et al., 2018), much is left to be understood in regards to their rates of health resource use and family-identified care needs. This is particularly relevant within the Canadian context. As such, there is a critical need to develop a more comprehensive understanding into the patterns of health resource use and care needs of children with medical complexity and their families to inform supportive health policies, programs, and services.

The following section will provide an overview of the current literature exploring health resource use and the health needs of children with medical complexity. This literature review will provide context and background to the research problem presented in this dissertation. This chapter will be divided into the following sections: 1) Conceptualization, prevalence, and characteristics of children with medical complexity;

2) Health resource use of children with medical complexity and their families; 3) Family-reported health resource needs; and 4) Use of conceptual and theoretical frameworks in the study of pediatric complex care. Lastly, this chapter concludes with a scoping review of the literature exploring how health administrative data are informing practice, policy, and research recommendations for children with medical complexity

To retrieve all potentially relevant literature, a number of strategies were employed. This included searching relevant electronic databases (CINAHL, PudMed, and EMBASE), reference lists of seminal articles in the field of pediatric complex care, and grey literature sources (Google, Google Scholar, pediatric health associations). Using a combination of Boolean operators 'AND' and 'OR' to separate key words and MeSH headings, a comprehensive search strategy was developed and adapted for implementation in each of the electronic databases (See Appendix A). Published literature in the field of pediatric complex care has grown dramatically over the last ten years (Cohen et al., 2018). As such, a date range was set between 1998-2021 to capture all potentially relevant articles. Further limiters included English language only. As there remains a dearth of research exploring children with medical complexity and their families in Canada, all relevant articles were explored regardless of their country of origin. While this literature review will remain focused on the population of children with medical complexity, relevant examples from the broader pediatric and adult literature have been included to augment certain areas where gaps exist in the field.

2.1. THE POPULATION OF CHILDREN WITH MEDICAL COMPLEXITY

Conceptualizing and identifying children with medical complexity both at an individual and population level has been a frequent discussion within the pediatric

complex care literature. The following sections will explore the conceptualization of children with medical complexity and their families.

A plethora of changes in environmental, social, and medical factors have contributed to the increased number of children living with a chronic condition over the past several decades (Perrin et al., 2014). With this increasing proportion of children with unique health concerns, it is essential that health practice, policy, and programs are designed to effectively meet their health care needs. In 1998, the United States' Maternal and Child Health Bureau recognized the crucial need to define this pediatric cohort as the first step in the development of a responsive health care system for these children and their families (McPherson et al., 1998). Adopting the terminology 'children with special health care needs', the Maternal and Child Health Bureau formed a working group composed of families, health care providers, and decision makers to create and recommend a comprehensive definition. Drawing upon learnings from a review of the literature and government policies, the working group chose to adopt a 'service-based approach' to characterize this pediatric population. They argued that adopting this approach would avoid a problematic and restrictive characterization that could come with condition-specific approaches. As such, their final recommendation was as follows: "Children with special health care needs are those who have or are at increased risk for chronic physical, developmental, behavioural, or emotional condition and who also require health and related services of a type or amount beyond that required by children generally" (McPherson et al., 1998, p. 138). This inclusive definition has now been widely adopted across research and policy initiatives focusing on optimizing health care for children with special health care needs and their families (Bethell et al., 2008;

Strickland et al., 2011, 2015; van Dyck et al., 2002). Within this population, however, presents a smaller cohort of children that many researchers, policy makers, and clinicians have identified as needing greater support and resources to manage their care needs (Bramlett et al., 2009; Burns et al., 2010; Cohen et al., 2011). These children have been largely recognized as those with the most complex chronic health conditions requiring high levels of specialty care to manage their medical, social, and care coordination needs.

Contrary to children with special health care needs, characterizing and defining this subgroup of children with the most extensive health care needs has been challenging. Within both the research and policy domains, this specific pediatric population has been referred to by a variety of terminologies, which has included children with complex chronic conditions (Feudtner, 2000), medically complex and fragile children and youth with special health care needs (Gordon et al., 2007), and children with health complexity (Cohen et al., 2018). Increasingly, however, is the adoption of the term 'children with medical complexity'. This terminology was created and proposed based on its 'person-first' approach, placing the child at the forefront and their health condition(s) secondary (Cohen et al., 2011).

Children with medical complexity encompass a population with a wide variation of pediatric conditions that can range from severe neuromuscular impairments, malignancies, complex mental health conditions and multisystem diseases (Cohen et al., 2018, 2011). Creating a clear definition and conceptualization of this unique pediatric population has been identified as a top research priority to advance the field of pediatric complex care (Agrawal, 2015). However, to date there has been difficulty in adopting a universal definition for this subgroup of children as their specific clinical, social, and

familial characteristics may vary across individuals. Characterizing this population can be highly subjective, with personal biases influencing our conceptualization (Berry et al., 2015). For example, what one family or care provider may perceive to be 'complex' can differ based on their previous experiences (Berry et al., 2015). Despite these challenges, there are two main definitions used in the literature to describe this unique population. The first definition was proposed by Feudtner, Christakis, & Connell (2000) to extend their research focusing on pediatric palliative care. Feudtner et al. (2000) referred to this pediatric cohort as children with complex chronic conditions and proposed the following definition: "Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center" (p.206). While this definition was originally created for research exploring pediatric palliative care, it has remained a commonly used definition to describe this population within a variety of research contexts (Amin et al., 2018; Berry et al., 2011; Brittan et al., 2015; Guertin, Côté-Brisson, Major, & Brisson, 2009). This definition, however, lacks reference to any family-identified characteristics and needs that may play an influential role in defining 'complexity'.

The second definition, proposed by Cohen et al. (2011), provides conceptual clarity and facilitates the amalgamation of learnings across the health research, program, and policy sectors. Using recommendations produced by a systematic review of pediatric chronic conditions, Cohen et al. (2011) proposed a definitional framework composed of four main characteristics unique to the population of children with medical complexity:

(1) the presence of one or multiple suspected or diagnosed complex chronic condition(s); (2) functional limitations impacting daily life that may require the use of medical technology; (3) high levels of family-identified needs; and (4) high rates of health resource use across multiple services and sectors. This definitional framework has helped to mitigate some of the challenges of defining children with medical complexity at an individual level by outlining shared characteristics irrespective of specific diagnoses. This definition also recognizes family-identified needs as a significant aspect to 'medical complexity' (Cohen et al., 2018; Cohen et al., 2011). Furthermore, this conceptualization remains broad and inclusive of complex mental health conditions as primary or co-morbid condition (e.g. schizophrenia, specific delays in development). In 2018, the Canadian Association of Pediatric Health Centres released national guidelines recommending the adoption of this definitional framework to promote consistency across Canadian initiatives (Canadian Association of Pediatric Health Centres, 2018).

While exploring specific conditions is beyond the scope of this work, it is important to note a lack of visibility of children and youth with gender dysphoria who are seeking gender-affirming care/interventions within this body of literature. To date, there has been a paucity of work with this important pediatric population and how identities may intersect with medical complexity.

In summary, to begin to create a health system responsive to the care needs of children with medical complexity and their families, a uniform and consistent conceptualization of this cohort is needed.

2.1.1. Prevalence And Characteristics

Determining the prevalence of children with medical complexity has been hindered by the difficulty in operationalizing the concept of 'medical complexity' at a population level (Berry et al., 2015). This is primarily due to the myriad of conditions that make up the population of children with medical complexity. That being said, researchers have utilized multiple methods to determine the number of children with medical complexity using both survey and health administrative data. In Ontario, Canada, provincial health data revealed that children with medical complexity represent 0.67% of the pediatric population (Cohen, Berry, et al., 2012). Within this small pediatric cohort, 30% of children were classified as having a neurological impairment, 65% as having a condition involving a single body system, and less than 7% as having a condition affecting multiple systems (Cohen, Berry, et al., 2012). Further, approximately 12% of children used some form of medical technology to manage their health condition (i.e. gastronomy tube) (Cohen, Berry, et al., 2012). The majority of these children were less than one year of age and only a small proportion were between the ages of 14-16 years old (Cohen, Berry, et al., 2012). These findings are aligned with prevalence estimates stemming from United States that suggest children with the most complex chronic illnesses represent less than 1% of their pediatric population (Neff et al., 2004). Interestingly, Japan observed an increase in the number of children with medical complexity receiving home care between the ages of 7-17 over a 5 year time frame (Yamaoka et al., 2018). This signals the need for more longitudinal research in Canada to assess demographic characteristics and prevalence trends over time.

In addition to using routinely collected health data, national surveys have also provided prevalence estimates and demographic characteristics of children with medical complexity. The National Survey of Children with Special Health Care Needs is a telephone-based survey conducted every 4 years across the United States (van Dyck et al., 2002). Using a random sampling technique, this survey aims to locate households with children under the age of 18 years old (van Dyck et al., 2002). If deemed eligible, general demographic characteristics are collected and parents/guardians are led through a screening process to determine if their child(ren) has/have special health care needs (van Dyck et al., 2002). Researchers have harnessed the data to explore the characteristics of children with medical complexity by stratifying and examining families who responded that their child was in need of high levels of medical care (Kuo et al., 2011). Using this method, researchers have estimated that children with the most complex health needs make up approximately 3% of the population of children with special health care needs and 0.4% of the total US pediatric population (Aboneh & Chui, 2017; Kuo et al., 2011). Children with the most complex health care needs also tend to be younger and have a lower familial socioeconomic status (Kuo et al., 2011, 2014). Ethnic and racial demographics within this population have been largely understudied. Analysis of national survey data in the US suggests that children with medical complexity are primarily Non-Hispanic White (72%) (Aboneh & Chui, 2017) and have a significantly smaller proportion of children who identified as Non-Hispanic Black (Kuo et al., 2014). However, more research is needed in this area to gain a greater understanding into the detailed demographics of children with medical complexity.

2.2. PATTERNS OF HEALTH RESOURCE USE

Children with medical complexity have been shown to use a disproportionate amount of health care resources (Agrawal et al., 2016; Chan, Rodean, et al., 2016; Cohen, Berry, et al., 2012; Coquillette, Cox, Cheek, & Webster, 2015; Edwards et al., 2012; Kuo et al., 2015; Neff et al., 2004; Walter, Ellis, & Yuan, 2018). While they account for less than 1% of the overall pediatric population, evidence has shown that they consume approximately one-third of all pediatric health care costs (Cohen, Berry, et al., 2012; Kuo et al., 2011; Neff et al., 2004). In addition to accounting for a large proportion of inpatient hospital admissions (Berry et al., 2014; Berry, Toomey, et al., 2013; Gold et al., 2016; Simon et al., 2010), children with medical complexity experience lengthy hospitalizations (Gold et al., 2016) and frequent outpatient visits (Cohen, Berry, et al., 2012; Yamaoka et al., 2018). The following section will describe literature exploring the health resource use of children with medical complexity.

2.2.1. Hospital-Related Services

Recent evidence has suggested that children with medical complexity are using an increasing amount of inpatient services (Berry, Hall, et al., 2013; Burns et al., 2010; Simon et al., 2010). Using health data from 28 US hospitals over a 5 year period, Berry, Hall, et al. (2013) sought to explore trends in pediatric inpatient use. This observational study determined that when compared to both children without chronic conditions and those with non-complex conditions, children with a significant chronic condition impacting 2 or more organ systems are the largest growing population within pediatric health centres. A median growth of 35.6% was observed between years 2004 – 2009 (Berry, Hall, et al., 2013). Further stratified based on health resource use, children with a

significant chronic condition affecting 2 or more body systems or a complex or progressive condition represented an estimated 20% of all pediatric patients, 50% of hospital days, and 50% of hospital costs (Berry et al., 2013). It is also important to note that this study revealed an increasing number of unique children with medical complexity, suggesting a growth in the overall population (Berry et al., 2013). Simon et al. (2010) observed similar findings, with their administrative data suggesting that US children with complex chronic conditions account for "10% of admissions, 25% of hospital days, 40% of hospital charges, 75% to 92% of technology-assistance procedures, and 43% of inpatient deaths" (Simone et al., 2010, p. 651). Further, Berry et al.'s (2017) retrospective analysis of the Kid's Inpatient Database in the US suggests that children with multiple chronic conditions consume over half of pediatric hospital care costs and account for 1 out of every 4 pediatric hospitalizations (Berry et al., 2017).

Examining admissions to pediatric intensive care units (PICUs), Edwards et al. (2012) uncovered that over 50% of admitted children had a diagnosed complex chronic condition. Children with complex chronic conditions also experience significantly high rates of mortality and long length of stays within the PICU context (Edwards et al., 2012). Additionally, recent evidence suggests that admission rates of children with medical complexity to both PICUs and intermediate care units have been increasing over time (Silverman & DeCourcey, 2016).

Only a handful of studies have examined emergency department use in this pediatric cohort. Studies that have examined this phenomenon have noted that amongst all children with chronic conditions, children with medical complexity experience higher rates of pediatric emergency department visits (Hudson et al., 2014; D. Kuo et al., 2015;

O'Mahony et al., 2013). Children with medical complexity have also been shown to experience higher rates of repeat visits, direct hospital admissions, visits related to adverse drug events, and longer length of stays within the department (Feinstein et al., 2014; O'Mahony et al., 2013). While these rates are higher in comparison to other pediatric populations (O'Mahony et al., 2013), findings from one Canadian study suggests emergency department visits are not a significant contributor to the health care costs accrued by children with medical complexity (Cohen et al., 2012). More research is needed to explore how emergency department resources are being used by this vulnerable population and their families.

2.2.2. Outpatient Care

It is now widely accepted that the ideal location of care for children with medical complexity is in their homes and home communities (American Academy of Pediatrics, 2002; Barone et al., 2020; Gordon et al., 2007). Despite data suggesting that 89% of hospitalized children with complex chronic conditions are discharged home (Berry, Hall, et al., 2013), there has been limited exploration of their health resource use outside of the hospital setting. Examining provincial health data over a 2-year period, one study in Ontario, Canada, showed that children with medical complexity were receiving outpatient care from a median of 13 physicians from 6 separate medical specialties (Cohen, Berry, et al., 2012). This study further revealed that 36% of children with medical complexity had used home care services (Cohen, Berry, et al., 2012). This rate of service utilization was much higher (64%-81%) amongst children who were dependent on technology, with nursing, therapy, and case management visits being the most frequently used home service (Cohen, Berry, et al., 2012). These findings are in alignment with other health

care contexts. Studies emerging from the US and Japan revealed that children with medical complexity had approximately 20 outpatient visits over a one year time frame (Kuo et al., 2015; Yamaoka et al., 2018). Yamaoka et al. (2018) also noted that the number of children receiving care at home had increased from 1994 - 2014.

2.2.3. Health Resource Costs

In addition to describing rates and percentages, researchers examining health resource use have displayed their findings in terms of health care costs (Cohen, Berry, et al., 2012; Kuo et al., 2015; Neff et al., 2004). Current evidence suggests that the hospital-related expenses account for the majority of health care expenditures for children with medical complexity (Cohen, Berry, et al., 2012; Kuo et al., 2015; Neff et al., 2004). For example, Neff et al. (2004) used Washington state health insurance data to reveal the staggering pattern of health care expenditures amongst the entire pediatric population. Over a one-year time period, children classified as 'healthy' used on average approximately $500 health care dollars (Neff et al., 2004). However, children living with the most complex chronic condition used on average over $75,000/year (Neff et al., 2004). They also noted inpatient costs accounted for the bulk (67%) of overall health care charges, with only 27% of costs going towards physician billings (excluding inpatient related-charges) and outpatient services (Neff et al., 2004). While the data used in this study are now 20 years old and health care programming and associated costs may have changed, these findings remain consistent with more current literature (Cohen et al. 2012).

In the Canadian context, Cohen and Berry et al. (2012) demonstrated that on average children with medical complexity used $53,000 in pediatric health care resources

over a 2-year period. Examining the population over the 2-year time frame, children with medical complexity expended approximately $840,000, representing one-third of the entire provincial pediatric health care spending. Interestingly, approximately 80% of these costs were accrued during hospital admissions, with only 16% of costs going towards home care and out-patient services (Cohen, Berry, et al., 2012).

Much of the current literature surrounding the costs of health resource use of children with medical complexity has focused on that of the health care system. It is, however, critical that we acknowledge the financial costs many of these children and families experience related to their ongoing care at home. Regrettably, there is a significant dearth of research in this area. One study by Walter et al. (2018) used private health insurances claims to explore the out-of-pock expenditures for families of children with medical complexity. The findings stemming from this work showed that in comparison to families of children without chronic conditions, families of children with complex chronic illnesses reported twice as many out-of-pocket expenses (Walter et al., 2018). While this study begins to shed light on the financial burden many families of children with complex chronic conditions experience, it only provides a partial picture due to its reliance on data from medical claims and the lack of family-reported outcomes.

2.3. FAMILY-REPORTED EXPERIENCES

Children with medical complexity are often discharged with comprehensive care plans and/or medical technologies requiring extensive oversight and management (Barton et al., 2021; Whiting, 2014). Once at home, much of this skilled medical care and care coordination becomes the primary responsibility of parents/primary caregivers (Barone et al., 2020; Elias et al., 2012; Kirk et al., 2005; Ray, 2002). While the previously discussed

studies provide critical information regarding health resource use, most of these studies were unable to capture the social, financial, and care management burdens many of these families face while overseeing their child's care. This leaves a critical gap in knowledge by limiting our ability to fully assess and understand their health needs and use of health resources. The following section will summarize current literature surrounding family-reported experiences and health needs.

2.3.1. Care Coordination

Despite the increasing attention to attend to the needs of this vulnerable population, approximately half of children and their families report having unmet health needs (Aboneh & Chui, 2017; Dyck et al., 2004; Kuo et al., 2011; Kuo et al., 2014; van Dyck et al., 2002). For example, in addition to using The National Survey of Children with Special Health Care Needs to identify prevalence and characteristics of children with medical complexity, this survey has been used to illuminate family-reported health care needs. These data have revealed that in comparison to families of children without a complex chronic health condition, families of children with medical complexity report significantly higher rates of unmet care needs (Aboneh & Chui, 2017; Kuo et al., 2014). These challenges are accompanied by reports of fragmented coordination of care and inadequate resources to successfully thrive at home (Aboneh & Chui, 2017; Allshouse et al., 2018; Ghose, 2003; Golden & Nageswaran, 2012; Kuo et al., 2014). Despite families identifying care coordination as a critical component of care for their child (Leyenaar, O'Brien, et al., 2017), this need continues to go unmet (Aboneh & Chui, 2017; Charlton et al., 2017). In a qualitative study examining families' perspectives on care coordination, Golden & Nageswaran (2012) revealed that families often assume the crucial

responsibility of care coordination for their child as a result of poor communication between their care team members. In turn, this not only created the potential for delays in care, but also generated significant stress for families. Given the complexity of their needs across multiple services and sectors, many families described managing their child's care as a 'full time job' (Golden & Nageswaran, 2012). While some health care providers such as pediatricians and nurses helped with aspects of care coordination, families reported the need for more support to help navigate across sectors, access health resources, and advocate for their child's needs (Golden & Nageswaran, 2012). Especially troubling were families reporting that they had moved closer to a health centre and changed employment to attain greater flexibility in their schedules to manage their child's care (Golden & Nageswaran, 2012).

To address this challenge in care coordination, Adams et al. (2013) aimed to explore parent perceptions and usefulness of comprehensive care plans. Care plans were defined as a "written document that outlines the major medical issues and care needs for a specific child and is created by the health care provider in collaboration with the family" (Adams et al., 2013, p. 2). Through conducting semi-structured interviews with families and focus groups with health care providers, Adams et al. (2013) identified care plans as a beneficial tool in the care of this pediatric population. Participants expressed that frequently updated care plans can help facilitate effective care coordination across multiple settings and services. Key components to the care plan would include, but were not limited to, medication lists, care team contacts, emergency care protocols, brief medical history, diagnoses, the child's photograph, and information about their preferences (Adams et al., 2013). In a subsequent study, Adams et al. (2017) explored the

meaning of care maps for children with medical complexity and their families and for their health care providers. Similar to their 2013 study, families identified care maps as a helpful tool to facilitate the communication of key components about their child's health care team, services, and needs. Furthermore, health care providers perceived care maps as being a beneficial tool in gaining an insight into the family experience and help facilitate effective partnerships.

2.3.2. Financial Challenges

Using family-reported survey data, Kuo et al. (2011) revealed that parents/guardians of children with the most complex care needs spend up to 20 hours/week delivering direct medical care for their child and an additional 2 hours/week on managing care coordination activities. This, coupled with the many health-related expenses accrued by families (i.e. prescription medication, assistive equipment), it is unsurprising that many families of children with medical complexity report experiencing significant financial challenges (Kuo et al., 2011; Looman et al., 2009). Approximately half of families of children with complex medical needs have reported financial burdens, leaving their employment and/or reducing their work hours (Kuo et al., 2011). Further, caregivers who reported poor communication with health care providers were at higher odds of having financial problems when compared to families reporting strong communication (Looman et al., 2009). In addition to these employment constraints, families report spending over $1000 yearly in health-related expenditures and needing supplementary income to cover these health-related expenses (Kuo et al., 2011). Care providers need to advocate for families and help identify potential resources to cover health-related expenses (Looman et al., 2009). However, despite these findings, little

research has been conducted surrounding what resources families are accessing or wish to access to mitigate financial hardship.

2.3.3. Family Health

Adequate support and resources need to be in place to optimize family health and functioning (Barnert et al., 2018). In a 10-year longitudinal Canadian study, families of children with chronic conditions reported more depressive symptoms and poorer overall health in comparison to families of children without health concerns (Brehaut et al., 2011). These findings are not in isolation, with additional longitudinal and qualitative studies revealing that families of children with medical complexity report high levels of stress, depression, feelings of isolation, and disturbed family functioning (Carnevale et al., 2006; Curran et al., 2018; Kirk et al., 2005; Toly et al., 2012). A commentary written by a mother of a child with medical complexity described the time required to manage her child's medical, educational, and social needs made it difficult to seek support from other families (Ghose, 2003).

In Winnipeg, Canada, Woodgate, Edwards, Ripat, Borton & Rempel (2015) employed ethnographic and photo voice methods to explore the experience parenting a child with complex medical needs. A total of 40 families participated in the study. Results from this research provide insight into the "intense parenting" that occurs when raising a child with complex needs. Families expressed that they had little time to focus on any tasks outside of caring for their child and their complex care needs. Parents also felt as if they had assumed the role of their child's health provider, case manager, and advocate. As a result, families reported experiencing negative effects on their own health. This was related to worrying about their child's current and future health, sleepless

nights, and the physical tasks required while caring for their child (Woodgate et al, 2015). These findings highlight the need to support family health as a whole. Researchers, health professionals and policy makers need to gain a greater understanding of what resources need to be in place to better support these children and their families.

2.3.4. Health Resource Use

In alignment with studies using health administrative data, families have reported bringing their child to approximately 11-15 outpatient physician visits over a one-year time frame (Cohen, Berry, et al., 2012; Kuo et al., 2011). However, a number of health resources are not captured in routinely collected health administrative data that are critical to supporting children and their families at home. This contributes to a gap in our understanding of relevant and important health resources for families. For example, respite care has been identified as one of the key components in the care of children with medical complexity and their families (Elias et al., 2012). However, families experience challenges in locating and accessing respite services in their community (Doig et al., 2009). Restrictive eligibility criteria, lack of funding, difficulty in accessing information, and long-wait lists are just some of the barriers being reported by children and families while trying to attain respite care services (Cramer & Carlin, 2008; Doig et al., 2009). This essential supportive resource is an unmet need experienced by many children and their families (Eaton, 2008). With one-third of families reporting challenges in attaining access to nonmedical services such as rehabilitation, child care, and early intervention services, greater attention needs to be paid to the resources families require to successfully care for their child in their home community (Kuo et al., 2011).

In the Canadian Maritime context, Charlton et al. (2017) sought to examine the health resources available to children with complex chronic health conditions in New Brunswick. In tandem with an environmental scan, semi-structured interviews were conducted with families and key stakeholders to identify barriers related to accessing health services. A multitude of services spanning across various sectors and settings were identified as being accessed by families in their home province. This included, but was not limited to hospital clinics, home health services, dental care, community centres, and financial support programs. Families also reported having to travel to surrounding provinces, primarily to the closest pediatric tertiary care facility in Halifax, Nova Scotia. Families identified various gaps in health resources related to mental health and allied health services such as physiotherapy, speech-language, and rehabilitation resources. Furthermore, families reported the need for more care coordination, less restrictive program requirements, increased provider training, and enhanced financial support to help reduce the challenges and delays in accessing health resources. As Charlton et al., (2017) states, "despite the range of services available for children with complex health conditions, this study suggests that the level of services is not keeping pace with the needs of children with complex health conditions and their families" (p.146).

2.4. CONCEPTUAL AND THEORETICAL FRAMEWORKS

With the rising attention directed towards pediatric complex care, ensuring a strong conceptual foundation across initiatives is essential. Conceptual frameworks provide a lens to guide an individual's perceptions and interpretations of a phenomenon or phenomena under inquiry (the term 'model' and 'framework' will be used interchangeably through this work) (Ivey, 2015; Polit & Beck, 2012). Conceptual

frameworks also play an integral role in bridging the gap between research, policy, practice and education by providing a common language to understanding and perceiving a phenomenon.

There is ambiguity in the literature surrounding the distinction between a conceptual and theoretical framework. While many researchers and authors use these terms interchangeably, Polit and Beck (2012) stipulate that theories and theoretical frameworks offer a detailed explanation into the perceived and predictive relationships between concepts, whereas conceptual models do not. However, authors have questioned whether a clear distinction between the two terminologies is paramount; requesting that the emphasis be placed on how the framework is being used and applied in research and clinical practice (Green, 2014). The following section will explore the use of conceptual and theoretical frameworks in the study and care delivery of children with medical complexity and their families.

2.4.1. Care Delivery Models

The disproportionately high rates of health care resource use by children with medical complexity have signaled the crucial need to improve the delivery of patient and family centered health care. Furthermore, the multitude of care team members involved in the care of these children and their families at any one time has led many families, researchers, and decision makers to emphasize the need to improve interdisciplinary care coordination and communication (Breneol et al., 2017; Canadian Association of Pediatric Health Centres, 2018; Cohen, Berry, et al., 2012; Leyenaar, Rizzo, et al., 2017). As such, there has been a spiked interest in creating and implementing effective models of service delivery for these children and their families (Pordes et al., 2018). For example, the

American Academy of Pediatrics released a policy statement reaffirming their 'medical home' as the optimal model of care for all children and families (American Academy of Pediatrics, 2002, American Academy of Pediatrics, 2022). This model has its foundations in primary care and proposes that all care should be "accessible, continuous, comprehensive, family centered, coordinated, compassionate, and culturally effective" (p.5). While some have speculated that this may be the ideal model of care for children with medical complexity and their families, our current health care structures are not designed to fulfil these principles (Berry, Agrawal, et al., 2013, p. 5). First, while primary care should be the 'hub' of care for these children and families, community health care providers outside of specialized pediatric facilities may be less familiar with caring for the vast array of rare pediatric diagnoses and needs (Berry et al., 2014). As such, some primary care providers may feel ill-equipped to provide the most optimal level of care for these children and their families (Berry, Agrawal, et al., 2013). Second, coordinating the complex care for these children and their families may be difficult to achieve within of a community practice context (Berry, Agrawal, et al., 2013).

In 2018, Children's Health Canada released national guidelines to inform the care management of children with medical complexity (Canadian Association of Pediatric Health Centres, 2018). While they acknowledge the medical home model proposed within the US health system, it remains unclear how this model could be adopted in the Canadian health care context. As such, the guidelines aim to provide evidence-informed recommendations to improve the health of Canadian children with medical complexity and their families. Additionally, they emphasize the critical need for more Canadian-based research exploring this population. These guidelines put forth six

recommendations: (1) identify children with medical complexity and their families to ensure appropriate services are provided; (2) build health system capacity to provide comprehensive and family-centered care in the community; (3) ensure each child and family have a key point of contact to facilitate care coordination and communication; (4) create and update comprehensive co-developed care plans; (5) empower families; and (6) develop strategies to ensure smooth transitions in care. As these recommendations begin to be operationalized and implemented into specific provincial contexts, implementation and evaluation research is critical to achieve the most optimal model of care delivery (Canadian Association of Pediatric Health Centres, 2018).

2.4.2. Research and Policy Models

Conceptual and theoretical frameworks are not commonly used in the study of children with medical complexity and their families. Many seminal research initiatives written by leading experts in the field of pediatric complex care lack reference to the use of conceptual frameworks guiding their research process (Berry et al., 2017; Cohen et al., 2011; Feudtner et al., 2000; Kuo et al., 2014; Simon et al., 2010). Authors that do reference the use of a conceptual framework supporting their research often report little detail about the framework itself, how it was developed, or how it guided and/or informed the research process (Gidengil et al., 2014; Nageswaran & Golden, 2016; Nageswaran, Golden, Gower, & King, 2018). This highlights the need for researchers to be more explicit when describing their use of conceptual and theoretical frameworks.

Within the adult literature, there are a number of frameworks being readily used in research related to individuals with medical complexity. A systematic review examined conceptual models related to multiple chronic conditions and medical

complexity in adults (Zullig et al., 2016). This resulted in the synthesis of 7 conceptual models and creation of the Cycle of Complexity Model, which highlights both social and medical components contributing to complexity in the adult population (Zullig et al., 2016). Other models used in the adult chronic care management literature include the Chronic Care Model, where emphasis is placed on key elements within the health care system to support quality care (Barr et al., 2003; Wagner et al., 2001). Currently, there are no widely adopted conceptual or theoretical frameworks being used in the study of children with medical complexity and their families, leaving this body of literature largely a-theoretical (Barnert et al., 2018). While the use of 'traditional' conceptual and theoretical frameworks may not be present in the current literature surrounding pediatric complex care, there are few frameworks present within the literature that are guiding the conceptualization of this cohort and their health.

To date, Cohen et al.'s (2011) definitional framework for children with medical complexity has been cited over 900 times in published literature. While there is no clear understanding regarding what comprises a definitional framework, its potential as a rudimentary conceptual framework can be argued. Conceptual frameworks provide a greater understanding of a concept under study; thus they conceivably provide a comprehensive definition. Unsurprisingly, Cohen et al.'s (2011) definitional framework has been primarily used to describe and define the population of children with medical complexity (Arthur et al., 2018; Barnert et al., 2017, 2018; Berry et al., 2011, 2014; Bogetz et al., 2015; Cady et al., 2015; Cohen, Berry, et al., 2012; Coller et al., 2016; Leyenaar, O'Brien, et al., 2017; McKissick et al., 2017; Nelson et al., 2016; Thomson et al., 2016). This framework has been used in a variety of different research contexts,

including guiding the eligibility for complex care programs (Cohen, Lacombe-Duncan, et al., 2012) and exploring the concept of 'complexity' itself (M. Brenner et al., 2018; Nelson et al., 2018). Other articles have attempted to operationalize this conceptual definition to identify children with medical complexity at a population level (i.e. nationally representative surveys, health administrative data) (Cohen, Berry, et al., 2012; Coller et al., 2016; Simon et al., 2018; Srivastava et al., 2016). However, there has been limited exploration into how this framework could be leveraged to inform the research process (i.e. formulation of the research question, data collection, data interpretation). As definitional frameworks do not have any guidelines as to their development, they may conceivably have some limitations in their use as a conceptual framework. More research applying Cohen et al.'s (2011) definitional framework throughout the research process is needed to fully understand its strengths and limitation as a comprehensive conceptual framework.

Cohen et al.'s (2011) definitional framework can also be found within the policy domain. For example, Children's Healthcare Canada's guidelines to inform the care management of children with medical complexity used Cohen et al.'s (2011) definitional framework to define and characterize their target population. They outlined a pre-determined criteria that facilitates the operationalization of Cohen et al.'s (2011) conceptual definition in practice to consistently identify this population across Canada. The adoption of this recommendation will help advance efforts to develop and standardize an optimal model of care delivery for children with medical complexity and their families (Canadian Association of Pediatric Health Centres, 2018). Further, similar to the research domain, other policies and reports have used Cohen et al.'s (2011)

definitional framework to describe their target population, such the Irish Health Service Executives Review of Policy and Practice in the Provision of Home Care to Children with Medical Conditions and American Academy of Pediatric Clinical Reports for discharging children with medical complexity (Elias et al., 2012; Health Service Executives, 2014; Kuo et al., 2016).

Work from Barnert et al. (2018) has demonstrated promising advancements into the conceptualization of health for children with medical complexity and their families through the inclusion of patient perspectives. Using an integrative approach, Barnert et al. (2018) synthesized learnings from a systematic review, observations, and interviews with families, clinicians, advocates, researchers, and policy makers from North America, to create a conceptual framework outlining 10 domains of health for children with medical complexity. These domains included: (i) basic needs; (ii) inclusive education; (iii) child social integration; (iv) child health-related quality of life; (v) long term child and self-sufficiency; (vi) family social integration; (vii) community system supports; (viii) health care system supports; (ix) high quality patient-centered medical home; and (x) family-centered care (See Table 2-1) (Barnert et al., 2018). In addition to a detailed conceptualization, each domain had at least 1 proposed measurable health outcome. Unfortunately, many of the domains outlined in this conceptual framework are not being addressed in current research targeting this unique cohort (Barnert et al., 2018); highlighting the crucial need for advancement in this area. It is also important to note that children with medical complexity were defined and conceptualized using Cohen et al.'s (2011) definitional framework (Barnert et al., 2018). Barnert et al.'s (2018) work offers another vital step in the development of a comprehensive framework to describe health

for children with medical complexity and inform practice and policy reform. While this framework may be limited by its primarily North American perspective, it is anticipated that this conceptual framework will continue to grow with its use and application in a variety of care settings.

Table 2-1: 10 Domains of Health for Children with Medical Complexity

Domain	Description of Domain
Basic Needs	"Children with medical complexity having their basic needs met" (p.6)
Inclusive Education	"Access to an education system that fully supports children with medical complexity and allows the opportunity for maximal participation in school" (p.6)
Child Social Integration	"The opportunity for fully social immersion and acceptance by a community that empowers children medical complexity" (p.6)
Current Child Health-Related Quality of Life	"Physical, emotional, and social aspects related to the health and developmental status of children with medical complexity" (p.6)
Long-Term Child and Self Sufficiency	"The presence of confident and self-reliant management of care of children with medical complexity" (p.6)
Family Social Integration	"Access to family social supports that allow the family to fully" (p.6)
Community System Supports	"Access to social and physical supports that allow children with medical complexity to navigate their homes and communities" (p.6)
Health Care System Supports	"Access to supports that allow children with medical complexity to obtain all needed health care services"

Domain	Description of Domain
High-Quality Patient-Centered Medical Home	"Access to and use of high-quality comprehensive and speciality health care" (p.6)
Family-Centered Care	"The presence of a beneficial partnership between providers, patients, and families that places families at the center of planning and decision-making related to the child or youth's health" (p.6)

(Barnert et al., 2018, p.6)

To date, this body of knowledge has lacked theoretical and philosophical guidance. A closer examination of current literature reveals the inclusion of concepts borrowed from other frameworks and disciplines. While the theoretical underpinnings of the conceptual frameworks discussed above were not explicitly addressed, each of these models follow principles outlined within the biopsychosocial model. This philosophical view expands the concept of disability beyond the confines of the dichotomous social and medical models by adopting the understanding that disability and health are affected by multiple factors.

The need for strong conceptual congruency across research and policy domains is critical. Frameworks such as Cohen et al.'s (2011) definitional framework of children with medical complexity and Barnert et al.'s (2018) 10 Domains of Health can provide this body of literature with a strong conceptual foundation. Regrettably, there still remains a dearth of research utilizing these framework to their fullest capacity as conceptual frameworks guiding the research process (Brenner et al., 2018; Cohen et al., 2018).

2.5. THE USE OF HEALTH ADMINISTRATIVE DATA

The following scoping review also appears in: Breneol, S., Curran, J.A., McCulloch, H., Shin, D.H., Montelpare, W., Martin-Misener, R., Macdonald, M., Vine, J., Stewart, S. (2022). Improving Health Care for Children with Medical Complexity Through the Use of Health Administrative Data: A Scoping Review. *Child: Care, Health and Development.* Submitted.

Statement of manuscript contribution: SB conceptualized the review concept with guidance and input from JAC, WM, RMM, MM, JV, and SS. SB implemented the search strategy, screened all potential literature, extracted data, synthesized findings, and drafted the manuscript. DHS and HM were the second reviewers and data extractors. All authors contributed to revising this manuscript.

2.5.1. Introduction

With a marked increase in children with chronic conditions over the past 50 years (Perrin et al., 2014), many health care professionals, researchers, and policy makers are now directing their attention and efforts towards children with medically complex needs (Cohen et al., 2018). High levels of health care utilization, fragmented coordination of care and difficulties in accessing services are just a few of the cited challenges being faced by these children and their families (Brehaut et al., 2011; Cohen et al., 2018; Kuo et al., 2011; Kuo et al., 2014). To date, there has been difficulty in identifying and conceptualizing pediatric 'medical complexity' at both the population and individual level (Berry et al., 2015; Lindley et al., 2021). In comparison to the adult population, where a small number of conditions encompass a large proportion of individuals with medically complex needs, the population of children with medical complexity is

comprised of diverse and wide-ranging disorders (Berry et al., 2015). In an attempt to unify this pediatric population irrespective of specific diagnoses, Cohen et al. (2011) developed a definitional framework outlining four domains characteristic to this cohort: (i) the presence of a complex chronic condition(s); (ii) high health resource use across multiple services and settings; (iii) functional limitations; and (iv) high family identified needs. While this provides a broad conceptual understanding of medical complexity, operationalizing this framework to a population level may pose unique challenges (Berry et al., 2015).

Population level exploration of children with medical complexity and their families is critical to identifying areas of practice and policy reform. Health administrative data are one source of population-level data that are being increasingly used for public health surveillance, examining health system utilization, and informing policy and practice recommendations (Cadarette & Wong, 2015; Hinds et al., 2016; Mazzali & Duca, 2015). These databases contain copious amounts of health data that are routinely collected and stored for administrative and billing purposes (Mazzali & Duca, 2015; Mohammed & Stevens, 2007). There are a variety of advantages to harnessing these data sources for research purposes, such as the potential for large sample sizes, ability to link multiple databases, and capacity to access multiple years of information (Mazzali & Duca, 2015). However, these advantages come with limitations, including the potential for misclassification of the phenomena of interest and the inability to control data collection procedures. This often leads to questions regarding data quality, as well as the possibility that databases may lack clinical variables of interest (Hinds et al., 2016; Mazzali & Duca, 2015). Previously demonstrated utility in studying patient populations

such as functional decline in adults (Rao et al., 2016) and end-of-life cancer care (Langton et al., 2014), suggests there is potential to leverage health administrative data to study children with medical complexity. It is currently unknown how health administrative data are being used to inform and develop health practice and policy recommendations for this vulnerable and emerging population.

2.5.2. Review Aim and Questions

The aim of this scoping review was to examine how health administrative data are informing the design of health practice and policy recommendations for children with medical complexity and their families. This review was driven by the following research questions: (i) How have health administrative data been leveraged to develop health care practice and policy recommendations for children with medical complexity and their families? (ii) How have included studies defined/identified children with medical complexity within health administrative data? and (iii) What are the reported strengths and limitations of using health administrative data for the study of children with medical complexity?

2.5.3. Methods

Scoping reviews are used to identify existing evidence in relation to a specific research area (Arksey & O'Malley, 2005; Joanna Briggs Institute, 2015). In contrast to systematic reviews, scoping reviews propose broad research questions to explore and examine key concepts and knowledge gaps (Arksey & O'Malley, 2005; Joanna Briggs Institute, 2015). To ensure a systematic and replicable process, this scoping review followed the steps outlined in JBI Methodology for Scoping Reviews (Aromataris &

Munn, 2020) and the reporting guidelines of the PRISMA Extension for Scoping Reviews (ScR) (Tricco et al., 2018).

2.5.3.1. Data Sources and Search Strategy

This review began with an initial search of PubMed to identify keywords, concepts, and citations in relation to this topic of interest. Relevant citations were further reviewed to identify potential index terms and MeSH headings. The final search strategy was then adapted and last implemented on July 21st, 2021 across three electronic databases: PubMed, CINAHL, and EMBASE (See Appendix A). Databases were searched from inception. Three high-impact pediatric journals (Journal of Pediatrics, Pediatrics, and BMC Pediatrics) were chosen to undergo a hand search of titles and abstracts for the past eight years (October 2013-October 2021) to identify any additional citations. Further, sources of unpublished, published, and grey literature were identified through a targeted Google search.

2.5.3.2. Eligibility Criteria

This review considered studies that examined children and youth with medical complexity as the population of interest. To ensure inclusivity to the variety of terminologies used to describe this cohort of children, articles were considered for inclusion if authors described their primary population of interest using any variation of the terms medically fragile or complex. Given that many youth with medical complexity face challenges in transitioning to the adult health care system (Campbell et al., 2016; Porepa et al., 2017), this review remained flexible in regards to upper age limits as long as authors described their primary population of interest as being within the field of pediatric care. Articles focusing on specific subpopulations (i.e., neurological disorders)

or broader populations of interest (i.e., children with special health care needs) were excluded. This review considered articles regardless of the specific health concept under study. The context under consideration was within the health care system. This review considered all care settings and geographical regions. Only empirical studies or quality improvement projects describing the use and analysis of routinely collected health administrative data were considered for inclusion. Abstracts were included if there was sufficient information to discern eligibility. Reference lists of relevant articles were examined for additional studies of interest. No date limitations were set but for resource limitation reasons the review was limited to English language only.

2.5.3.3. Screening

All identified citations were uploaded into Covidence Systematic Review Software (Covidence Systematic Review Software, 2018). This review followed a two-stage screening process to identify relevant articles. Two independent reviewers reviewed all title and abstracts against the eligibility criteria. Next, two reviewers independently screened the full texts of all remaining articles to determine final inclusion. Any disagreements in either stage of the review process were resolved through consensus. Reasons for exclusion during the full-text screening stage were recorded and reported in the PRISMA ScR.

2.5.3.4. Data extraction

A standardized data extraction form was adapted from that provided by JBI (See Appendix B) (Joanna Briggs Institute, 2015). One independent reviewer extracted all data from included articles, with a second reviewer validating extracted data. Any disagreements in extracted data were resolved through consensus.

Critical appraisals are not a required step in the scoping review methodology (Joanna Briggs Institute, 2015; Tricco et al., 2018). The goal of this review was to understand the use of health administrative data in the study of children with medical complexity, thus critical appraisal was not conducted.

2.5.3.5. Data analysis

Data were analyzed and are presented in both tabular and narrative formats. Findings were thematically grouped to address each review question and narratively reported.

2.5.4. Results

The implemented search strategy resulted in 7552 unique citations. A total of 92 articles and conference presentation abstracts were deemed relevant for inclusion in this review (Figure 1).

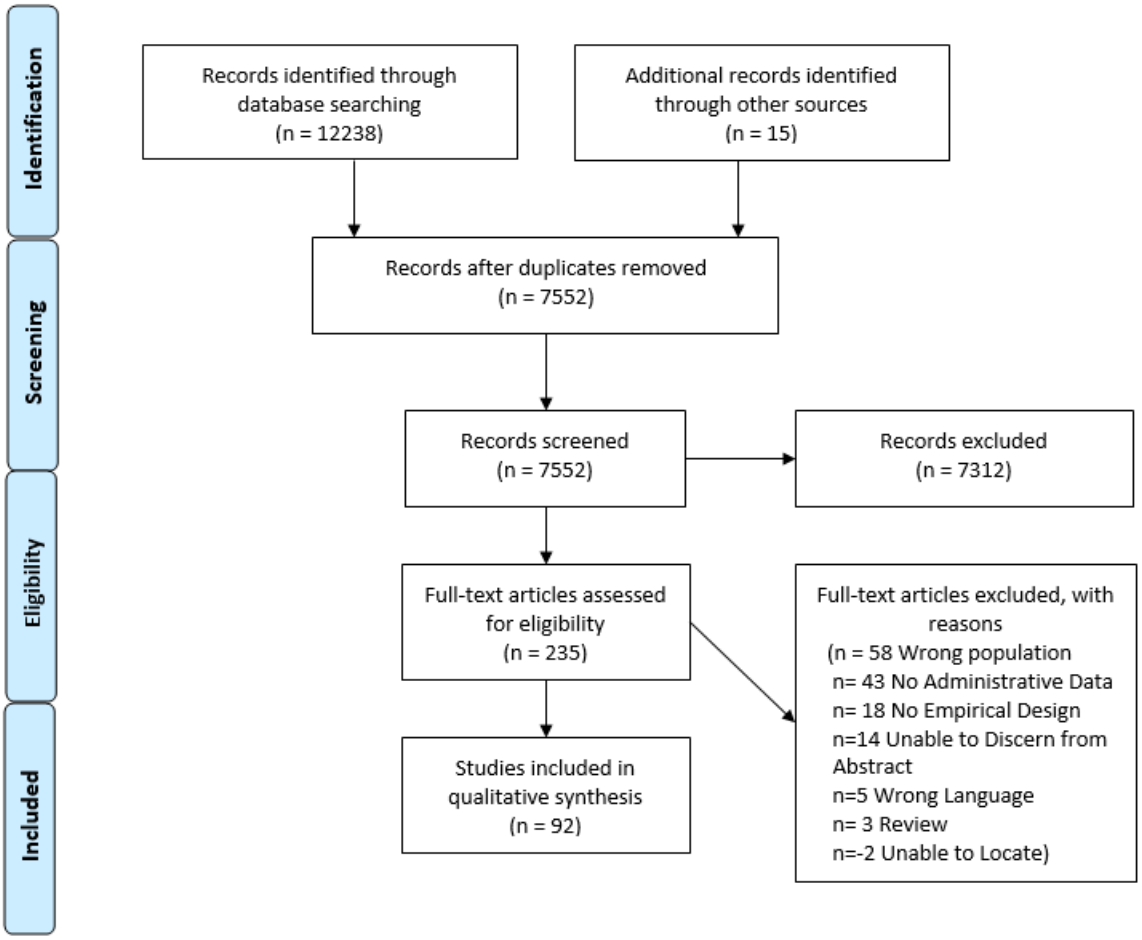

Figure 2-1. PRISMA Diagram

2.5.4.1. Characteristics of Included Studies

A variety of research methods were employed in included studies (See Table 2-2). Retrospective cohort designs (n=59) were most common followed by retrospective case series (n=6), cross-sectional designs (n=6), and development and/or validation studies (n=6). Most studies used health administrative data from the United States of America (US) (n=68), with other articles using data from Canada (n=10), Portugal (n=3), Australia (n=3), Korea (n=2), Brazil (n=1), Belgium (n=1), Japan (n=1), Sweden (n=1), Netherlands (n=1), Spain (n=1), and an amalgamation of 15 different countries (n=1). All

included articles were published between 2000-2021. A variety of data sources were reported, including medical claims, hospital, regional, and national databases. Only five studies combined the use of health administrative data with family reports by way of surveys, focus groups, or interviews (Arthur et al., 2018; Canadian Institute for Health Information, 2020; Cohen, Lacombe-Duncan, et al., 2012; Coller, Klitzner, et al., 2018; Kingsnorth et al., 2015).

2.5.4.2. Definition/Identification of children with medical complexity

A variety of terminologies, conceptual definitions, and identification methods were used across all included studies to determine and describe 'medical complexity' (See Table 2-3). The majority of articles (n= 34) referred to this population of interest using Feudtner et al.'s (2000) working definition for children with complex chronic conditions. Another commonly used description was Cohen et al.'s (2012) conceptual definition for children medical complexity (n=14). Further, most authors identified their study cohort in health data using the original and revised Pediatric Complex Chronic Conditions (CCC) Classification System, which is comprised of diagnostic codes associated with complex chronic conditions (n=51) (Feudtner, 2000; Feudtner et al., 2014). Other methods of identification included the Pediatric Medical Complexity Algorithm (PMCA) (n=8), Clinical Risk Groups (n=8), hospital-based programs for complex patients (n=6), pre-determined lists of International Classification of Diseases (ICD) diagnostic codes (n=6), health service use thresholds (n=5), Chronic Conditions Indicators (n=2), Adjusted Morbidity Groups Classification System (n=1), hospital registries (n=1), and point of care complexity screening algorithms (n=1). Seven studies used a mix of strategies listed above to identify their cohort.

Six studies were included that developed, validated, or updated an algorithm to identify children with medical complexity in health administrative databases. First, Neff et al. (2015) assessed the sensitivity and specificity of using Clinical Risk Groups to stratify children into three condition categories: complex chronic, noncomplex chronic, and nonchronic. Sensitivity and specificity were strong across all conditions except for poor sensitivity in classifying noncomplex chronic condition (Neff et al., 2015). Second, the Pediatric Medical Complexity Algorithm (PMCA) has been validated to identify and classify children by varying levels of medical complexity: complex chronic conditions, noncomplex chronic condition, and without chronic conditions (Simon et al., 2014, 2017, 2018). The most current version, PMCA 3.0, has 'good' to 'very good' sensitivity for classifying children with complex chronic conditions and without chronic conditions, but poor sensitivity for children with noncomplex chronic conditions (Simon et al., 2018). Third, Feudtner et al. (2000; 2014) evaluated the updated version of their CCC Classification System (Feudtner, 2000; Feudtner et al., 2014). Fourth, Lindley et al. (2021) compared the predictive performance of the original and revised versions of the CCC Classification System and found good agreement between measures (Lindley et al., 2021).

Table 2-2. Characteristics of Included Studies

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Amin et al. (2018)	-Retrospective Cohort Design -Original Research Paper	-To determine the prevalence of persistent obstructive sleep apnea post adenotonsillectomy in children with complex chronic conditions and to identify predictors of persistent postoperative obstructive sleep apnea to help guide selection of surgical candidates	-Having a complex chronic condition was a strong predictor of persistent obstructive sleep apnea post adenotonsillectomy	-Adenotonsillectomy is considered the first-line therapy in healthy children >2 years old by the American Academy of Pediatrics; however, other surgical procedures or non-surgical management may need to be considered as first-line treatment for this cohort	- None noted
Ananth et al. (2015)	-Retrospective Cohort Design -Original Research Paper	-To describe the characteristics, hospital use, and costs for children with life threatening complex chronic conditions in the last year of life, compare resource use by type and number of conditions, and examine medical interventions received in the terminal admission	- Hospital use in the last year of life varies considerably type and number of conditions - 2/3^{rds} of children had multiple conditions, which was a major factor for increased hospitalizations, days spent in the hospital, and cost	- Clinicians may find this information useful to optimize care - Anticipatory consideration of the time they could spend in the hospital might aid in advance care planning - Clinicians can use this information to better inform and discuss possibilities of future hospital use based on prognosis and to ensure that hospital interventions are in accord with child and family wishes	- Explore hospitalization in greater detail to improve our understanding of how hospital care can be best utilized near the end of life - Examine specific disease trajectories - Examine the effect of medical interventions on the child and family experience at the end of life - Care delivery model that integrates primary, specialty, and surgical providers should be examined

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Arthur et al. (2018)	-Retrospective Cohort Design -Original Research Paper	- To explore the construct validity of a claims-based measure of continuity of care as a potential indicator of care coordination quality for children with medical complexity	- Higher continuity of care score was associated with lower odds of ED utilization and higher probability of caregivers reported receiving needed care coordination services	- Implementation of a continuity of care measure in commercial health plans and state Medicaid agencies could stimulate quality improvement efforts at the practice and health plan levels	- Convergent validity of continuity of care with other previously validated care coordination measures could help establish continuity of care as a measure of care coordination quality.
Avritscher et al. (2019)	-Post Trial Follow-Up -Quality Improvement Project	-To assess the sustainability of the benefits relative to usual care of a medical home providing comprehensive care for high-risk children with medical complexity after comprehensive care was created and expanded as a standard practice	-Mean total health system costs per child-year were lower post-trial for those in the comprehensive care group -Usual care costs did not change significantly -The rate of children with serious illnesses in practice were similar to those in trial	-Benefits identified seem to only be achievable in academic centres with adequate staffing and resources. -Real world implementation and expansion of the program suggests scalability of comprehensive care -Inadequate reimbursements is a barrier for implementation and this needs to be in place for providers to see large potential reductions in expenditures	-Explore the outpatient pharmacy costs
Bennett et al. (2020)	-Retrospective Cohort Design -Original Research Article	-To utilize hospital electronic medical record data for children placed in foster care and a matched control group to compare: 1) health care utilization rates for primary care, subspecialty care, ED visits, and hospitalizations; 2)	-Disproportionately higher rates of children with CCCs -Those with CCCs have higher utilization and charge (cost) outcomes	-Emphasize the importance of the role of care coordination -Medical home may be one model of care from which this population could benefit	-Need to integrate child welfare and hospital data to improve our ability to understand patterns of health resource use and provide optimal care coordination

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
		overall charges per patient-year; and 3) prevalence of CCCs and their effect on utilization.			-Evaluate specific diagnoses for inpatient admissions
Bergman et al. (2020)	-Prospective Cohort Design -Original Research Article	- To assess the impact of a multicenter care management program on spending and use in CMC	- 4.6% decrease in the total per-member per-year spending, 7.7% decrease in inpatient spending, and 11.6% decrease in ED spending	- Expansion of this model to reach all CMC will require more of a consultative and educational approach to these practices - This model of care demonstrated a reduction in total spending for CMC cared for in both hospital-based complex care clinics and pediatric primary care providers	- Determine which components of the intervention most contributed to improvement in outcomes - Determine the feasibility of scaling and spreading this intervention to other integrated health care systems as well as the sustainability of improvements over time
Berry et al. (2011)	-Retrospective Cohort Design -Original Research Paper	- To describe the demographic and clinical characteristics of patients who receive care within different structured outpatient and inpatient complex-care clinical programs for CMC; and describe their indications for and use of inpatient resources	- Hospitalized CMC experienced multiple, lengthy hospitalizations - Reasons for hospitalizations suggest that certain hospitalizations may be avoidable with improved delivery of care	- Reduce early hospital readmissions - Early readmission to technological malfunction may be avoidable with implementation of high quality operative and procedural techniques -Identify best practices for inpatient respiratory management and improve provider's attitudes and skills	- To understand the patient and health system factors that influence inpatient resource utilization and determine which factors are ameliorable with improved delivery care

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
				-Administrators of complex care programs may consider recruiting staff with expertise in neurological impairment - Complex care programs should track their health care utilization	
Berry et al. (2017)	-Retrospective Cohort Design -Original Research Paper	- To assess the fraction of USA hospital discharges, bed days, and hospital charges attributable to children with multiple chronic conditions, and determine which chronic conditions have the largest impact on hospital care for children	- Account for 1 of every 4 acute-care hospitalizations and 1 of every 2 dollars of pediatric hospital care costs - Hospitalized cohort were more often older and non-Hispanic African American - Mental health conditions and at least 1 additional chronic condition accounted for the most hospital days	- Hospitals could benefit from exploring the efficiency and effectiveness of inpatient care delivery - States, federal agencies, and others may benefit from assessing systems of care to ensure that their hospital use is appropriate and delivered at the highest level of quality	- Investigate how to best provide high-quality inpatient care to children with mental health conditions with additional medically based comorbidities
Berry et al. (2013)	-Retrospective Cohort Design -Original Research Paper	- To assess trends for children who were presumed to be relatively healthy without a chronic condition and for children with chronic conditions of varying medical	- Cohort of hospitalized children with significant chronic conditions that affected 2 or more body systems or were complex or	- Develop financial strategies to reimburse the costly inpatient needs of children with CCCs - Develop structural plans to accommodate growth in inpatient resource use	- Characterization of hospitalization trends in non–children's hospitals

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
		complexity within a national cohort of children's hospitals	progressive increased at the highest rate - High prevalence of primary conditions associated with neurologic impairment and comorbidities	- Encourage the identification of children with CCCs who may benefit from proactive hospital and community care integration	
Berry et al. (2014)	-Retrospective Cohort Design -Original Research Article	- To improve our understanding of the opportunities and challenges for better care management and reduced health care spending for CMC with Medicaid	- Account for half of Medicaid spending on hospital care for all children and is increasing over time - Small subset of the population accounted for the most cost - Spending for primary and home care is small	- The implementation of care management activities to facilitate a smooth discharge and prevent readmission - Neurological conditions may greatly benefit from targeted care management	- Examine healthcare encounters over time -Compare effectiveness of different care management approaches and learn how to disseminate such practices - Develop a national publicly available data base of healthcare cost and utilization to assess longitudinal trends
Berry, Glotzbecker, et al. (2017)	-Retrospective Cohort Design -Original Research Paper	- To assess health care resource use and spending on behalf of CMC undergoing spinal fusion and to simulate a global payment reallocation with increased use of primary care followed by decreased hospital use	-Initial and subsequent hospital readmissions were responsible for nearly $8 of every $10 spent on perioperative care - Primary care health services accounted for a negligible amount of spending	- Preoperative aspects of outpatient care may help optimize the children's health ahead of surgery - Examination of clinical guidelines regarding preoperative laboratory testing in the absence of clinical indications should occur - Encourage multidisciplinary communication to advance their	- Investigate the role of primary care in perioperative care to assess its value and potential impact on cost savings - Investigate the utility and value of this preoperative testing with cost savings

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
			- Multiple preoperative primary care visits were associated with lower length of stay and spending	knowledge of perioperative health resource use and spending	
Bjur et al. (2019)	-Repeated Cross-Sectional Design -Original Research Article	- To determine the prevalence and incidence of children with multiple complex chronic conditions living in a mixed urban-rural community and examine temporal trends over the past 15 years.	- Five-year prevalence and incidence rates increased from 1237 to 1961 per 100,000 persons, and 256 to 335 per 100,000 persons, respectively - Tend to be slightly more prevalent in lower socioeconomic status and with a racial minority background	- Findings can be used to inform health policy at national and local levels because these children have high health care expenditures and payer source is more likely to be public	- Assess the impact of pediatric CCCs on financial outcomes of families and the total national health care costs - Investigate factors of health equity
Brittan et al. (2015)	-Retrospective Cohort Design -Original Research Paper	- To analyze the association between post-discharge outpatient follow-up visits and 30-day readmissions in Medicaid-enrolled children with CCCs	- Children with an outpatient visit between 4- 29 days of discharge had lower odds of readmission than those without such visits - Children with at least 1 outpatient visit within 3 days of discharge had greater odds of readmission than those without a 0-3 day visit	- Provides a foundation for developing recommendations on outpatient follow-up - Recommendations for longer term post discharge follow-up - Should be family-centered	- Develop a more detailed description of outpatient visit characteristics - Explore the relationship between differing follow-up times and readmission end points to clarify future guidelines

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Brown et al. (2021)	-Retrospective Cohort Design -Original Research Article	- To examine concomitant recent trends in (1) medical complexity, (2) LOS, and (3) readmission rates among patients admitted to U.S. children's hospitals	- There was an increase in the proportion of hospitalized patients with complex chronic conditions (40.1 to 41.5%) or receiving intensive care (10.8 to 12.3%) - Mean length of stay for the patients with complex chronic conditions was stable -14-day readmissions declined for those with CCCs	-Pediatric hospitals are managing inpatient more efficient despite increase complexity -Small changes in length of stay can meaningfully impact hospital systems -Specific conditions' length of stays are increasing, such as mental health. -Hospitals should continue to explore how to improve care transitions, enhance care coordination, and intervene on the social determinants of hospital use	- Exploration of specific condition groups and individual diagnoses with reduced readmission rates could provide understanding for how lower readmission rates can be achieved despite increasing medical complexity
Burns et al. (2010)	-Retrospective Cohort Design -Original Research Article	- To determine changes in the prevalence of hospital admissions for medically complex children over a 15-year period	- Hospital admission rate increased significantly - Hospitalization rates of children with diagnoses in multiple CCCs increased	- Improve tracking of children with special health care needs to better understand the patients and their health care and care-coordination needs - Improving resident education in the care of these children both acutely and in their communities.	- Investigate the value of a hospital based medical home

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Buser et al. (2020)	-Retrospective Cross-Sectional Design -Original Research Article	- To perform a detailed analysis of the asylum-seeking children with frequent visits, detailing their underlying medical conditions and analyzing care provided	-4% of patients met the inclusion criteria -In children less than 2, genetic disease and nutritional problems were most common -In adolescents, orthopedic disease and mental health problems were most common	- Specific nutritional treatment programs in refugee camps and an active screening for malnutrition with early interventions are needed - Mental health screening should be included in routine health screening -Psychosocial and therapeutic treatment should be adapted to the needs of asylum-seeking adolescents - Primary care access into local health care system in a setting may offer the possibility of improved integrated care	-None noted
Carrilero et al. (2020)	-Cross-Sectional Design -Original Research Article	- To describe the pathologic patterns of CMC and their socioeconomic inequalities to better manage their needs, plan healthcare services accordingly, and improve the care models in place	-71% of CMC had at least one parent with no employment -Four comorbidity class were identified: oncology, neurodevelopment, congenital/perinatal, and respiratory	-More disadvantaged children represent higher proportion of CMC and children are subject to inequalities from the beginning of their lives -Creating more efficient multidisciplinary teams according to each comorbidity class -Introduce policies to support both their health and financial situation	-Explore the correlation between socioeconomic inequalities and CMC status

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Casey et al. (2011)	-Retrospective Cohort Design -Original Research Article	- To evaluate the effect on all state Medicaid costs of a children's hospital-based multidisciplinary clinic that provides comprehensive and coordinated care for medically complex children	- Clinical intervention resulted in significantly fewer inpatient stays with significantly shorter lengths of stay - Increased number of outpatient claims but decreased ED visits - Aggregate cost savings were $1179 per pay per month for the year after the first clinic visit	- Expand medical homes beyond this complex population - The medical home should be seen as the concept of comprehensive, coordinated, and accessible care that all children deserve - The expanded multidisciplinary team offers the opportunity to have the coordinated and comprehensive care	- Determine whether the coordinated and comprehensive care resulted in better patient quality of life and health status for the child and family
Chan et al. (2016)	-Retrospective Cohort Design -Original Research Paper	- To examine the proportionate use of critical care resources among children of differing medical complexity admitted to pediatric intensive care units in tertiary-care children's hospitals	- Disproportionate usage of resources and therapies - Patients with CCCs use the vast majority of resources and therapies and those with progressive conditions use more than half of all resources	- Standardized critical care processes and research protocols should be designed with the inclusion of CMCs, by subtype - Standardized care protocols based on frequent planned procedures and common admission diagnoses - Standardizing use of palliative care services	- Investigate procedural admissions for quality, outcomes, and cost-effectiveness to optimize resource use

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
CIHI (2020)	-Retrospective Cohort Design -Pan-Canadian Report	-To provide a better understanding of the landscape of CMCs across Canada along with the differences across provinces and territories	-Variation across provinces and territories -In 2015-2016, the age adjusted rate was 948 per 100,000 -Require higher use or primary health are, hospital care, and ED care -Use multiple medications -Data on home care is limited	-Families and health care providers want better data to describe this population to facilitate advocacy and improved service delivery -Integrated care coordination may be one way to optimize care delivery and decrease hospital stays	-Perform ongoing monitoring and surveillance efforts for this population -Assess trends over time -Explore the role mental health disorders have in this population
Cohen et al. (2010)	-Mixed Methods Descriptive Single Centre Pre or Post Evaluative Design -Original Research Article	- To evaluate the impact of a complex care clinic in a children's hospital on healthcare utilization, parental and primary care provider perceptions of care and parental quality of life	-Number of days admitted to hospital decreased from a median of 43 to 15 days, and outpatient visits increased from 2 to 8 -Mean parental quality of life-scores for three domains improved related to mental health -Primary care providers found the clinic helpful for families -Parents reported improved continuity of care, family-	-Continue the development of ambulatory complex care programmes from the perspectives of patient, families, and health systems -Establishment of a complex care clinic is unlikely to be associated with large additional costs -Areas for continued improvement include optimizing communication, minimizing bureaucracy and duplication of services, and providing appropriate remuneration for providers.	-Conduct a rigorous evaluation of the clinic using a randomized controlled trial design or a non-equivalent groups design -Explore the impact of a complex care programme for those that were not admitted to hospital

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
			centeredness of care, and comprehensiveness and thoroughness of care, but still experienced frustrations with access to services and miscommunication	-Complex care clinics in a children's hospital may improve the quality of care	
Cohen et al. (2012)	-Retrospective Cohort Design -Original Research Article	- To evaluate health care utilization and costs in a population based sample of CMC in Ontario, Canada.	- Cohort constitutes approximately 0.67% of the pediatric population - Account for 1/3rd of health care spending on all children - Mortality rates were not high	- Care coordination interventions - Extend clinical programs/care management interventions to the home and community care setting may be warranted - Care coordination of an important issue	- Determine which interventions can decrease costs associated with unnecessary utilization and improve outcomes
Cohen, Lamcombe-Duncan, et al. (2012)	- Before/after Intervention Study Design - Mixed methods -Original Research Article	- To evaluate the effectiveness of a novel community–based complex care clinic integrated with a tertiary care facility	- Families experienced long-term cost savings - Care was more likely to be delivered in a community setting - Families and health care providers were highly satisfied with reports of family-centeredness of care	- Support the development of complex care models focused on tertiary care-community based partnerships - Accountable Care Organizations may prove vital to financially integrate organizations across the care continuum - Encourage formal partnerships between children's hospitals and community hospitals in care coordination, together with family	- None noted

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
				engagement and the primary care providers	
Coller et al. (2018)	-Retrospective Cohort Design -Original Research Article	- To evaluate ambulatory care sensitive hospitalizations for children with noncomplex chronic disease and CMC, and identify associations with ambulatory care characteristics	- Among CMC, hospitalizations for these conditions may be less sensitive to ambulatory care	- Tailor interventions to the patient's underlying medical complexity continuum - Ambulatory care sensitive hospitalizations for non-complex chronic may be more "modifiable"	- Examine interventions with a focus on provider continuity, identify different ambulatory care constructs, or target a different set of conditions altogether - Prospective study to refine measurement quality of ambulatory care
Coller et al. (2019)	-Retrospective Cohort Design -Original Research Article	-To evaluate factors associated with admission from ED encounters for CMC and to quantify the hospital admission rate as well as variation in adjusted hospital admission rates across EDs	-Hospital admission happened in around 26% of ED visits -Variation exists in hospitalization rates for children with medical complexity	-Children going to the ED overnight may have various limited resources that results in the need for hospital care -Communities with high quality comprehensive care could support more clinicians to safely discharge these patients from the ED, resulting in lower admission rates	-Clarify how information on prior use affect future decision making on disposition -Unravel the racial/ethnic and other sociodemographic disparities in ED admissions -Explore differences in provider training which is associated with admission rate variation -Explore underlying causes of rate variations and intervention that could reduce admissions -Undertake a longitudinal study

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Coller Klitzner et al. (2018)	- Randomized Controlled Trial -Original Research Paper	- To examine the effect of a caregiver coaching intervention, Plans for Action and Care Transitions, on hospital use among CMC within a complex care medical home at an urban tertiary medical center	- Reduced hospital use for those already enrolled in a mature complex care program - Those enrolled in the intervention had fewer all-cause 30-day readmissions and were less likely to experience at least 1 readmission	- Programs with existing staff to implement this intervention activities and those with newer payment models may find this intervention more sustainable - Even after enrollment in an established medical home with care coordination, potential to further reduce hospital use may be possible	- Determine whether such findings are replicated in different populations and complex care clinical settings
Coquillette et al. (2015)	-Retrospective Cohort Design -Original Research Paper	- To quantify hospital-wide social work services utilization by CMC compared to non-medically-complex children to inform the development of family-centered care models that support these vulnerable patients and families	- CMC were more than six times as likely to use social work services -On average, social workers spent more than eight times as many hours with CMC	- Proactively address social work services to the primary care setting, further increasing the capacity needed to support these families - Nonlicensed support staff such as patient navigators or resource specialists may fill a subset of social work roles -Incorporation of personnel who can help families negotiate complex systems and access concrete resources -Develop written or electronic resources could assist families with certain common needs	- Understand factors driving increased use and outcomes of social work interventions in terms of patient satisfaction and health outcomes

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Edwards et al. (2010)	-Retrospective Cohort Design -Original Research Paper	- To estimate the risks associated with CCCs for children admitted to pediatric intensive care services using regression methods modeled on a nationally representative sample	- 23% had a documented CCC -Having a CC increased the odds of death during PICU admission by 43% compared to not having a CCC	- None noted	- Intensivists must continue to devote efforts to study and mitigate the impact of CCCs on children
Edwards et al. (2012)	-Retrospective Cohort Design -Original Research Paper	- To estimate the prevalence of chronic conditions among children admitted to U.S. pediatric intensive care units and to assess whether patients with CCCs experience mortality and prolonged length of stay risk beyond that predicted by commonly-used severity-of-illness risk-adjustment models	- Children with CCCs comprised just over one-half of admissions - Have significant risk for pediatric intensive care unit mortality and prolonged length of stay beyond that predicted by commonly-used severity-of-illness models	- Attention and focus needs to be paid to the needs of this population - For PICU mortality benchmarking, additional inclusion of high-risk complex chronic conditions subcategories into future risk-adjustment models would improve model accuracy	- Investigate whether CCC rates are lower in the community - Examine whether inclusion of CCCs into risk-adjustment models has more impact when a more diverse sample of PICUs is evaluated
Feinstein et al. (2014)	-Retrospective Cohort Design -Original Research Paper	- To describe the characteristics of adverse drug events related ED visits, including association with CCC status; determine the implicated medications; and determine if CCC status increased the risk of adverse drug events-related admission	- Increased risk of ED visits related to adverse drug events - Highest rates attributed to psychotropic agents, antimicrobial agents, anticonvulsants, hormones/steroids, and analgesics	- There is little evidence to guide best practice in children CCCs even for routine medications	- Understand the reasons for the increased risk of adverse drug events among children with CCCs to enhance monitoring strategies - Examine additional ambulatory settings

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Feudtner et al. (2000)	-Retrospective Cohort Design -Original Research Paper	- To determine if an increasing proportion of pediatric deaths were attributable to an underlying CCC, what they typical age of the death, and weather this age is increasing	- Substantial portion of pediatric deaths is attributable to an underlying CC - Deaths with an underlying CCC declining less rapidly than deaths without	- Comprehensive planning of support care service should use a unifying concept to assess this population and its needs - Pediatric supportive care service must be flexible due the dynamic and individual characteristics of families - All families should be offered supportive care services	- A greater understanding into the need of this population
Feudtner et al. (2001)	-Retrospective Case Series -Original Research Paper	- To identify trends over the past 2 decades in the pattern of deaths attributable to pediatric CCCs, examining counts and rates of CCCs-attributed deaths by cause and age at the time of death, and to determine the average number of children living within the last 6 months of their lives	- Approx. 15 000 infants, children, adolescents, and young adults die from complex chronic conditions each year - Mortality rates are declining across all age groups	- Focus on the pediatric to adult transition - Two types of palliative care services may be required depending on patient characteristics (i.e. cancer vs congenital conditions)	- None noted
Feudtner et al. (2002)	-Retrospective Cohort Design -Original Research Paper	- To describe the demographics of children who die in children's hospitals and to describe the prevalence of CCCs among these cases	- Children's hospitals care for a substantial number of dying patient with varying characteristics - Children who die with CCCs experience both longer periods of mechanical ventilation	- Palliative care services need to accommodate a wide range of symptoms and coordinate with many specialty services - Identifying these children in a timely manner could provide a method of estimating an average daily census to guide	- None noted

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			and of hospitalization before death	staffing required to meet the family needs - Prepare palliative care teams for the challenges of limiting or withdrawing care	
Feudtner et al. (2003)	-Retrospective Cohort Design -Original Research Paper	- To describe the hospital care received in the last year of life by children and young adults who died and the proportion with CCCs and whether the use of hospital services increased as the date of death drew nearer	- Children with CCCs were significantly more likely to be hospitalized and to experience mechanical ventilation or some other procedure - Older children and young adults, a substantial proportion spent weeks to months in the hospital during the last year of their lives	- For infants, a hospital-based service system may be well situated to deliver timely and continuous end of life services the timely delivery of effective hospital-based - For older children, outpatient, community-based, or at-home services would typically be necessary to care for them when they are not hospitalized -Multi-sectoral collaboration is key	-Determine ways to incentivize health care providers and institutions to deliver complementary care and measure the degree to which such care is delivered - Evaluate patient-outcomes
Feudtner Silveria et al. (2002)	-Retrospective Case Series -Original Research Paper	- To test whether deaths attributable to underlying CCCs were increasingly occurring at home and to determine what features were associated with home deaths	- A greater proportion of deaths with an underlying CCC occurred at home - Likelihood of a death at home appears significantly modified by both biological and	- None noted	- Deepen our understanding of how biological, technological, social, and cultural factors influence where children with CCCs die

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			social factors and geographic regions		
Forjaz de Lacerda et al. (2017)	- Retrospective Cohort Design -Original Research Paper	- To examine the epidemiological situation of pediatric deaths in a country without pediatric palliative care, describing trends in cause and place of death and factors related to home-death in children with CCCs	- Without pediatric palliative care provision, there was a long-term trend of dying away from home - Overall, we found that CCCs were attributing to an increasing portion of pediatric deaths	- Neonatology should be a greater priority for pediatric palliative care - Implementation of pediatric palliative care services to ensure adequate home support for families	- None noted
Friedel et al. (2019)	-Retrospective Cohort Design -Original Research Paper	- To identify, over a 5-year period (2010–2014), the number of children and adolescents (0–19 years) living with a complex chronic condition, and also their referral to pediatric liaison teams	- 22 721 children/adolescents were diagnosed with-a complex chronic condition --1.7% were referred to a pediatric liaison team	- The number of referrals to pediatric liaison teams is probably insufficient or being reserved for the most complex conditions	- Estimate the type of care needed by children with CCCs - Understand why children are not being referred to the pediatric liaison team
Gay et al. (2016)	-Retrospective Matched Cohort Design -Original Research Paper	- To assess the impact of post discharge home health services on future hospital use in medically complex children	-Home health cases had higher percentage of CCcs, technology assistance, and neurological impairment than the controls -30-day readmission rates were lower for	-Home health may provide both anticipatory guidance and early direct treatment of health decline for those with CCCs -Home health care may be particularly effective at limiting hospital use for those with CCCs	-Investigate why home health was associated with lower hospital resource use over time -Explore how quickly home health is equipped to respond to children's health needs when compared to

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			home health patients, particularly those with CCCs -12 months after the index admission, home health patients average fewer admissions, fewer days in the hospital, and lower hospital costs	-Shifting care to nonhospital environments may have the potential for significant cost savings for hospitals and integrate medical systems -Care in the home environment may provide great comfort and satisfaction based on fewer missed days of work, reductions in travel time and expense, and the reassurance of familiar surroundings.	primary and other types of care -Compare financial data across the care continuum when assessing the impact of home health in children -Assess how much clinical stability plays a role in the impact of home health on children with complex chronic conditions -Prospective analyses with rigorous designs to assess the impact of home health on hospital use -An all-encounter dataset would allow a much-needed assessment across the continuum
Gold et al. (2016)	- Retrospective Cohort Design -Original Research Paper	- To assess the impact of, risk factors for, and variation across children's hospitals regarding long length of stay hospitalizations in CMC	- Small percentage of CMC experience long length of stay account for the majority of hospital bed days and cost in children's hospitals	- Optimize the efficiency of hospital care - Clinical practice guidelines should be created to include children with medical complexity	- Examine intensive care use and whether admission or duration can be safely prevented or abbreviated

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Gordon et al. (2007)	- Pre/Post Intervention Design - Original Research Article	- To evaluate the impact of a tertiary care center special needs program that partners with families and primary care physicians to ensure seamless inpatient and outpatient care and assist in providing medical homes to medically complex and fragile children and youth with special health care needs	- Cohort consumed large amount of resources - Large decrease in resource use post-intervention - Associated with a more than 50% decrease in hospital days and a $10.7 million decrease in tertiary care center payments	- Encourage partnering with the family and primary health care provider - Ensure familiarity with the child's condition - Ensure close involvement during hospitalizations - Ensure proactive outpatient care - Results should promote the use of this type of model	- Replication in other contexts
Guertin et al. (2006)	-Retrospective Cohort Design -Abstract	- To determine, among children dying of a CCC, whether sociodemographic characteristics, specific cause of death, or health care use in the last six months of life are associated with the present dying in an ED	- 13.8% occurred or were confirmed in the ED and this percentage increased over the study period	- None noted	- None noted
Guertin et al. (2009)	- Retrospective case series - Original Research Article	- To determine the percentage of deaths occurring or confirmed in an ED among children dying of CCCs and identify factors associated with that percentage	- Death occurred or was confirmed in the ED for 13.8% of children aged 1–19 years who died of CCCs	- Health services planning and training programs in pediatric palliative care need to take end-of life in the ED into consideration	- None noted

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Håkanson et al. (2017)	-Retrospective Cohort Design -Original Research Paper	- To examine where children with CCCs die and to investigate associations between places of death and sex, cause of death and country	- Hospital was the most common place of death in all countries - In general, deaths caused by neuromuscular diseases and malignancies occurred at home more often than other complex chronic conditions	- Create awareness of pediatric palliative care among several pediatric specialties to expand educational efforts and encourage them to embrace palliative care. - These benchmarks act as a starting point to further explore strategies and policies that contribute to increasing the proportion of home deaths different countries	- None noted
Hannan et al. (2021)	-Retrospective Cross-Sectional Design -Original Research Paper	- To (1) describe the prevalence of medical complexity, specifically the presence of CCCs, in a nationally representative sample of very low birth weight infants (VLBI) and (2) determine the association of specific demographic, hospital, and clinical factors with complex chronic conditions and with either complex chronic conditions or death	- Clinical factors accounted for the highest proportion of the model's ability to predict CCCs or death at 93.3% and 96.3%, whereas demographic factors were 11.5% and 2.3% and hospital factors were 5.2% and 1.4%, respectively - 37.5% of VLBW infants had CCCs	-The prevalence of medical complexity is increasing and as such has impacts on clinical providers -Home health care may decrease hospital use post-discharge and is an area of potential cost savings. -Infants born in rural areas are more likely to have CCCs or die, potentially due to low resources to provide intensive care -Need to address social determinants of health to narrow the disparities at the clinical and patient level	-Impact of VLBW infants with medical complexity on the supply and demand of a home nursing -To mitigate the differences that we have demonstrated across practice locations and regions -Understanding the differences and potential disparities is essential to providing ongoing high-level, equitable care

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				-Deliver the highest-level of care to all potentially complex neonates	
Jamorabo et al. (2015)	- Retrospective Cohort Design -Original Research Article	- To describe mortality trends for Rhode Island resident children aged 0–17 years, along with the demographics, subtypes, sites of death, and comorbidities of those with CCCs	- Pediatric mortalities due to CCCs have recently declined, but still the leading cause of pediatric deaths	- Multidisciplinary approach is essential - Adequate support for families that are coping with their children's illnesses at home	- Determine types interventions that children with CCCs receive at present - Examine family and patient perspectives
Johnston et al. (2019)	- Retrospective Population-Based Design -Original Research Article	- To assess socioeconomic and clinical disparities in the rates of medically intense end-of-life care or intensive care admission in the last 30 days of life for all California children with a CCC	- Average age at death was 11.8 years -Neuromuscular, malignancy, and cardiovascular were the most common categories of CCCs -66% died in hospital, 35% had ≥2 intensity markers -Low-income neighbourhood and age 15-21 years was associated with increased odds of hospital death, medically intense intervention, and ≥2 intensity markers	-Programs aimed at improving end-of-life care may be most successful if they are focus on children CCCs who are cared for by the same subspecialty -Treatment preference discussions during periods of great health may be helpful -Improving end-of-life care will require cultural, literacy-sensitive, and systems approaches -Need for both inpatient and community-based palliative care to meet the needs of this growing population	-Determine if disparities in end-of-life are due to patient and/or family preference, system and/or provider biases -Understand how intensity of end-of-life of care in children with CCCs affects family bereavement -Explore interventions used to ensure that patient with CCCs are receiving goal concordant end-of-life care -Explore subspecialties with the highest rates of high-intensity end-of-life care

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Jurgens et al. (2014)	- Retrospective Cohort Design -Original Research Article	- To determine 30-day hospital readmission rates in children with CCCs discharged from a rehabilitation and transitional care hospital and to identify factors associated with increased risk of readmission	- Similar readmission rates between facilities - Various factors influenced increased risk of readmission	- Subacute hospitals may want to consider enhancing discharge-planning processes for patients with high number of discharge medications	- Examine interventions such as transportation assistance for outpatient appointments, increased home nursing support, ongoing case management, and enhanced communication with the outpatient medical teams
Kalzen et al. (2018)	-National, Prospective, Closed Cohort Design -Original Research Article	- To investigate whether multiple admissions compared to single PICU admissions were associated with poor survival over time after being admitted to PICU facilities and to investigate if the presence of a CCC would further impair prognosis	- Highly significant difference in survival between single and multiple admission patients, intensified by the presence of a complex chronic condition -Multiple admissions with a complex chronic condition was the worst outcome	-None specifically noted	-Larger dataset and multicenter studies
Keim-Malpass et al. (2021)	-Retrospective Cohort Design -Original Research Article	- To examine the prevalence of concurrent hospice care overtime and investigate the relationship between medical complexity and concurrent hospice care among Medicaid children	-34% used concurred hospice -Medical complexity was unrelated to concurrent hospice care -CCCs were negatively related; whereas technology dependence, multiple CCCs, and	-Children with functional limitations, high health care use, and substantial needs might benefit especially from nursing home care as they near the end of life -Those caring for this population should raise awareness about the potential	- Qualitative examination of the needs of these families in hospice care -Examine the implementation of concurrent care at the clinical level -Examine this population with an earlier dataset

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			mental/behaviorual disorders were positively associated	benefit of concurrent care when appropriate	
Kieran et al. (2019)	-Retrospective Cohort Design -Original Research Article	-To describe the clinical profile, resource use, prevalence, and both in-hospital and post-discharge outcomes of neonates with complex medical needs and to assess the feasibility of sustaining the use of the neonatal complex care team	-Major underlying condition include genetic abnormalities, extreme prematurity, neurological abnormality, and congenital anomalies -Interventions included mechanical ventilation, parenteral nutrition, and technology dependence on discharge. -During 1st year of discharge, emergency visits were 44% and inpatient admission were 58%	-Findings can help clinicians objectively prognosticate mortality and re-hospitalization rates when caring for this population -Provides evidence that some elements of feasibility of implementing an innovative model of care, where neonates with complex care needs are managed primarily with complex care team -Findings can be used for counseling parents, health resource planning, and targeting care coordination efforts	-Future models of care should incorporate cost-effective analyses and impact on outcomes as part of the evaluation
Kim et al. (2017)	- Retrospective case series -Original Research Article	- To investigate the scale, time trends, disease composition, regional distribution, and unmet needs of children dying from CCCs	- 1/3 of pediatric deaths were observed in children with CCCs - Could have benefited from pediatric palliative care	- None specifically noted	- Identify the prevalence, incidence, and healthcare utilization and needs, of families with children with CCCs

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Kim et al. (2018)	-Retrospective Cohort Design -Original Research Article	- To investigate the scale, time trends, disease composition, regional distribution, and unmet needs of children dying from CCCs	-34% of deaths were due to complex chronic conditions -Pediatric deaths due to CCCs has declined from 2005 to 2014 -Overall, malignancy was the most common cause of death, but in infants, it was neonatal disorders	-No institutes in Korea meet the minimum standards for specialized pediatric palliative care, hospital might plan to expand their programs -Findings provide the fundamental characteristics of the population potentially in need of pediatric palliative care -Resources to conduct proper services in accordance with Korean culture need to be made more available -Health authorities must consider supporting the establishment of specialized palliative care centres in regional tertiary teaching hospitals -Benefits of early implementation of pediatric palliative care for children with CCCs	-Investigate the other factors that affect how patients suffering from CCCs and their families choose the place of death -Explore and develop an appropriate system of pediatric palliative care for Korean families and educate medical personnel based on their needs -Identify the prevalence, incidence, and health care utilization, as well as the pediatric palliative care service needs, of patients and their families
Kingsnorth et al. (2015)	- Descriptive mixed methods design - Original Research Article	- To explore the implementation process of the Integrated Complex Care Model: a voluntary partnership between acute, rehabilitative and community care aimed at system integration through a	-Enablers included leadership and electronic care plan use and communication -Barriers included assumptions about partner organizations,	- Engagement of primary care is critical in moving forward with an integrated model of care - Inter-organizational teams - Implementation of an accessible system-wide	- Examine how engaged organizations functioned internally

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		key worker model to improve care co-ordination for CMC	differing organizational structures and client information systems, limited engagement of primary care -Parents perceived an electronic care plan as a facilitator	information management solution - Implementation of stakeholder engagement strategies - Ensure adequate resourcing of key worker time	
Lacerda et al. (2014)	-Retrospective Cohort Design -Abstract	- To examine the epidemiology of dying for children and adolescents in PT, focusing on deaths due to CCCs, to understand trends in place of death and determine factors associated with home death	-Pediatric deaths due to CCC have increased from 1/4 to 1/3 of all deaths in the last 25yrs. -The home death proportion remains low, with a long-term trend of death away from home	-Regional, seasonal, and age need to be considered in the development of policy and services to facilitate home deaths for families who wish so.	- None noted
Lacerda et al. (2019)	-Observational, Longitudinal, Retrospective Design -Original Research Paper	- To evaluate hospital inpatient care in the National Health Service (mainland Portugal) by pediatric patients (0 – 17 years) with CCCs	-15% contained at least one CCC code - CCCs represented ~30% of hospital days, ~40% of costs, and 87% of deaths. -Malignancy was the most frequency category, neonatal had highest median length of stay, median	-Quality improvement programs could lead to a reduction of inpatient episodes, length of stay and costs, as well as to an improvement of the clinical outcomes and family satisfaction -Educating clinicians is critical -Integration of pediatric palliative care programs, involving multidisciplinary teams and providing guideline	-Evaluate the increasing trends in inpatient, length of stay, and cost

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			expense, and median number of deaths	and/or healthcare both to inpatient and outpatients -The welfare of these families should be a concern of the whole society -These patients should be a major target for the development of reforms aimed at care coordination and timely planning of care -The development of patient-oriented health care models and family well-being is an obligation of pediatrics and a reflection on the user's needs is mandatory	
Lindley at al. (2019)	-Retrospective Nonexperimental Design -Original Research Article	- To examine-the prevalence of CCC and investigate the infant characteristics related to a CCC classification	-40% were classified as having a CCC -African Americans had lower odds of being classified with a CCC -Those with health insurance were less likely to have a CCC -Nonurban residence increased the odds of having a CCC	-It is important to pay particular attention to disparities in care, especially in regard to ethnicity/race	-Validate and compare to two version of Feudtner's classification system -Researchers need to be vigilant about disparities inherent in measures used for studies -Examine the relationship between race and geographic location among children at end of life -Include infants in pediatric end-of-life research specifically by: extending

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					length of stay; including diagnosis of prematurity at any time during the hospital stay and; high technology is a sign of ongoing complexity
Lindley et al. (2013)	-Retrospective Cohort Design -Abstract	- To understand the demographic and health characteristics, health care utilization, and expenditures among Medicaid children with CCCs at end of life	- Neuromuscular conditions were most common - >50% of the children had two or more CCCs - Only 25% of children in the study accessed care designed to meet the physical and psychosocial needs	- Support current policy aimed to improve access to and ultimately quality of EOL care by eliminating a criterion for hospice - Change in federal law to allow concurrent curative care may be an important health care reform policy initiative aimed at improving the utilization and quality of care	- Explore and clarify the influence of child demographic and health characteristics on end of life care utilization -Future research might compare the influence of health care reform provisions on access to hospice and home health care using longitudinal data
Lindley et al. (2016)	-Pooled Cross-Sectional Design -Original Research Article	- To understand the clusters of CCCs present among children in the last year of life	-Four latent classes were yielded: medically fragile (31%); neurological (32%); cancer (25%); and cardiovascular (12%) -Three classes were characterized by a 100% likelihood of having a CCC coupled with a low or moderate	-Additional private insurance may mitigate the financial burden these families encounter by covering medical services, medical transportation, equipment and supplies, and treatments not covered or fully covered by Medicaid -End-of-life care for children that targets specific clusters	-Examine the impact of the primary diagnosis on health state and health resource use by using other data source such as death certifications and/or electronic records -Compare the quality of care delivered to these four classes for children by specialists and pediatricians

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			likelihood of having the other eight conditions	should perhaps consider different patterns of care	
Lindley et al. (2020)	-Retrospective -Nonexperimental Design -Original Research Article	- To assess the agreement and validity of the original and the modified complex CCC system	- The modified CCC classification system was sufficiently different from the original system - The Kappa Statistic (k = 0.125) suggested only a slight agreement between the original and modified systems	-None noted	-Explore at a conceptual level to understand why differences in identification exist between the modified and original classifications -Additional testing for end-of-life outcomes and service referrals -Additional testing to determine the magnitude of the difference
Lindley et al. (2021)	-Retrospective Nonexperimental Design/Validation -Original Research Article	-To compare the predictive performance of the original versus the revised CCC classification measure among infants	-Both measures poorly discriminated palliative care consultation and any inpatient procedure, however the original measure was more accurate -Good agreement between outcomes for both measures	-None noted	-Explore prospective design that captures diagnoses in real time and as they change -No clear advantage to using either classification system -Chose the first or second based on your conceptualization of CCCs (original aligns better with life-limiting health conditions; revised aligns best with medical complexity) -Include infants in nursing research

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Maenge et al. (2017)	- Quality Improvement -Original Research Article	- To examine the complex care clinic impact on total cost of care and utilization by analyzing Geisinger Health Plan claims data obtained from 83 Medicaid patients (medically complex Adolescents and young adults with special care and health needs) enrolled in complex care clinic	- Reduced total medical care costs and acute care utilization	- Result show key features of a complex care management model to meet the needs of this population	- Use family-centered outcomes
Marshall et al. (2017)	- Retrospective Cohort Design -Abstract	- To examine the prescription utilization and cost patterns of children with special healthcare needs having CCCs	- The ten most expensive drugs to all exceed a daily dispense cost of $1,450	-None noted	- Compare differences in prescribing trends and costs between the pediatrician-led model and usual care model
Moura et al. (2017)	- Retrospective Cohort Design -Original Research Paper	- To estimate the extent of hospitalizations for CCCs in Brazil	- Incidence rate of hospitalizations of 331 per 100,000 inhabitants	- Results support decisions on resource allocation	- Identify the dimension of children with CCCs in Brazil - Compare the evolution throughout the time
Murtagh et al. (2014)	- Retrospective Cohort Design -Original Research Paper	- To generate nationwide estimates for ED use by children and young adults with CCCs and to evaluate if being of the age for transition to adult care significantly affects	- Majority of visits are to general EDs - No difference between rate of admission or transfer and total charges by site of care	- None noted	- Develop and validate measures for the quality of emergency care delivered to medically complex children and young adults across

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		the site of care and likelihood of hospital admission			both pediatric and general EDs
Neff et al (2016)	- Retrospective Cohort Design - Original Research Article	- To determine the rates of central line-associated blood stream infections in 3 similar children's hospitals according to patient complexity groups by using the 3M Health Information Systems' clinical risk groups (CRGs)	- Variation in rates based off clinical risk group status:-0.008% in nonchronic, 1.604% in progressive chronic or dependent on technology, 1.162% in malignancies, and 0.625% in complex chronic	-Hospitals reports need to be adequately risk adjusted to control for patient characteristics	-Include a broader sample -Compare age stratification (less than 1, and greater than 1) at various level of infant/NICU admission -Compare chronic condition risk adjustment tool and age stratification to line days and admission to intensive care units in a large multihospital dataset
Neff et al. (2015)	- Development and testing of algorithm - Original Research Article	- To stratify children using available software, Clinical Risk Groups, in a tertiary children's hospital, Seattle Children's Hospital, and a state's Medicaid claims data, Washington State, into 3 condition groups: complex chronic disease; noncomplex chronic disease, and nonchronic disease	- Good to excellent specificity and sensitivity classifying children in the groups CCCs and no condition -Very good specificity in identifying children with noncomplex chronic condition -Poor sensitivity in classifying children with noncomplex chronic condition	- Further severity gradation might help in developing tiered levels of payment and targeted care coordination	- Improve performance in hospital data, CRGs should be limited to those with inpatient encounters

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O'Mahony et al. (2013)	- Retrospective Cohort Design -Original Research Paper	- To characterize the use of and disposition from a tertiary pediatric ED by children with chronic conditions with varying degrees of medical complexity	- Comprise 20% of pediatric ED visits - With increasing medical complexity, the rates of both hospital and PICU admission, pediatric ED LOS, and repeat ED visits all increased	- EDs must be prepared to care for children with chronic medical complexity - Interventions such as individual pre-arrival identification, the use of individual emergency care plans, and expedited admissions processes may be of benefit	- None noted
Parente et al. (2021)	-Non-experimental descriptive design -Original Research Paper	- To describe a point-of-care screening algorithm to identify CMC with high health care use, a group that may benefit the most from improved care coordination	-The screening algorithm is a feasible measure to prospectively identify CMC - CMC had greater per-patient and per-encounter hospital use than other children	-The screener tool offered an efficient approach to identify CMC in both inpatient and outpatient settings that may benefit from enhanced care coordination via a complex care program	-Did not validate the screener tool since no gold standard from CMC exists -More comprehensive datasets may be able to detect difference in use for children with greater than 4 complex chronic conditions
Peter et al. (2011)	Pre/post clinical cohort evaluation -Original Research Paper	- To evaluate an ambulatory care coordination program for children with complex care needs	- Significant reductions in hospital utilization with subsequent cost savings	- Implement a systems approach to care - Coordination approaches provided by a team of nurses integrating specialist care and primary care -Pediatric nurses can be influential in advocating for such programs where	- Examine family perspectives

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				there is an identified gap in service -A 24-hour telephone support and the familiarity of the nurses with their children's conditions	
Phillips et al. (2019)	-Retrospective Cohort Design -Original Research Paper	-To investigate, for children and youth, the effects of chronic disease on the likelihood of ambulatory care–sensitive conditions	-Approximately 18% of discharges had one CCC -The presence of any CCCs was protective against ambulatory care-sensitive conditions hospitalizations for a non-chronic condition	-The role of chronic disease in children/youth differs from that in an adult or older persons -Findings are most likely a result of more intense primary care for those with CCCs	-Further analyses with alternative databases should better clarify the dynamics underlying these results
Pulcini et al. (2021)	-Retrospective Cohort Design -Original Research Article	- To assess the quantity and characteristics of low-resource ED visits in children with a complex chronic condition	-16% of ED visits were low-resource -Highest odds for experiencing a low ED visit was those 0 year, living <5 miles away, during the day/evening	-Health system initiatives could focus on curtailing ED use in the most at risk children identified by this study	-Explore factors related to design processes and systems that will best address the urgent health care needs of children with CCCs -Explore how ED clinicians perceive and use hospitalization history in children with CCCs -Explore pre-ED health care use to understand illness trajectories and describe

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					patterns of health service use -Assess opportunities for diversion of ED care to outpatient and community settings -Explore health resource use at community Eds
Ralston et al. (2015)	- Retrospective Cohort Design -Original Research Paper	- To identify and characterize CMC from the resident pediatric population of Maine, Vermont, and New Hampshire and present utilization rates for children receiving care in 4 academic children's hospitals and 2 regional hospitals	- Hospital-specific resource utilization exhibited a high degree of variation, particularly around high cost, discretionary services	- Identify best practices for this growing patient population	- Evaluate directed management programs
Rasooly et al. (2020)	-Retrospective Cohort Design -Original Research Article	- To quantify state-to-state variability in distribution of posthospitalization home nursing to commercially insured CMC and to rank-order states	- ~10% of the sample received home nursing - Using home care varied across regions -Adjusted median home nursing days ranged from 6.6-24.5 days	- Development of standards and policies to providing home nursing care to reduce disparities in who receives it and how much they receive - May be opportunity to provide more support for families	-Evaluate potential drivers of the variation among states and over time
Shumskiy et al. (2018)	-Retrospective Cohort Design -Original Research Article	- To determine the number of years with ≥1 well child visits for Medicaid-insured CMC between 2010 and 2014 and to assess correlations between	- Small numbers underwent consistent well child visits over time - Modestly higher odds of acute unplanned	-Primary health care providers may not feel equipped to provide well child visits -State Medicaid programs could use the findings to develop policies and programs that may	- Identify reasons for underutilization - Identify reasons for consistent well child visits are associated with fewer hospitalizations

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
		years of well-child visits with hospitalizations for CMC	hospitalization were noted among children without consistent visits	enhance primary care services - Through increased awareness of underutilization of well child visits, pediatric primary care practices may be prompted to monitor use proactively promote and enable children who are underusing them	- American Academy of Pediatrics could consider surveying primary care pediatricians to better understand their perceptions
Silber et al. (2018)	- Retrospective Matched Cohort Design - Original Research Article	- To determine whether style-of-practice differs between similar Medicaid and Non-Medicaid children with CCCs undergoing surgery	- Small differences in the practice style - Small pattern indicating higher resource use and in-hospital mortality	- None noted	- Monitor both Children's and non-Children's Hospitals with respect to practice style and mortality differences across Medicaid and Non-Medicaid patients.
Silber et al. (2019)	-Retrospective Cohort Design -Original Research Article	- To examine variation in resource utilization by the hospital for children with medically diverse CCCs admitted for 40 different principal diagnoses	-Intensive care unit utilization was 111% higher; hospital length of stay was 25% higher, and cost per patient varied by 47%	-This system provides a bench marking system of resource use -Allows hospitals to now focus on patient groups along the risk continuum needing improvement	-As data system improve, the use of a Template may provide benefits for those seeking to improve care
Silverman et al. (2016)	- Retrospective Cohort Design -Abstract	- To estimate the prevalence of CMC admitted to USA pediatric/infant intensive care units and describe clinical characteristics, and estimate bed growth over the past 5 years	- PICU bed growth increased by 2.7%, visits increase 18.2% in those with an intermediate care unit and 13.3% among PICU-only hospitals	- Increase in hospitalizations over the study period, far outpacing bed growth -Children with CCCs accounted for most of the visits in both hospital cohorts	- Assess the effect of infant critical care unit on pediatric ward admissions for children with CCCs

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
			-Children with chronic conditions accounted for the majority of PICU visits		
Simon et al. (2010)	- Retrospective Cohort Design -Original Research Paper	- To determine whether the proportion of pediatric inpatient use that is attributable to patients with a diagnosis of one or more has increased overtime and to assess the degree to which complex chronic condition hospitalizations are associated with attributes that are consistent with heightened medical complexity	- The proportion of inpatient pediatric admissions, days, and charges increased over the study period -Admissions used 22.7% to 26.1% of pediatric hospital days, used 37.1 to 40.6% of pediatric hospital charges, accounted for 41.9% to 43.2% of deaths, and 73% to 92% of different forms of technology-assistance procedures	- CCCs are an increasing proportion of inpatient care and resource use	- Improve methods to identify medically complex children, monitor the trends of increasing use, and assess whether current systems of care are meeting their needs
Simon et al. (2014)	- Retrospective Observation Study - Development and validation study -Original Research Article	- To develop an algorithm based on International Classification of Diseases, Ninth Revision, Clinical Modification (ICD-9-CM), codes for classifying children with chronic disease according to level of medical complexity and to	- Good sensitivity for correctly categorizing children with CCCs - Excellent sensitivity for correctly categorizing children without chronic disorders	- Use of PMCA will be critical to target resources and services such as care coordination to children with the most needs	- Validate in other populations and contexts

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
		assess the algorithm's sensitivity and specificity	-Poor sensitivity for correctly categorizing children with non-complex chronic condition		
Simon et al. (2017)	- Development and Validation Study - Original Research Article	- To refine Pediatric Medical Complexity Algorithm and evaluate its performance based on the duration of eligibility and completeness of Medicaid data	-Resulted in improved face validity, but minimal improvement in performance compared with PMCA version 1.0	- Use of PMCA will help address the legislative mandate to assess disparities by special health care need status	- Validate in other populations and contexts
Simon et al. (2018)	- Development and Validation Study - Original Research Article	-To modify the Pediatric Medical Complexity Algorithm (to include both International Classification of Diseases, Ninth and Tenth Revisions, Clinical Modification codes for classifying children with chronic disease by level of medical complexity and to assess the sensitivity and specificity of the new version 3.0 for correctly identifying level of medical complexity	- Includes both ICD-9 and ICD-10 codes - Very good sensitivity for correctly categorizing children with CCCs - Good sensitivity for correctly categorizing children without chronic conditions - Poor sensitivity for correctly categorizing children with noncomplex chronic conditions	- Use of PMCA will help address the legislative mandate to assess disparities by special health care need status	- Validate in other populations and contexts - Test the hypothesis that children with complex chronic disease experience poorer quality of care

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Simon et al. (2017)	- Cluster Randomized Controlled Trial - Original Research Article	- To assess whether CMC exposed to a hospital based comprehensive case management service experience improved health care quality, improved functional status, reduced hospital-based utilization, and/or reduced overall health care costs	- Improved quality of care, did not change functional status or hospital-based utilization, and increased overall health care costs	- Tiered care providing care coordination services commensurate with disease severity may be a more cost-effective approach for CMC	- Focus on refining (1) the population of CMC who most benefit from intervention(s), (2) the intervention approach, (3) the intensity and duration of intervention needed, and (4) outcomes to include not only utilization and cost but also similar quality of care measures
Srivastava et al. (2016)	- Retrospective Cohort Design -Original Research Paper	- To describe the hospital costs, hospital types and differences across states and territories for CMC cared for in Australian public hospitals	- Small proportion of total childhood hospitalizations - Account for almost 1/3 of total hospital costs of all children. -Children's hospitals account for approximately 1/5 of the total hospital costs for all children	- Create linked Australian databases to track individual patients across the continuum of care in order to better understand outcomes of care	-Identify types of CMC and reasons for admission between children's and non-children's hospitals - Compare the current paediatric costing models in to those that include children with medical complexity - Measure the variation in costs of select medical and surgical pediatric conditions across hospital types - Care quality provided must be measured for a more informed health policy

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Stephens et al. (2017)	-Retrospective Cohort Design -Original Research Paper	- To examine the prevalence of diagnosis and treatment for constipation among children receiving Medicaid and to compare healthcare utilization and spending for constipation among children based on number of CCCs	- Approximately 6.8% of children in the study have a CCC and account for 33.5% of total constipation spending, 70.3% of inpatient constipation spending, and 19.8% of ED constipation spending - Constipation prevalence was 11.0% for children with 1 CCC, 16.6% with 2 CCCs, and 27.1% with 3 CCCs	-May be an opportunity to reduce spending on unnecessary ED visits for constipation through improved outpatient and community care management for children in Medicaid -Reduction of x-ray use in the ED for constipation may represent an area for potential practice improvement -Better constipation screening and treatment in the primary care setting -May have a significant effect on the health of children with CCCs	-Prospective examination of risk factors for admission for constipation and hospital-level variation in practice on treatment of constipation -Understand which patient necessitate admission for constipation treatment
Van Doren et al. (2015)	- Retrospective Cohort Design - Original Research Paper	- To characterize the frequency, cost, and hospital reported outcomes of cachexia and debility in children and adolescents with CCCs	- Cachexia was more frequently listed on discharge summaries for pediatric CCCs hospitalizations than debility. - Cachexia and debility were listed together only rarely as comorbidities	- Results can aid in the development of treatments for these conditions and assist in identification of cachexia and debility at earlier stages	- Confirm the burden of cachexia and debility

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
Verlaat et al. (2019)	- Retrospective Cohort Design - Original Research Article	-To determine if CCCs or other factors are associated with death in high-risk PICU-patients.	- No association was found between CCCs and non-survival - Lower Glasgow coma scores at admission was associated with lower mortality	-None noted	-Further explore the role of CCCs in PICU patients with different risk profiles
Walter et al. (2018)	- Retrospective case series - Original Research Article	-To examined issues for families with CMC enrolled in USA private health plans	- Children with CCCs had higher health service utilization and out-of-pocket expenditures than those without any chronic condition	- Insurance with high deductibles has not provided the needed financial protection for families with children of complex health needs - Health plan benefit design be provided in plain language to meet the literacy levels in the general population - Families need to understand the implications of health plan choice on out-of-pocket expenses	- Evaluate how strategies could be implemented to alleviate such family financial and social burdens.
Wang et al. (2009)	- Retrospective cohort design - Original Research Article	-To quantify the effect of socioeconomic status on health outcomes during the first year after newborn discharge among infants with CCCs insured through a universal health plan	- There was a socioeconomic gradient in risks of mortality and hospitalization who participated in a universal health insurance system	- Important to note socioeconomic status in clinical practice	- Stratify by socioeconomic status further investigations

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
White et al. (2020)	- Retrospective Cohort Design - Original Research Article	-To describe interfacility transfer among children with CCCs and determine if interfacility transfer was associated with health outcomes	- Of those with a CCC code, 13% had an interfacility transfer -Those who were transferred in and ultimately discharged from the from the receiving hospital had an adjusted length of stay rate ratio of 1.6, were more likely to received critical care services, and had a higher in-hospital mortality	-Children with CCs undergoing interfacility transfer is a particularly vulnerable subpopulation among pediatric patients who are hospitalized -Need to better equip non-children's hospital to care for children with CCCs -Targeted efforts to coordinate transitions of care, including interfacility transfers, specifically for patients age <1 year with CCCs may be particularly effective at improving outcomes and health care use among patients with CCCs -Systematic improvements in the pediatric transfer process more than most pediatric subgroups	-Explore the role of hospital proximity, parent decision-marking, and disease severity on presentation -Assessment of the role racial, ethnic, and payer-source factors have on interfacility transfer to better design intervention that improves health equity -Prospective assessment using multidisciplinary clinical data across a diverse cohort to inform intervention to improve outcomes
Yamada et al. (2020)	- Retrospective Design - Original Research Paper	-To investigate the prevalence and background of CMC and its secular trend in Japan.	-During the study period, prevalence increased 1.9 times from 0.98 to 1.88 per 1000 population -The number of individuals living at home increased,	-The number of children who need follow-up cardiac is likely to increase -Tube feeding is expected to increase the survival rate -It is necessary to provide medical care, as well as collaborate with the welfare	-Future investigations are needed as prognosis of disease will change over time -Look at underlying disorders, medical care, and devices to design individualized services and

Author/ Year	Study Design/ Paper Type	Aim/Objectives	Results	Practice/Policy Recommendations	Research Recommendations
			accounting for 90% of cases -The number of children with congenital, perinatal, and cardiac disorders increased	and education to share the accurate needs of CMC	education according to severity of CMC -Establish a common definition to perform prospective multifaceted evaluation -Future studies must provide appropriate individualized medical, welfare, administrative, and education about the different types of CMC
Zurynski et al. (2019)	- Pre and Post Implementation Cohort Design - Abstract	-Evaluated the impacts of Care Coordination on tertiary hospital service use and family outcomes for children with medical complexity	-557 hospital encounters were prevented in 6 months -ED visits decreased by 40% -Day-only-admissions decreased by 42% - Over 2 years, an estimated AU$4.9 million was saved for the pediatric network, out of pocket costs to families were reduced by AU $586,644, reduced over 50,000 kms of travel, and 370 school absences were prevented	-The model has clear benefits for pediatric hospital networks and families	-Ongoing evaluation needed -Better engagement with the primary care sector

Table 2-3. Terminology, Definitions, and Methods of Identification to Children with Medical Complexity

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Amin et al.	2018	Canada	**Children with chronic conditions:** a condition that lasts more than 12 months, involves 1 or several organ systems, and requires a high level of specialty care	- 0-18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9) - Further categorized into having single or multiple system involvement	None
Ananth et al.	2015	USA	**Life-threating complex chronic conditions:** conditions described as those that last at least 1 year, are severe enough to necessitate subspecialty pediatric care, and bear the highest risk of mortality out of all chronic conditions in childhood.	- 1-18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Arthur et al.	2018	USA	**Children with medical complexity:** no definition provided	- 0-17 years old - Pediatric Medical Complexity Algorithm (ICD-9) - Had at least 4 primary care visits -Had a health care provider listed among participating pediatricians in the Medicaid program	None
Avritscher et al.	2019	USA	**High risk, chronically ill, children with medical complexity:** significant chronic conditions that require care from a multitude of providers in different settings	-Had ≥2 hospitalizations, or ≥1 PICU admission during the year before enrollment and >50% estimated risk of hospitalization	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Bennett et al.	2020	USA	**Children with complex chronic conditions**: as those medical conditions expected to last longer than 12 months and involve either several organ systems or one organ system severely enough to require recurrent readmissions to children's hospitals	-0-18 years old -Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Bergman et al.	2020	USA	**Children with medical complexity:** medical fragility, substantial functional limitations, increased need for health care services, and increased health care costs	- 0-21 years old - Identified at each participating hospital site using their own eligibility criteria were prospectively enrolled if their diagnosis were compatible with 3M Clinical Risk Groups categories 5b through 9 (based on low enrolled, ended up excluded 19-21 year olds from this analysis)	None
Berry et al.	2014	USA	**Children with medical complexity:** Expensive, complex, and chronic conditions expected to last longer than 1 year that often lead to functional limitations, which as often severe; substantial health services needs to maintain health, including numerous clinicians, medications, equipment, therapies, and surgeries; and high resource utilization; and high morbidity and mortality	- 0-18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Berry et al.	2011	USA	**Children with medical complexity:** Complicated, fragile chronic disease or multiple chronic medical problems that lead clinicians to consider them medically complex; require high intensity coordination care from primary, community and multiple-specialty providers	- At least one health encounter with a structured complex-care clinical program during their life - One or more hospitalization between July 2006-2008 - Individual diagnosis codes were categorized using Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Berry et al.	2017	USA	**Children with Multiple Chronic Conditions**: a chronic condition is defined as a condition that lasts 12 months or longer and has one or both of the following effects it places limitations on self-care, independent living, and social interaction and/or it results in the need for ongoing intervention with medical products, services, and special equipment	- Agency for Healthcare Research and Quality Chronic Condition Indicator (CCI)	None
Berry et al.	2013	USA	**Children with chronic conditions:** affecting 2 or more body systems and/or a complex or progressive chronic condition	- Clinical Risk Groups v 1.7	None
Berry, Glotzbecker, et al.	2017	USA	**Children with medical complexity:** no definition provided	- 5-18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Bjur et al.	2019	USA	**Children with multiple complex chronic conditions:** any child with >2 complex chronic condition categories previously defined by Feudtner et al.	-0-17 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Brittan et al.	2015	USA	**Children with chronic conditions:** presence of chronic medical conditions, often in comorbid combination, that are expected to last at least 12 months and are likely to lead to inpatient utilization	- 6 months - 18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Brown et al.	2021	USA	**Children with medical complexity/complex chronic conditions**: are defined as medical conditions expected to last at least 12 months, involve several organ systems or 1 system severely enough to require specialty pediatric care, and have a high probability of hospitalization	- Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Burns et al.	2010	USA	**Multiple chronic conditions**: (1) diagnoses in more than 1 chronic condition category, defined by organ system, without requiring the presence of any single specific condition or (2) children with a single specific diagnosis, CP or bronchopulmonary dysplasia (BPD), and the presence of a diagnosis in 1 of the chronic-condition categories	- Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Buser et al.	2020	USA	**Children with medical complexity:** children with complex chronic conditions in need of frequent health care visits. This includes all children and adolescents with serious chronic conditions, substantial	-Those with frequent visits: as in >10 visits in 24 months. Because some children had only recently arrived, the criteria of ≥1.5 visits per month with at	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			functional limitations, increased health and other service needs and increased health care costs	least 5 visits or 7 cumulative days of hospital admission was also included	
Carrilero et al.	2020	Spain	**Children with medical complexity:** complex acute and chronic conditions, numerous and varied comorbidities, a broad range of mental health and psychosocial needs, major functional limitations, and a higher rate of mortality	- >15 years old - Adjusted Morbidity Groups classification system (Catalan acronym GMA)	None
Casey et al.	2011	USA	**Multiple chronic conditions:** least 2 serious chronic conditions and were followed up by at least 2 pediatric subspecialists	- Those enrolled in a clinic program and linkable with Medicaid data	None
Chan et al.	2016	USA	**Children with complex chronic conditions**: those with either chronic conditions in ≥2 organ systems, a progressive condition associated with decreased life expectancy, malignancy, or continuous technology dependence.	- <19 years old - Pediatric Medical Complexity Algorithm	None
CIHI	2020	Canada	**Children with medical complexity:** share 4 characteristics: complex chronic conditions, functional limitations, high health care utilization and a high need for caregiving	-0-24 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-10)	Noned
Cohen et al.	2012	Canada	**Children with medical complexity:** complex underlying chronic health conditions that are typically associated	- ICD-10 diagnostic codes within 3 clinical categories of neurological Impairment, complex chronic conditions, and technological assistance	Cohen's conceptual framework of children with medical complexity

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			with significant functional status limitations		
Cohen et al.	2010	Canada	**Medically complex and fragile children (MCFC):** a severely affected subset of children and youth with special health care needs who problems are multiple in terms of health/developmental needs, services, sectors of care, and locations of care.	- >18 years old -Caregivers were referred to the clinic by attending hospital-based pediatricians on inpatient general wards or ambulatory clinics	-Evaluation of the program was based on the conceptual framework of the six domains of high-quality care by the Institutes of Medicine
Cohen, Lamcombe-Duncan, et al.	2012	Canada	**Children with medical complexity:** substantial family-identified needs, characteristic complex and/or chronic conditions, functional limitations, and high health care use	-Those enrolled in the clinic program - Diagnoses were categorized into Feudtner et al.'s Complex Chronic Conditions Classification, Srivastava's definition of neurological impairment, and technology assistance	None
Coller et al.	2018	USA	**Children with medical complexity:** severe chronic conditions, major functional limitations, high health resources utilization, and substantial health service needs	- <21 years old - Pediatric Medical Complexity Algorithm	None
Coller et al.	2019	USA	**Children with medical complexity**: lifelong, complex chronic conditions associated with multi-morbidity, severe functional limitations, myriad health care needs, and high resource use **Complex Chronic Conditions:** diagnosis groupings expected to last >12 months and involved either a single organ system severely enough to require specialty	- 0-19 years old - Feudtner et al.'s Complex Chronic Conditions Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			pediatric care and hospitalization, or multi organ systems.		
Coller Klitzner et al.	2018	USA	**Children with medical complexity:** no definition provided	- <18 years old - Enrolled in the Pediatric Medical Program - Categorized based on Feudtner et al.'s Complex Chronic Conditions Classification	None
Coquillette et al.	2015	USA	**Children with medical complexity:** chronic conditions that result in significant functional limitations and/or dependence on medical technology and who require high levels of home-based and hospital-based services	- Existing registry of medically-complex patients developed by the hospital	None
Edwards et al.	2010	USA	**Children with complex chronic conditions:** no definition provided	- Modified Delphi Methods	None
Edwards et al.	2012	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- A modified version of the Feudtner et al.'s Complex Chronic Conditions Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Feinstein et al.	2014	USA	**Children with complex chronic conditions:** who have a chronic or life span-shortening disease process, require lifelong medical care, and often rely on supportive technology (such as tracheostomy, ventilator, or gastrostomy tube) and multiple prescription medications	- Feudtner et al.'s Complex Chronic Condition Classification - Excluded malignancies	None
Feudtner et al.	2000	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- Feudtner et al.'s Complex Chronic Condition Classification	None
Feudtner et al.	2002	USA	**Children with complex chronic conditions:** no definition provided	- Feudtner et al.'s Complex Chronic Condition Classification	None
Feudtner et al.	2003	USA	**Children with complex chronic conditions:** as any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- >25 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Feudtner et al.	2007	USA	**Children with complex chronic conditions:** medical conditions that can be reasonably expected to last at least 12 months (unless death intervenes) and that involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- >20 years - Feudtner et al.'s Complex Chronic Condition Classification	None
Feudtner et al.	2001	USA	**Children with complex chronic conditions:** no definition provided	- >24 years - Feudtner et al.'s Complex Chronic Condition Classification	None
Feudtner et al.	2014	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- Feudtner et al.'s Complex Chronic Condition Classification V1 and V2	The conceptual definition provided by Feudtner
Feudtner Silveria et al.	2002	USA	**Children with complex chronic conditions:** any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and	- >25 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			probably some period of hospitalization in a tertiary care center		
Forjaz de Lacerda et al.	2017	Portugal	**Children with complex chronic conditions** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center.	- >17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Friedel et al.	2019	Belgium	**A complex chronic condition:** can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either different organ systems or one organ system severely enough to require specialty paediatric care and probably some period of hospitalisation in a tertiary care centre	- 0-19years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Gay et al.	2016	USA	**Children with medical complexity:** have complex chronic conditions that that are often associated with significant functional impairment, myriad healthcare needs, and high resource use; conditions expected to last >12 months and involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and hospitalization. 3 CCCs include medical technology to maintain a child's health status	- 0 -17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Geurtin et al.	2006	Canada	**Children with complex chronic conditions:** no definition provided	- 1-19 years old - unknown	None
Geurtin et al.	2009	Canada	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center.	- 1-19 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Gold et al.	2016	USA	**Children with medical complexity:** have a complex and chronic health conditions that often involve multiple organ systems and severely affect cognitive and physical functioning	- Clinical Risk Groups	none
Gordon et al.	2007	USA	**Medically complex and fragile CYSHCN** : complex chronic conditions that involve several organ systems and require multiple specialists, technological supports, and community services	- Those enrolled in the special needs program	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Håkanson et al.	2017	Belgium, Canada, the Czech Republic, France, Italy, Korea, Mexico, New Zealand, Spain (Andalusia), Sweden, & the USA	**Children with complex chronic conditions:** no definition provided	- 1-17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Hannan et al.	2021	USA	**Children with medical complexity:** assessment of high medical and family needs, functional limitations, high health care resource use, and underlying chronic conditions that are severe and/or associated with medical fragility	- Less than a year old - Feudtner et al.'s Complex Chronic Condition Classification	-None
Jamorabo et al.	2015	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 1-17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Johnston et al.	2019	USA	**Children with complex chronic conditions:** involve different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 1 – 21 years old - ICD-9 or International Classification of Diseases, 10th Revision code for a CCC	None
Jurgens et al.	2014	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- >18 years - Feudtner et al.'s Complex Chronic Condition Classification	None
Kalzen et al.	2018	Sweden	**Children with complex chronic conditions:** no definition provided	-0-16 years old - Feudtner et al.'s Complex Chronic Condition Classification (v2)	None
Keim-Malpass et al.	2020	USA	**Medically complex children:** multi-organ system problems, chronic childhood conditions, limitations in functional status, and ongoing use of medical technology, along with extremely high health care resource use and substantial needs	- >21 years old - Feudtner et al.'s Complex Chronic Condition Classification was used to create a measure for technology dependence; complex chronic condition; multiple complex chronic conditions; mental/behavioral disorders	-Following Kuo et al.'s definition for medical complexity

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Kieran et al.	2019	Canada	**Children with complex medical needs:** complex chronic conditions requiring specialized care, substantial healthcare needs, functional limitations, high use of health resources, and hospitalizations	-Neonates -Underlying diagnoses contributing to having complex medical needs was establish a priori by consensus among the research team members -Five broad categories: prematurity of less than 29 weeks and its complications; neurological problems; congenital anomalies; genetic or chromosomal disorders; other.	None
Kim et al.	2017	Korea	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- >20 years old - Feudtner et al.'s Complex Chronic Condition Classification: Modified from ICD-10 codes to KCD codes	None
Kim et al.	2018	Korea	**Complex chronic conditions:** any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty	- >1-19 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			pediatric care, and probably some period of hospitalization in a tertiary care center		
Kingsnorth et al.	2015	Canada	**Children with medical complexity:** depend on medical technology and highly specialized care to optimize their quality of life and universally require multiple services from multiple sectors, in multiple locations	- >18 years old - Identified by health care providers perceived to be complex with at least one criterion met from each of the following categories: technology dependent and/or uses high intensity care, fragility, chronicity and complexity	None
Lacerda et al.	2014	Portugal	**Children with complex chronic conditions:** no definition provided	- 0-17 years old - unknown	None
Lacerda et al.	2019	Portugal	**Complex chronic conditions:** any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 0-17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Lindley et al	2019	USA	**Children with complex chronic conditions:** No definition provided	- > 1 year - Feudtner et al.'s Complex Chronic Condition Classification	None
Lindley et al.	2013	USA	**Children with complex chronic conditions:** no definition provided	- >20 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Lindley et al.	2016	USA	**Children with multiple complex chronic conditions:** conditions that are reasonably expected to last at least 12 months and to involve either several different organ systems or one organ system severely	- 0-20 years old - Feudtner et al.'s Complex Chronic Condition Classification	The conceptual relationships among the complex chronic conditions, the latent classes, and the covariate predictors for the latent class model estimated in these analyses
Lindley et al.	2021	USA	**Children with complex chronic conditions:** medical conditions that would reasonably be expected to last at least 12 months and involve either several organ systems or 1 organ system requiring specialty pediatric care	- > 1 year - Feudtner et al.'s Complex Chronic Condition Classification	The first classification system is based off the conceptualization of a life-limiting illness whereas the second is based on medical complexity
Lindley et al.	2020	USA	**Children with complex chronic conditions:** medical conditions that would reasonably be expected to last at least 12 months and involve either several organ	- > 1 year old - Feudtner et al.'s Complex Chronic Condition Classification version 1 and 2 (comparison study)	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			systems or a single organ system requiring specialty pediatric care		
Maenge et al.	2017	USA	**Medically complex adolescents and young adults with special care and health needs:** no definition provided	- <15 years old - Typically use 20 or more medications - Rely heavily on 3+ specialties, 3+ times in the past 2 years, and 2+ genetic medicine visits - Be technology-dependent - Have certain diagnoses - Have a history of frequent ED visits and inpatient admissions - Referred to complex care management - Self-identified by the patient or caregiver.	None
Marshall et al.	2017	USA	**Children with complex chronic conditions:** no definition provided	- Enrolled in the Specially for Children pilot study	none
Moura et al	2017	Brazil	**Children with complex chronic conditions:** functional limitations, specialized care needs and technological dependence	- >18 years old - Feudtner et al.'s Complex Chronic Condition Classification	Cohen's conceptual framework of children with medical complexity

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Murtagh et al.	2014	USA	**Children with complex chronic conditions:** Include children with multisystem disease states, technology dependence, or complex medication regimens	- >24 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Neff et al	2016	USA	**Complex chronic conditions: h**ave significant chronic conditions in .1 body system, such as diabetes type 1 with a neurologic disorder, or developmental delay with complex seizure disorders	-Clinical Risk Groups: Study group 1: nonchronic (CRG status groups 1 and 2) • Study group 2: noncomplex chronic (CRG status groups 3–5) • Study group 3: complex chronic (CRG status groups 6 and 7) • Study group 4: malignancies (CRG status group 8) • Study group 5: progressive chronic or dependent on technology or transplantation (CRG status group 9)	None
Neff et al.	2015	USA	**Complex chronic disease:** no definition provided	- Clinical Risk Groups: complex chronic disease (C-CD), noncomplex chronic disease (NC-CD), and nonchronic disease (NC).	None
O'Mahony et al.	2013	USA	**Children with medical complexity:** lifelong multi-organ system conditions, who have increased technology dependence and are cared for by multiple subspecialists	- Clinical Risk Groups	None
Parente et al.	2021	USA	**Children with medical complexity:** clinically heterogeneous group of children characterized by a wide range of multiple chronic conditions, a need for technology	- A point-of-care complexity screening algorithm using the Children with Special Health Care Needs Screener	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
			assistance, a need for multiple subspecialists, and high resource use.		
Peter et al.	2011	Australia	**Children with complex healthcare needs**: chronic conditions that involve several organ systems and/or require multiple specialists, technological supports, and community services incur very high healthcare costs	- Those enrolled in the clinic program: (1) require care coordination and (2) frequently utilizes the hospital services (i.e., more than four ED presentations, or more than two hospital admissions, or longer than 14 days LOS within the past year, or, in the case of infants, is at risk of significant future hospital utilization)	None
Phillips et al.	2019	USA	**Complex chronic conditions**: no definition provided	->18 years old - Feudtner et al.'s Complex Chronic Conditions Classification (ICD-9)	None
Pulcini et al.	2021	USA	**Children with complex conditions:** medically fragile because of significant chronic health problems, functional limitations, intense health care needs, and high health resource use	- Feudtner et al.'s Complex Chronic Condition Classification (ICD-10)	None
Ralston et al.	2015	USA	**Children with medical complexity**: any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or one system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 30 days - 18 years old - Pediatric Medical Complexity Algorithm: further refined the chosen ICD codes based an initial review of the cohort	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Rasooly et al.	2020	USA	**Children with medical complexity:** chronic conditions, medical fragility, and substantial functional limitation. Complex chronic conditions represent defined diagnosis groupings expected to last longer than 12 months and involve either a single-organ system severe enough to require specialty pediatric care and hospitalization, or multiple-organ systems.	- 0-18 years old - Feudtner et al.'s Complex Chronic Condition Classification (ICD-10)	None
Shumskiy et al.	2018	USA	**Children with medical complexity:** myriad of healthcare needs, high healthcare resource use, and significant impairments in functioning	- 1-14 years old - Feudtner et al.'s Complex Chronic Condition Classification - Chronic Condition Indicator (CCI) system developed by the Agency for Healthcare Research and Quality - Restricted to children using ≥1 chronic medication	None
Silber et al.	2019	USA	**Children with complex chronic conditions:** no definition provided	-1-18 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Silber et al.	2018	USA	**Children with complex chronic conditions:** no definition provided	- 1-18 years old - Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Silverman et al.	2016	USA	**Children with medical complexity:** no definition provided	- Clinical Risk Groups	None
Simon et al.	2010	USA	**Children with complex chronic conditions:** Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 0-18 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Simon et al.	2017	USA	**Children with medical complexity:** no definition provided	- (1) Clinical Risk Group - (2) Identification by participating primary care providers: had to have a hospitalization or ED (ED) visit at 1 or more times in the enrollment period and have a dominant chronic condition"	None
Simon et al.	2014	USA	**Children with medical complexity:** no definition provided	- 0-18 years old - Pediatric Medical Complexity Algorithm: ICD-9 Codes	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Simon et al.	2017	USA	**Children with medical complexity:** no definition provided	- 3 months- 18 years old - Pediatric Medical Complexity Algorithm 2.0: ICD-9 Codes	None
Simon et al.	2018	USA	**Children with medical complexity:** no definition provided	- 0-18 years old - Pediatric Medical Complexity Algorithm: ICD-9 and ICD-10 Codes	None
Srivastava et al.	2016	Australia	**Children with medical complexity:** have chronic and severe health conditions, substantial health service needs, major functional limitations and high health resource utilization	- Cohen's ICD-10 Codes	None
Stephens et al.	2017	USA	**Children with complex chronic conditions:** any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- 1-17 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
Van Doren et al.	2015	USA	**Children with complex chronic conditions:** no definition provided	- ICD-9 Codes based on complex chronic conditions	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Verlaat et al.	2019	Netherlands	**Children with complexity chronic conditions:** any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center	- >18 years old - A modified Feudtner's list was used to classify diagnoses	None
Walter et al.	2018	USA	**Children with medical complexity:** the most severe, complex, and life-threatening health problems	- 0-17 years old - Pediatric Medical Complexity Algorithm (ICD-9)	None
Wang et al.	2009	Canada	**Children with complex chronic conditions**: Any medical condition that can be reasonably expected to last at least 12 months (unless death intervenes) and to involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care and probably some period of hospitalization in a tertiary care center.	- 0-1 years old - Feudtner et al.'s Complex Chronic Condition Classification	None
White et al.	2020	USA	**Children with complex chronic conditions:** no definition provided	- Feudtner et al.'s Complex Chronic Condition Classification	None

Author	Year	Country	Complexity Terminology/Definition	Method of Identification	Model/Framework
Yamada et al.	2020	Japan	**Children with medical complexity:** children with chronic conditions, health care needs, functional limitations, and multiple services needs	- 0-20 years old - Children who have the items described in the severe motor and intellectual disabilities scoring system except for the usage of nebulizer, hyperhidrosis due to hypertonicity requiring frequent changing of clothes and position	Cohen's conceptual framework of children with medical complexity
Zurynski et al. (2019)	2019	Australia	**Children with medical complexity:** long-term health conditions, often involving multiple organ systems and resulting in complex health care needs	-Unknown	-Unknown

2.5.4.3. Practice and policy recommendations

Most included articles discussed the relevance of their results to clinical practices, policy development, and/or research initiatives (Table 2-2). Thirteen studies showed promising results in improving the health care delivery and health outcomes through the implementation of structured clinical complex care programs (n=12) or transitional care interventions (n=1). Each of these studies identified key components for health care providers, policy makers, and researchers to consider for inclusion and adoption in their own clinical context. Some recommendations of note included the implementation of comprehensive care management strategies, nursing care coordinators, and partnerships with primary health care.

Most other studies included in this review used health data to identify population prevalence and characteristics, health system utilization, and associated economic costs. Results from included studies uniformly suggest that children with medical complexity consume high levels of resources within the health care system, despite representing a small proportion of the pediatric population (Berry et al., 2014; Berry, Hall, et al., 2013; Cohen, Berry, et al., 2012; O'Mahony et al., 2013). Medical complexity also appeared to be slightly more prevalent in individuals who identify as member of the Black, Indigenous, and People of Colour community and/or individuals with a lower socioeconomic status (Bjur et al., 2019; Carrilero et al., 2020). These findings resulted in a variety of practice and policy recommendations including, but not limited too: attending to social inequities; increasing social work support; developing structural hospital

expansion plans; increasing palliative care; implementing discharge follow-up; increasing provider education; and intensifying care coordination efforts.

2.5.4.4. Strengths and limitations to using health administrative data

A complete list of strengths, limitations, and data sources can be found in Table 2-4. Examples of reported strengths included: access to large publicly available datasets; less costly and time consuming in comparison to survey or medical record abstraction; less vulnerable to recall bias; linkable datasets; and access to multiple years of data.

Many of the limitations discussed by authors pertained to using health administrative data in general, such as the potential for inaccurate/inconsistent coding/documentation, inability to control for all potential confounders, and difficulty in concluding causality. Authors often noted the difficulty and inability to determine the quality and/or appropriateness of care received. For example, although Ananth et al. (2015) revealed a wide variation in hospitalizations in the last year of life, authors stipulated that future research is needed to examine if these hospitalization were preventable or unwarranted. Authors often highlighted the lack of data for health resource use outside of the hospital setting.

One major limitation noted by most authors was the difficulty in identifying such a multi-dimensional concept as 'medical complexity' using only diagnostic codes in the pediatric population. Individual clinical patient level data captured in detailed medical records or discussed during patient encounters may provide more insight into their complexity status or degree of functional limitations. Furthermore, administrative data often lacked family-identified needs, psychosocial complexity, race/ethnicity, and cultural factors.

Table 2-4. Strengths and Limitations of Health Data Use

Author	Year	Country	Strengths	Limitations	Data Sources
Amin et al.	2018	Canada	- None specifically noted	- Small sample size	- The sleep laboratory database at SickKids
Ananth et al.	2015	USA	- None specifically noted	- Unable to draw conclusions about quality or appropriateness care, quality of life, or comfort at the end of life - Unable distinguish expected from unexpected deaths - Poor sensitivity for palliative care code - Unable to assign a primary diagnosis for children with multiple long term-CCCs because of the CCC classification system does not contain this capability - No data on outpatient, community care or hospice - No data from non-children's hospital	- Pediatric Health Information System
Aritscher et al.	2019	USA	-None specifically noted	-Unavailability of outpatient pharmacy costs -Costs for homecare were not included because they are influenced by baseline, were subject to change during the study period, and are challenging to estimate -Generalization unable to discern	-Hospital logs -Claims data from the 16 Memorial Hermann Health System area hospitals -Billing data from Texas Medicaid
Arthur et al.	2018	USA	- Less costly than survey or medical record abstraction	- Unable to infer causation - Limited generalizability to the entire population (data for only Medicaid-insured populations in 2 states)	- Medicaid Administrative Data

Author	Year	Country	Strengths	Limitations	Data Sources
			- Less susceptible to recall bias in comparison to surveys	- Consent of use of administrative data in the survey cohort was low - Patient-centered medical homes were being established thus could have been an unmeasured confounder	- Family Experiences with Coordination of Care survey field test
Bennett et al.	2020	USA	-Strengths were associated with using electronic medical records data and not the hospital administrative data	-Relying soling on Medicaid claims data results in an under-ascertainment of foster care status and reliance on eligibility codes. -Single urban children's hospital and single urban child welfare system and as such may not be generalizable -May have missed other health services outside of their study location -Unable to determine exact dates of entry and exit of foster care	-The Electronic Healthcare Records -Hospital administrative data
Bergman et al.	2020	USA	-Prospective study to demonstrate system-level impact	-May not be representative of the greater population -Neonatal intensive care unit hospitalizations were included and baselining spending would have been predictable greater before the intervention -Only included those in a hospital-based complex care clinic and their associated primary care provider	-Health care claims data, provided by 9 claims providers consisting of stat Medicaid agencies, managed care organizations, or hospital health plans

Author	Year	Country	Strengths	Limitations	Data Sources
Berry et al.	2014	USA	- None specifically noted	- Limited to ICD-9 codes to distinguish a complex population - Cross-sectional data cannot assess the probability of the cohort experiencing future healthcare expenditures - Unable to distinguish which patients need improved care management - Continuous years of data may be preferably when compared to the four discrete time points in a 10-year period; - Outpatient visits were aggregated - Generalization unable to discern	- The Truven Marketscan Medicaird Database - Agency for Healthcare Research and Quality Healthcare Cost and Utilization Project's Kids' Inpatient Database
Berry et al.	2011	USA	- None specifically noted	- Not intended to be generalizable - Clinical data was not attainable	- Pediatric Health Information System
Berry et al.	2017	USA	- None specifically noted	- Unable to draw conclusions about quality or appropriateness care, quality of life - Cross-sectional data thus unable to assert year-to-year variation in hospital resource - Clinical data was not attainable - Unable to look at resource use between children's and non-children's hospitals - No patient level data - Possible inaccuracies of the ICD-9-CM codes	- Agency for Healthcare Research and Quality Healthcare Cost and Utilization Project's Kids' Inpatient Database

Author	Year	Country	Strengths	Limitations	Data Sources
Berry et al.	2013	USA	- None specifically noted	- No data on non-children's hospital - No outpatient data - No cost data - No information on referrals	- Pediatric Health Information System
Berry, Glotzbecker, et al.	2017	USA	- None specifically noted	- Data may not reflect the true cost of health care delivery - May be variation in the categorization of Medicaid spending across contexts - Can't discern state level data - May not be generalizable to children with private insurance or other contexts	- Truven MarketScan Medicaid Multistate Database
Bjur et al.	2019	USA	-Population-based study -Self-contained health care environment -Applied objective, validated, and individualized measures	-No one agreed system for classifying this population -Detailed information for the regional context	-National Institutes of Health Record Linkage System
Brittany et al.	2015	USA	-None specifically noted	-Did not include reliable measures for technology assistance -Incomplete adjustment for confounding by severity without clinical severity measures -No details if outpatient visits were scheduled or nonurgent -May not be generalizable -Unable to categorize admissions as planned or preventable	-Colorado Medicaid Analytic Extract Data from the Centres for Medicare and Medicaid

Author	Year	Country	Strengths	Limitations	Data Sources
				-Unable to determine separate effects of primary vs specialty or surgical outpatient care -Not confident that home health services or technology assistance could be accurately determined using the data	
Brown et al.	2021	USA	-Provides data to inform resource management and decrease indirect downstream impacts (i.e. family impacts)	-Limits evaluation of complete patient-level clinical data and confirmation of diagnostic coding accuracy. -Combining similar hospitalizations may obscure different between individual diagnosis and treatment group -May have captured planned admissions -Restricted to one hospital and unable to discern hospital use in other settings and may limit generalizability	- Pediatric Health Information System (PHIS)
Burns et al.	2010	USA	- Data collected over 15 years from a large number of hospitals and a large number of patients - Representable to the entire country	- Limited to ICD codes to identify a complex cohort - Data was originally collected for reimbursement purposes - Coding practices may vary between contexts - No patient level data	- Nationwide Inpatient Sample
Buser et al.	2020	Switzerland	-No missing values for most variable was an indicator of a quality dataset	- Immunization data is incomplete -Retrospective analysis limited the data -Data collected outside the hospital were not available	-Administrative Records -Medical Records

Author	Year	Country	Strengths	Limitations	Data Sources
Carrilero et al.	2020	Barcelona	-Population-based individual income data -Robust dataset -Included all children in the study setting and provides a realist view of the current health status of the population	-No specific agree criteria for defining children with medical complexity -Limited data to capture full income status -Private health care provide not available	- The central registry of insured persons - Catalan Health Surveillance System (CHSS) database
Casey et al.	2011	USA	- Data on all clinical contacts and associated costs	- No clinical data - Unable to match some subjected to Medicaid data due to inconsistencies in identifiers between the dataset - May not be generalizable to children with private insurance or other contexts	- Hospital medical records - Arkansa Medicaid Data
Chan et al.	2016	USA	- None specifically noted	- Limited to ICD codes to identify a complex cohort - No patient level data - Limited to children's health centers, thus limiting generalizability - Coding practices may vary between contexts	- Pediatric Health Information System
CIHI	2020	Canada	-Gain a pan-Canadian landscape	-Data coverage for home care for pediatric clients and outpatient specialty clinics were limited -Lack of private service data (school-based program, social and community services, etc.)	-Canadian Institute of Health Information Discharge Abstract Database and Morbidity Database

Author	Year	Country	Strengths	Limitations	Data Sources
					-National Ambulatory Care Reporting System -Patient-Level Physician Billing -National Prescription Drug Utilization Information System -Home Care Reporting System -Canadian Vital Statistic Death Database -Statistics Canada -Semi-Structure Qualitative Interviews
Cohen et al.	2012	Canada	- None specifically noted	- Unable to capture patient who were not hospitalized during the study period - Not complete rehabilitation data, private drug, home care coverage, and providers that bill fee-for-service - Unable to capture family-identified needs, psychosocial complexity, and a direct measure of functional status; - Sensitivity and specificity of the ICD-10 codes used has not be determined	- Discharge Abstract Database - The National Ambulatory Care Reporting System - The Ontario Health Insurance Plan Physical Billing
Cohen et al.	2010	Canada	-Multifaceted evaluation of the programmes from parents, providers, and health databases	-None specifically noted	-Hospital databases -Caregiver semi-structure interviews -Standardized questions

Author	Year	Country	Strengths	Limitations	Data Sources
			-Health system impacts		-Primary care provider questionnaires
Cohen, Lamcombe-Duncan, et al.	2012	Canada	- None specifically noted	- No control group - Data on only two study centers - Participants were recruited due to having 'unmet needs'	- Hospital administrative data - Structured interviews and focus groups with parents
Coller et al.	2018	USA	- None specifically noted	- Unable to infer causality - Single-center design limits generalizability - No data on ambulatory encounters outside of their site or those without documentation - No data regarding the visit content	- Administrative Data - Chart review of a random sample
Coller et al.	2019	USA	-Identify explanatory variables and eventually design interventions to decrease excessive variation	-Admission to another hospital than the one affiliated to the ED was not measure and could have resulted in an underestimation -The definition of randomly selected index visit may have distorted admission rates -Limited to its cross-sectional nature -Severity such as physiological distress and functional impairment is not measured -Unable to determine if admissions were truly preventable or excessive	-Pediatric Health Information System

Author	Year	Country	Strengths	Limitations	Data Sources
				-No provide or family perceptions of the child's health status or admission -No community health services data were available	
Coller Klitzner et al.	2018	USA	- None specifically noted	- Limited in generalizability due to identification of cohort through program enrollment - No data on health care utilization outside of the institutional (if caregiver did not report)	- In-Hospital Administrative Data -Caregiver reports
Coquillette et al.	2015	USA	- None specifically noted	- Retrospective data - Data on a single site - Possibility for inconsistent documentation - Unable to stratify based on characteristics of complexity - Consistency and accuracy of data unable to evaluate	- Hospital-wide database of social work encounters
Edwards et al.	2010	USA	- None specifically noted	- None specifically noted	- Virtual Pediatric Intensive Care Unit Performance System
Edwards et al.	2012	USA	- None specifically noted	- No patient level data - Unable to control for contextual characteristics - Limited to PICU outcomes	- Virtual Pediatric Intensive Care Unit Performance System

Author	Year	Country	Strengths	Limitations	Data Sources
Feinstein et al.	2014	USA	- None specifically noted	- Unable to determine the true nature or severity of recorded adverse health outcomes - No prescription data to compare it too	- ED Sample - Abstracted Billing Records from Health care Cost and Utilization Project
Feudtner et al.	2000	USA	- None specifically noted	- No patient level data - Specific to one state context	- Vital Statistics
Feudtner et al.	2002	USA	- None specifically noted	- Data collected from administrative practices - Data limited to children's hospitals - Limited generalizability - Cannot discern future trends	- National Association of Children's Hospitals and Related Institutions data
Feudtner et al.	2003	USA	- Population level data	- Limited to service arising from a hospital admission - No perspectives from the family or health care provider - Unable to account for migration - Unable to discern causality	- Death and Illness History Database
Feudtner et al.	2007	USA	- None specifically noted	- Possible error in classifying diagnosis and race or ethnicity	- National Center for Health Statistics Multiple Cause of Death Files
Feudtner et al.	2001	USA	- None specifically noted	- Potential for inaccurate coding	- National Center for Health Statistics

Author	Year	Country	Strengths	Limitations	Data Sources
				- Unable to determine which patient would have benefited from supportive care services	
Feudtner et al.	2014	USA	- Assess population-level temporal trends in the proportion of morbidity and mortality associated with CCCs -Assess patterns of healthcare utilization among patients with CCCs -Perform individual-level risk adjustments for patients' CCC status in studies of healthcare processes and outcomes.	-No single classification system is likely to serve all the different goals of research or quality improvement projects that study the needs of children with complex chronic conditions	-CDC Multiple Cause of Death Data
Feudtner Silveria et al.	2002	USA	- None specifically noted	- No data on specific biological, technological, social, and cultural factors - Limited generalizability to one state	- Vital statistics
Forjaz de Lacerda et al.	2017	Portugal	- None specifically noted	- No data on process of care - No data on diagnosis to death - Measurement biases due to modifications in diagnostic, reporting, or coding processes over time - Potential uncontrolled confounder	- The National Institute of Statistics

Author	Year	Country	Strengths	Limitations	Data Sources
				- Small differences and weak association have statistical significance in a large dataset	
Friedel et al.	2019	Belgium	- Multi-center data and as such possible to extrapolate results at a national level	-Reliance on ICD-9 codes and unable to consider ever evolving medical advancements -Could only access aggregated data -Missing data due to an mandatory reporting of pediatric liaison team referrals only starting in 2010	-Existing health administrative data
Gay et al.	2016	USA	-None specifically noted	-Not positioned to assess the impact of home health care on total health care spending as they do not contain outpatient, community, ED, or other health services data necessary to assess this impact -The payments made for home health reported are not equivalent to the reported costs of hospital care -May not have captured al the cohort -Unable to determine truer reasons for home health use -Unable to account for potential confounding factors	- Billing records of BAYADA Home Health Care and the Case Mix database from the Children's Hospital Association
Geurtin et al.	2006	Canada	- None specifically noted	- None specifically noted	- Medical Claims Database

Author	Year	Country	Strengths	Limitations	Data Sources
Geurtin et al.	2009	Canada	- None specifically noted	- None specifically noted	- Linked administrative datasets
Gold et al.	2016	USA	- None specifically noted	- Unable to determine if care delivery was preventable or unnecessary - Unable to capture social or familial attributes, transportation availability, home equipment needs, and local availability contexts - No contextual information - Small differences and weak association have statistical significance in a large dataset	- Pediatric Health Information System
Gordon et al.	2007	USA	- None specifically noted	- Data only from single - No data on outpatient costs of community resources, home care, medical equipment, and pharmaceuticals.	- Hospital Administrative data
Håkanson et al.	2017	15 countries	- None specifically noted	- Coding may differ based on context - Unable to determine quality of care	- International Place of Death project (IPoD) database
Hannan et al.	2021	USA	-Highlights areas for cost savings -Large nationally representative analysis	-Limited by data elements collected and causation cannot be assessed nor implied -Relying on ICD codes and limited by the reliability -Number of entries had missing information for race and ethnicity	-Healthcare Cost and Utilization Project Kids' Inpatient Database

Author	Year	Country	Strengths	Limitations	Data Sources
				-Cannot link patients to future encounters and as such cannot assess ongoing health care use -Variables may be underestimated -The CCC classification system, was not developed from the neonatal population	
Jamorabo et al.	2015	USA	- None specifically noted	- Unable to discern health care delivery; - No data from health care professions or families - No data on insurance coverage	- Vital Statistics
Johnston et al.	2019	USA	-Population variations in intensity of end-of-life care -Highlight critical disparities	-May not be generalizable outside region -Data reported whether an intervention occurred during an admission, not the day it occurred -Did not capture procedures outside the hospital or ED -There are other important aspects of end-of-life care not included in this dataset (access to home hospice, symptom control, etc)	-California Office of Statewide Health Planning and Development (OSHPD) private discharge data linked to death certificates
Jurgens et al.	2014	USA	- None specifically noted	- No data on nonadherence with follow-up appointments - Unable to control for all confounder - Single centre data	- Administrative Data

Author	Year	Country	Strengths	Limitations	Data Sources
Kalzen et al.	2018	Sweden	-Understand factors associated with pediatric intensive care unit admissions could facilitate better allocation of resources, contribute to sharing correct advice and expectations and identify important targets for future research efforts -Only 5.3% lost to follow-up	-May have missed some children not admitted to the PICUs in the study -Small cohort in the dataset -Lack of data concerning the physical location of death and detailed circumstances of the event outside of the PICU -The number of multiple admissions may be underestimated by the study design -Cannot evaluate very late admissions	-Chart review -Swedish File of National Registration -Swedish Intensive Care Registry
Keim-Malpass et al.	2020	USA	-None specifically noted	-There may be unobserved confounding factors -Measurement error may exist within the medical complexity variables -Cannot be generalized those not under Medicaid -Data was between 2011-2013	-Medicaid Analytical Extract Files
Kieran et al.	2019	Canada	-Identify areas for improving care	-Data was limited to 1 year in 8 patients and encounters were limited to pediatric hospitals -Re-hospitalization rates are likely underestimated -Single-center -Retrospective design -Feasibility assessment was not comprehensive and impact on outcomes	-Repository of electronic discharge summaries -Electronic and paper charts -The Canadian Neonatal Network database -Health Authority Database

Author	Year	Country	Strengths	Limitations	Data Sources
				such as parental satisfaction, length of stay, unmet needs, and health care costs were not measured	
Kim et al.	2017	Korea	- None specifically noted	- No data on socioeconomic status	- Cause of Death Statistics
Kim et al.	2018	Korea	-National wide understanding of trends of pediatric deaths	-Variables such as socioeconomic positions, family income, education of parents, are not included -Cross-sectional design	-Korean Statistical Information Services of Statistics Korea
Kingsnorth et al.	2015	Canada	- None specifically noted	- Lack of sociodemographic diversity	- Semi structured interviews - Focus group - Document review - Administrative databases.
Lacerda et al.	2014	Portugal	- None specifically noted	- None specifically noted	- Death certificate data
Lacerda et al.	2019	Portugal	- Obtain population level data as a starting point for the revision of health care -Needs estimation is easily available and involves a large dataset	-Correct codification cannot be ensures and may underestimate the number of episodes - Severity or stage were not available - Anonymized data made it impossible to link with other datasets	-Administração Central do Sistema de Saúde (ACSS), from which anonymised data are released to the National School of Public Health

Author	Year	Country	Strengths	Limitations	Data Sources
			- Can be used for comparison in future years	- Indirect costs are not taken into consideration (extra costs to family, loss of income, work/school absenteeism)	
Lindley et al.	2013	USA	- None specifically noted	- No data on cause of death, symptoms, and family characteristics - Limited generalizability - Infants less than 1 year of age may be less likely to have been enrolled in Medicaid prior to death	- California Medicaid program's Medicaid Analytic Extract (MAX) files.
Lindley et al.	2016	USA	-Identification of co-occurring conditions for future studies -This information may guide the design of intervention specific to multiple complex chronic conditions tailored at the end of life -Novel method for latent class analysis for multiple complex chronic conditions	-Limited generalizability to those not in the study population or setting -No casual or temporal explorations -Incomplete record keeping may resulted in an underestimate of the study population	-California Medicaid Data
Lindley et al.	2021	USA	-Improving the ability to select the most appropriate measure of illness severity for infants	-Those with changing diagnoses may not be the final diagnosis reported at death -The nature of the dataset may have introduced bias when the ICD codes were recorded	- Healthcare Cost -Utilization Project (HCUP) Kids' Inpatient Database (KID)

Author	Year	Country	Strengths	Limitations	Data Sources
			-The dataset used it the largest publicly available all payer pediatric inpatient database in the US -Large, national sample size, and improve external validity -Provides valuable insight into the performance of two classification measure for pediatric nursing research in predicting patient outcomes	-Limited generalizability to the US or other health care institutions	
Lindley et al.	2020	USA	-Improving knowledge about classifying children is critical to advancing science	-Limited generalizability -Only discharge diagnoses were included in the dataset (not admission diagnoses) -Infant deaths outside the hospital were not included in the data	- Healthcare Cost -Utilization Project (HCUP) Kids' Inpatient Database (KID)
Lindley et al.	2019	USA	-Can provide information to target research and interventions	-Race and ethnicity data is often incomplete or missing in secondary data -The datasets have a disproportionate representation of states with a high population that contains hospitals with more annual discharges -Represent hospital discharges, not patients	- Healthcare Cost -Utilization Project (HCUP) Kids' Inpatient Database (KID) -Agency for Healthcare Research and Quality

Author	Year	Country	Strengths	Limitations	Data Sources
				-Limited generalization to other pediatric contexts -Discharge information, not admission complaints -Deaths outside of the hospital were not captured	
Maenge et al.	2017	USA	- None specifically noted	- None specifically noted	- Health plan claims data
Marshall et al.	2017	USA	- None specifically noted	- None specifically noted	- Texas Medicaid administrative claims data
Moura et al	2017	Brazil	- Important to use health data to identify issues and improve data capture	-No data on private institutions -Not obligatory to record secondary causes - Unable to capture diagnosis mistakes, repeated hospitalizations, and patient transfers	- Ministry of Health's National Hospital Information System
Murtagh et al.	2014	USA	- None specifically noted	- Unable to track specific patients over time - No clinical data - Risk for misclassification bias - Contextual differences may differ data	- Nationwide ED Sample (NEDS) - Healthcare Cost and Utilization Project (HCUP) by the Agency for Healthcare Research and Quality

Author	Year	Country	Strengths	Limitations	Data Sources
Neff et al	2016	USA	-Illustrates the use of a risk adjustment tool in determining rates for public reporting -A novel way to measure the risk of central line blood infection in children that reflect complexity of conditions	-May not be representative -Cannot compare to line days, as this is not included in administrative data	-Children's Hospital Association Comparative Case Mix Data Program through a standard universal billing submission process
Neff et al.	2015	USA	- None specifically noted	- Definition for the 3 groups in the gold standard were developed by consensus and subjective in nature - Only 1 reviewer to review charts limiting an evaluation of reviewer reliability - Gold standard sample in this study is limited to children who were hospitalized or used the ED	- Data of encounters for managed care - Claims for fee for service
O'Mahony et al.	2013	USA	- None specifically noted	- May under or over capture cohort - Single setting limiting possible generalizability - Inability to control for all contextual confounders - Unable to identify social complexity	- Hospital administrative data

Author	Year	Country	Strengths	Limitations	Data Sources
Parente et al.	2021	USA	-None specifically noted	-Reliance on retrospective analysis of diagnostic codes from administrative databases -Inability to identify CMC who will have a persistent high resource use -Limited utility at point of care -Data on hospital use was only at one institution and did not include outpatient, urgent care, subspecialty, or ED costs.	-Electronic Medical Records -Hospital administrative data
Peter et al.	2011	USA	-Assess effectiveness of a care coordination approach -Identify system barriers and address issues including improve care	-Limited to 10 months due to funding constraints	-Hospital Morbidity Data System (HDMS)
Phillips et al.	2019	USA	-All data used in the research are available in widely distributed public use databases	-From one single state, during a limited time period -Hospital discharge data do not contain information about health services a patient received prior to hospitalization, current condition status on admission, or services received during hospitalizations	-Texas Inpatient Public Use Data File -The Area Health Resource Files
Pulcini et al.	2021	USA	-Assess opportunities for improving health services	-May only generalize to pediatric hospitals -True preventability of the encounter cannot be determined without clinical level data	-Pediatric Health Information System Database

Author	Year	Country	Strengths	Limitations	Data Sources
				-Peri-ED use outpatient health services are not included -No data on whether study participants had a medical home or complex care program that coordinates their care -May be confounding factors that are unable to account for given the retrospective nature of the data	
Ralston et al	2015	USA	-Assess the full continuum of care -Evaluate directed management programs to assess best practices sooner	-No causality -Lack validated measures on which to assess improved patient outcomes for this population -Limited ability to classify severity of illness -Potential for coding and selection bias -There are other methods to identify this cohort in population-level data -Assignment to hospitals may have misattributed some patients -Cannot distinguish if the variation in care is drive by inpatient, or emergency care and cannot link the variation encountered to patient outcomes -May not be generalizable	-The database drew from 6 sources: the commercial all payer claims database and the Medicaid claims databases from each of the 3 states
Rasooly et al.	2020	USA	-Measure the quantity of services distributed -National perspective on home nursing services	-Children with commercial or private insurance may have lower odd of receiving home care and as such funding may not be generalizable to all children	-Truven MarketScan Commercial Insurance Dataset

Author	Year	Country	Strengths	Limitations	Data Sources
			-Claims data from a rigorous dataset	-Results are comparable to those commercial insured -Cannot differentiate between skilled nursing home visits and private-duty nursing visits -Limited by available of patient and clinical information within administrative data -Race, ethnicity, or socioeconomic factors were missing -No information on hospital, facility, or clinician was available	
Shumski et al.	2018	USA	-To optimize performance of well-baby visits -Can use the finding to develop policies and programs to enhance primary care services	-Unable to distinguish reasons for observed patterns -Unable to control ICD coding -Possible that some individual may have received care in other types of primary care sites -Unable to assess important attributes of medical complexity including severity, medical fragility and functional impairments -Assessed only disability from data on the reason for Medicaid enrollment and as such could has missed children enrolled for other reasons -Unable to distinguish children support with a Medicaid waiver	-MarketScan Medicaid Database

Author	Year	Country	Strengths	Limitations	Data Sources
				-Children with private insurance were not included -May not be generalizable	
Silber et al.	2018	USA	-None specifically noted	-None specifically noted	-Pediatric Health Information System (PHIS)
Silber et al.	2019	USA	-Provides benchmark and a view of resource consumption	-Poor coding or inconsistency in coding may lead to bias -Lack of socioeconomic status	-Pediatric Health Information System (PHIS)
Silverman et al.	2016	USA	-None specifically noted	-None specifically noted	-Pediatric Health Information Systems (PHIS)
Simon et al.	2017	USA	- None specifically noted	- Single center data - Limited power for utilization outcomes - No data to conduct subgroup analysis - Unable to capture any utilization outside of the facility	- Hospital Morbidity Data System - Inpatient discharge summary data
Simon et al.	2010	USA	-Useful pragmatic tool to identify a population of medically complex children -Findings provide can better inform system of care for medically complex children	-An adequate description of medically complex children has been elusive -Used attributes of medical complexity, rather than a direct measurement of complexity -Absence of patient identifiers limits the descriptions and inferences about admission, not patients	-Healthcare Cost and Utilization Project Kids Inpatient Databases

Author	Year	Country	Strengths	Limitations	Data Sources
			-Evaluate both the quality of and evidence in care	-Increase in prevalence may be due to increased documentation -ICD-9 codes do not capture functional limitations, family needs, social factors, or the use of outpatient services or home technology -	
Simon et al.	2014	USA	- None specifically noted	- Potential for incomplete data set - No data on encounters that do not result in a claim	- WA-Medicaid and Seattle Children's Hospital
Simon et al.	2017	USA	- None specifically noted	- Potential for incomplete data set - No data on encounters that do not result in a claim	- WA-Medicaid and Seattle Children's Hospital
Simon et al.	2018	USA	- None specifically noted	- Potential for incomplete data set - No data on encounters that do not result in a claim - Unable to capture social, emotional, and economic factors	- Seattle Children's Hospital Discharge Data
Srivastava et al.	2016	Australia	- Large administrative databases results in generalizable information for Australian public hospitals - Used previously published codes to identify the cohort	- Only examined one year of data - No patient level data - Unable to track the costs of repeated hospitalizations - No data on outpatient encounters	- Admitted Patient Care (APC) National Minimum Data Set - National Hospital Costing Data Collection

Author	Year	Country	Strengths	Limitations	Data Sources
Stephens et al.	2017	USA	- Assess areas of practice/policy implications	-May have underestimated the number of visits for outpatient treatment for constipation as laxatives are not covered in all states -Retrospective in nature and may have missed constipation if other diagnostic codes were used -May not be generalizable to those not under Medicaid or other geographical areas	- Truven MarketScan Medicaid claims database
Van Doren et al.	2015	USA	- None specifically noted	- Reliance on ICD coding rather than symptomology - Potential for miscoding in dataset	- Kids' Inpatient Database
Verlaat et al.	2019	Netherlands	- None specifically noted	-None specifically noted	-Netherlands National Pediatric Intensive Care Unit Database
Walter et al.	2018	USA	- Large data set - Applied the Pediatric Medical Complexity Algorithm - Able to compare rates of utilizations across different health plans	- Limited generalizability to of all privately insured children or other countries without similar health care structures - No data on socioeconomic status, race, education and other source of health insurance coverage - Unable to discern causality may not be generalizable to other countries without similar insurance and health care delivery structures	- Watson/Truven AnalyticsSM MarketScan

Author	Year	Country	Strengths	Limitations	Data Sources
Wang et al.	2009	Canada	- None specifically noted	- This cohort has a range of diagnoses that may all have different influences - Potential for misclassification of neighborhood - No clinical data	- The hospital discharge abstract database - Registered persons database - Canadian Census
White et al.	2020	USA	-Describe the frequency of interfacility transfer among CCC and identify transfer status as an independent risk factor for long length of stay, receipt of critical care services and in-hospital mortality using a nationally representative data set	-Unable to individually identify patients and link data across hospitalizations -Inability to assess disease severity, before and after transfer -Length of stay in the transferring hospital was unable to be assessed -Multiple definition for classifying children with medical complexity	-Healthcare Cost and Utilization Project Kids' Inpatient Database
Yamada et al.	2020	USA	-None specifically noted	-Definitions are not uniform for CMC -Retrospective data -Severity data came from electronic medical records	- National health insurance claims data - Electronic Medical Records
Zurynski et al.	2019	Australia	-None specifically noted	-None specifically noted	-Hospital administrative data

2.5.5. Discussion

This scoping review identified various means of leveraging health administrative data to inform and support the development of health policy and practice recommendations for children with medical complexity and their families. Despite the development of a definitional framework to help unify this pediatric population (Cohen et al., 2011), there remains a variety of terminologies and definitions used to describe and conceptualize this population of interest. While a previous review examining transitional care programs for these children and youth identified a trend towards the adoption of Cohen et al.'s (2011) definitional framework, this review did not uncover any particular trends (Breneol et al., 2017). Rather, most studies referred to Feudtner et al.'s (2000) definition for children with complex chronic conditions. This may be due to the high proportion of studies originating from the US and therefore, a tendency towards the use of a US developed definition. Establishing conceptual agreement across initiatives should be promoted to assure that tools being developed to identify these children at a population-level are measuring the same concept. For example, when Lindley et al. (2021) evaluated the original and revised CCC Classification System, they speculated that the modifications appeared to reflect a change in the theoretical constructs underpinning the system; from 'life-limiting illness' to 'medical complexity' (Lindley et al., 2021). Greater efforts are needed to explore these theoretical constructs and ensure the use of one uniform conceptualization to encourage the synthesis of related knowledge and facilitate consistency across policy, clinical, and research initiatives.

Lists and algorithms using diagnostic codes were the most common strategy in identifying children with medical complexity within health administrative data. The

predictivity ability, sensitivity, and specificity of some of these algorithms have been evaluated and showed promising results in identifying this unique pediatric population (Berry et al., 2017; Feudtner, 2000; Feudtner et al., 2014; Lindley et al., 2021; Simon et al., 2018). However, concerns about focusing solely on medical diagnoses persists. Functional limitations and family-identified needs are two of the four characteristics of children with medical complexity (Cohen et al., 2011). Unfortunately, health administrative data may not always provide the necessary clinical and individualized information to examine these characteristics. Interestingly, only five studies included in this review used family reported outcomes to supplement health administrative data (Arthur et al., 2018; Canadian Institute for Health Information, 2020; Cohen, Lacombe-Duncan, et al., 2012; Coller, Klitzner, et al., 2018; Kingsnorth et al., 2015). Combining family completed surveys with health administrative data may be one avenue to address these gaps; however, few studies have examined the feasibility and accuracy of this method. One study showed promising results while exploring the application of linking a non-diagnosis specific screening tool to identify children with special health care needs to provincial level data (Arim, 2015). All of this considered, until future research is pursued to develop a strategy that considers all dimensions of children with medical complexity at a population level, diagnostic code lists and algorithms are a promising method of identification given their open availability and positive evaluation.

The advantages to using health administrative data when compared to using survey data merit further exploration. The National Survey of Children with Special Health Care Needs is one example of a national-wide survey implemented to better understand children with medical complexity and their health resource requirements

(Bramlett et al., 2009; Coller et al., 2016; van Dyck et al., 2002). Nation-wide surveys have benefits, such as using family-reported variables not captured in health administrative data, and as such holds promise in getting a more comprehensive picture of the lived experiences of these children and family. However, they can be very costly and time-consuming to conduct (Arthur et al., 2018). Further, research comparing self-reported health care utilization and medical records has shown a tendency for individuals to underreport health service use, especially amongst the highest-resource users (Ritter et al., 2001). This, coupled with the advantages of having a large, readily available and linkable dataset, further strengthens the justification to use health administrative data in the study of children with medical complexity.

Included studies were primarily heterogeneous in nature with respect to their primary research aim, highlighting the wide-ranging possibilities for health administrative data usage in understanding the impact medical complexity has on children and families. For example, by linking multiple administrative data sources Cohen and Berry et al. (2012) determined that while children with medical complexity represented less than 1% of the pediatric population, they accounted for over one-third of pediatric health care spending (Cohen, Berry, et al., 2012). These findings highlight the critical importance for practice and policy reform to address this small population's disproportionate health care system usage. One potential strategy noted by authors is the need to intensify care coordination efforts (Cohen, Berry, et al., 2012). Health administrative data was also used as an important tool to evaluate clinical initiatives. Casey et al. (2011) demonstrated that their multidisciplinary care clinic, providing comprehensive care management for children with medical complexity, resulted in a

significant decrease in inpatient use and cost savings one year following the implementation of their initiative. Presenting this economic case to decision makers has the potential to enact meaningful practice and policy changes. Researchers, clinicians, and policy makers are encouraged to consider the benefits of using health admin data to inform strategic directions for practice and policy reform and evaluate associated system and individual level impacts for children with medical complexity and their families.

2.5.6. Limitations

Given the wide variety of terminologies used to describe this population of interest, the search strategy may not have captured all relevant studies. Inclusion of studies written in English only may have limited the scope of this review and may explain the large portion of studies arising from North America. With these limitations in mind, the implementation of a broad search strategy and eligibility criteria likely captured a large portion of studies in this area of interest.

2.5.7. Conclusion

It is critical that we continue to improve health care systems to address the care needs of children with medical complexity and their families. As medical technologies and treatment continue to advance, this population of interest will continue to grow and have major implications for the healthcare system. Much is left to be uncovered about this population and health administrative data have the potential to contribute to advancing this work. With careful application of methods and understanding of the advantages and disadvantages to using health administrative data, health researchers can leverage its strengths to examine the impact of medical complexity on children and their

families within the health care system and to inform the development of health policy and program initiatives.

2.6. Literature Review Conclusions

Literature exploring pediatric complex care has been steadily increasing over the past two decades (Cohen et al., 2018). Emerging evidence reveals that while children with medical complexity represent a small proportion of the overall pediatric population, they consume a disproportionate amount of health care resources. Despite this high resource use, families of children with medical complexity are reporting inadequate support, challenges accessing services, and unmet health care needs. These findings highlight a clear disjuncture between our current health care structures and the care needs of children with medical complexity and their families. It is critical that we begin to develop and evaluate family-centered strategies tailored to meet the needs of this population. However, to achieve this, we must first address two major knowledge gaps within our current literature. First, the majority of studies exploring health resource use and/or the care needs of children with medical complexity and their families rely primarily on quantitative or qualitative data alone. Health administrative data can provide critical information related to the prevalence, characteristics and health resource use for this vulnerable population. However, families often require access to a multitude of psychosocial, medical, and financial resources to care for their child at home that go uncaptured by routinely collected health data. It is critical that we gain a greater understanding regarding the health resources valued and used by families. Qualitative methods have the power to explore families' experiences and needs to successfully thrive in their home community. As such, combining population level data with the stories and

experiences of individual families could assist with developing a more comprehensive understanding of the health resource use and health needs for children with medical complexity. Second, much of our current literature has been conducted within United States' health care systems. While we can gain learnings from these studies, it is critical that we begin to gain a greater understanding regarding this population within Canada. Evidence is starting to emerge over the past 10 years describing children with medical complexity in Canada, but further contextual work is needed to understand potential variation, needs and opportunities from a provincial and regional perspective. The following study has been designed to address this knowledge gap by examining both health administrative and family-reported data to achieve an in-depth understanding into the health resource use and care needs of children with medical complexity and their families in three Canadian provinces.

Chapter 3: PROTOCOL

The research protocol in the following section also appears in: Breneol, S., Macdonald, M., Montelpare, W., Stewart, S.A., Martin-Misener, R., Vine, J., Curran, J.A. (2022). Children with medical complexity in the Canadian Maritimes: A mixed methods study protocol. *JMIR Research Protocols*. 11(4):e33426. doi: 10.2196/33426

Statement of manuscript contribution: SB conceptualized the study with input from JAC, MM, WM, SS, RMM, and JV. SB submitted all data access requests and research ethics board applications. SB drafted the manuscript. All authors contributed to revising the manuscript.

3.1. INTRODUCTION

Ongoing developments in the medical field have improved survival rates and management of children with complex chronic health conditions (Cohen et al., 2018; Perrin et al., 2014). Frequently described as 'children with medical complexity', these children are often diagnosed with a wide array of pediatric conditions (Breneol et al., 2017; Cohen et al., 2011). Recognizing the need for conceptual agreeance across clinical and research initiatives to distinguish this unique pediatric population, Cohen et al. (2011) presented a definitional framework for children with medical complexity. Rather than proposing a diagnosis specific definition, this framework describes a non-categorical and inclusive approach to conceptualizing medical complexity in children (Cohen et al., 2011). Cohen et al. (2011) identified four intersecting domains specific to children with medical complexity: 1) The presence of a diagnosed or suspected complex chronic condition; 2) significant family-identified needs; 3) functional limitations that are often severe and may require the use of technological assistance; and 4) high health

resource use (Cohen et al., 2011). This definitional framework is now being used widely to describe this vulnerable and important pediatric group across policy, clinical, and research sectors (Breneol et al., 2017; Canadian Association of Pediatric Health Centres, 2018).

Literature exploring pediatric complex care has been steadily increasing over the past two decades (Cohen et al., 2018). Evidence emerging over this time suggests that while children with medical complexity represent a small proportion of the overall pediatric population, they consume a disproportionate amount of health care resources (Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). Despite approximately 89% of these children being discharged home from inpatient settings (Berry, Hall, et al., 2013), their resource-intensive needs are infrequently met by the current health system, leaving a substantial burden on families to provide expert medical and supportive care and facilitate care coordination activities for their child. Furthermore, these families often report inadequate support, difficulty accessing services, and various other unmet health care needs (Aboneh & Chui, 2017; Brehaut et al., 2011; Curran et al., 2020; Kuo et al., 2011; Kuo et al., 2014; Toly et al., 2012). These findings highlight a clear disconnect between currently available health resources and the care needs of children with medical complexity and their families.

It is critical that we begin to develop and evaluate family-centered strategies tailored to the needs of this population. However, to achieve this, we must address major knowledge gaps within the literature. First, much of the empirical research has been conducted within the United States' (US) health care system (Berry et al., 2017; Berry, Hall, et al., 2013; Kuo et al., 2011; Kuo et al., 2014), with only two main reports

published in the last 10 years examining the prevalence and health resource use of Canadian children with medical complexity (Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). While US studies are informative, it is critical to gain greater regional and jurisdictional understanding of the prevalence, clinical characteristics, health resource use, and health needs of this population within Canada. Second, the literature exploring this pediatric population primarily relies on routinely collected health administrative data (Agrawal et al., 2016; Berry et al., 2017; Berry, Hall, et al., 2013; Cohen, Berry, et al., 2012). While this data source has several strengths, such as having access to large population samples across various timeframes, there are important limitations to the use of health data to consider. Largely, families utilize a range of health resources not captured by health administrative data alone (i.e. private respite care services, local community-run health programs, private physiotherapy, etc). This leads to a significant gap in our understanding of the extent of health resource use and care needs of children with medical complexity and their families. Qualitative research methods are designed to explore this gap whereby researchers speak directly to children and families about their lived experience. As such, combining health administrative data with richly descriptive qualitative reports from families is one strategy to fully explore their health resource use and care needs. Employing a mixed methods approach could provide researchers, clinicians, families and decision makers with a detailed and comprehensive understanding into the prevalence, clinical characteristics, health resource use, and health care needs of children with medical complexity and their families. Without this information, decision makers may not have

all the necessary information to create family-oriented recommendations to support the health of these children and their families.

There remains a significant gap in our understanding into the true extent of health resources utilized by children with medical complexity and their families living in their home communities. Greater efforts are needed to map health resource use across the public, private, and community sectors to provide the foundational knowledge needed to develop evidence-informed recommendations and strategic directions to support the health and needs of children with medical complexity and their families. As such, the purpose of this research was to use both health administrative and family reported data to gain an in-depth understanding into the prevalence, health resource use, and care needs of children with medical complexity and their families in the Canadian Maritimes [(Prince Edward Island, (PEI), Nova Scotia (NS), and New Brunswick (NB)]. To achieve this objective, the following research questions will be addressed: 1) What are the prevalence and clinical characteristics of children with medical complexity in the Canadian Maritimes?; 2) What is the health resource use as described by health administrative data for children with medical complexity?; 3) What are the family-reported experiences, health resource use, and health needs of children with medical complexity and their families? and; 4) In what ways do the family-reported experiences, care needs, and health resource use converge and diverge with the characteristics and health service use as reported by health administrative data among children with medical complexity and their families in the Canadian Maritime Provinces?

3.2. METHODS

To achieve our research aim, an explanatory sequential mixed methods design (quan → QUAL) will be employed (Creswell & Clark, 2011). The weighting and emphasis of this mixed methods approach was placed on the qualitative phase to amplify the voice of families at the forefront of this work (Creswell & Clark, 2011). Ethics approval has been obtained from the IWK Health Centre's and Health PEI Research Ethics Board (#1026835; #1024934).

3.2.1. Phase One

3.2.1.1. Design

To understand the prevalence and health resource use of children with medical complexity, we will conduct a secondary analysis of routinely collected health administrative data. Access to these data will be obtained through the Health Data Nova Scotia (HDNS) Secure Data Repository Platform and the pediatric tertiary care facility's Decision Support Services. To achieve this study objective, a two-phased process will occur. First, discharge data from the Maritimes' only pediatric tertiary care facility will be used to identify and characterize children with medical complexity in the Maritimes. Next, the health card number of all Nova Scotia residents identified within the cohort will be linked to the provincial's health administrative datasets to examine their health resource use.

3.2.1.2. Study Setting

The primary site of this research is the only pediatric tertiary care facility located in the Canadian Maritimes, providing a unique opportunity for multi-jurisdictional research given their mandate to care for children, youth, and families in all three

provinces. This site was chosen for this research as children's hospitals have been identified as the main care site for children with complex chronic health conditions and can provide a representative sample of this population in the three provinces (Berry, Hall, et al., 2013; Bogetz et al., 2015). Based on the limitations in cross-provincial data linkages, health resource use will only be explored in Nova Scotia (NS). NS data is the sole source of health resource use by children with medical complexity and will be relied upon to extrapolate usage in the other Maritime Provinces (PEI and NB).

3.2.1.3. Data Sources

Five health care databases will be accessed in this study: the pediatric tertiary care facility's discharge data, MSI Physician Billings (MED), National Ambulatory Care Reporting System (NACRS), Canadian Institute for Health Information Discharge Abstract Databases (CIHI-DAD), Vital Statistics – Death (VITAL).

3.2.1.4. Study Population and Identification

All children/youth between the ages of 0-18 years old discharged from the pediatric tertiary care facility between April 1st 2004 and March 31st 2014 and meet Cohen et al.'s (2011) definitional framework for children with medical complexity (Cohen et al., 2011), will be included in the analysis. We know from previous published literature that children with medical complexity are small in numbers. As such, to ensure a cohort suitable to power a regression analysis, we will examine prevalence over a 10-year time period. This timeframe also provides health resource data for up to 5 years (up to March 2019). Cohen et al.'s definitional framework will be operationalized through the application of the Pediatric Medical Complexity Algorithm 3.0 (Simon et al., 2018). The Pediatric Medical Complexity Algorithm (PMCA) is a validated algorithm to identify and

classify the pediatric population based on level of medical complexity within health administrative data (Simon et al., 2018). An individual child will only be included once in the cohort. If a child was discharged more than once during the study period, the earliest discharge date with a complex chronic condition will be used as the index date to begin tracking health resource use. Our final cohort will be all children who are classified by the algorithm as a child with a 'complex chronic condition'. Health card numbers for the identified sample will be retrieved from the pediatric tertiary care facility's discharge database by a data analyst and sent to a health system partner organization for encryption to preserve confidentiality. These encrypted health card numbers are then sent directly to the provincial health data repository for linking with MED, NACRS, and CIHI-DAD, and VITAL for all Nova Scotia residents. At no point during this process does the research team have access to the unencrypted or encrypted health card numbers. A 3:1 matched control cohort will be identified by using age, sex, and postal code as matching variables. Matched cases will be used to provide a comparator population and to control for potential confounders that may influence health resource use (Gordis, 2014; de Graaf et al., 2011). Once the cohort is identified, their health resource use will be followed up over a 5-year period or up to the age of 18.

3.2.1.5. Measures

Variables related to patient demographics will include age, sex, urban/rural residence, organ system involvement, and care team characteristics. Race and ethnicity data were not accessible, as they are not included as routinely collected variables in the health administrative datasets. Variables related to health care utilization will include inpatient hospital visits, outpatient hospital visits, home care services, emergency

department visits, and transfers between care locations. These health data will encompass both tertiary and community care hospitals (See Appendix C for full variable list).

3.2.1.6. Data Analysis

To address our first research objective, the prevalence of children with medical complexity from 2004-2014 will be estimated using prevalence rate calculations. The estimated prevalence rate will be obtained by dividing the number of cases of children with medical complexity identified by the PMCA with the total number of children estimated in the Statistics Canada Census Data for Nova Scotia (2016). Prevalence will be further stratified based on: age, sex (as assigned at birth), clinical diagnosis category (categorized by the PMCA (Simon et al., 2018)) and geographical location (rural/urban). Urban and rural residence will be determined by the first three digits of their postal code. Age will be analyzed categorically (0-11 months, 1-4 years, 5-9 years, 10-13 years, and 14-18 years). Descriptive statistics will be used to describe the characteristics of children with medical complexity (age, sex, clinical diagnosis category and rural/urban location).

To address the second research objective, health resource use for both case and control cohorts will be explored using descriptive and inferential statistics. Descriptive statistics including mean (± standard deviation), median (interquartile range), and percentage (count) will be used to describe the number of services received, types of medical specialties, and health resource use for children with medical complexity over a 5-year follow-up period. Health resources will be grouped by inpatient admissions, emergency department visits, length of stay, location of care, outpatient services, home

care use, and ambulance transfers. Rates of health resource use and length of stay will be further stratified by clinical diagnosis category, age, sex, urban/rural location.

To explore for any associations between child characteristics and health system use, a negative binomial regression analysis will be run. The primary outcomes of interest will be counts of hospital readmission as defined by any type of inpatient admissions (i.e. intensive care admissions), emergency department visits, and outpatient community services defined as primary care visits, home care services, and clinic services. Predictors of interest are age, geographical location, and sex. Lastly, to explore the hazard ratios for time to and between health resource use a Prentice, Williams and Peterson (PWP) gap time model will be used. This will illuminate patterns of health resource use within the identified cohort. All data analysis will occur using the statistical software program, STATA version 9.3 (STATA, 2018).

3.2.1.7. Anticipated outputs

There are three main outputs from this first phase. First, we will have a detailed description of the prevalence of children with medical complexity in three Canadian provinces. Second, we will have an understanding into the formal health service use of this vulnerable population. Third, results from this phase will inform participant recruitment and the development of a theoretically-based interview guide for use in Phase 2 to capture family identified health resource use and needs (described in Phase 2 methods).

3.2.2. Phase Two

3.2.2.1. Design

A case study design will be employed to examine the health resource use and health needs of children with medical complexity and their families in each of the Maritime provinces. While the definitional framework for children with medical complexity advances our characterization of this pediatric cohort, there remains little understanding regarding the key health outcomes and their measurability for this population (Barnert et al., 2018). Case study research is an approach to developing and generating a rich description of complex phenomena in the 'real-world context' and can elicit the answer to how, what, and why questions (Yin, 2014). For example, how do children with medical complexity and their families use the health resources, what types of services are accessed, what gaps and areas for improvement exist, and why these patterns may be occurring. Each case will be informed by three sources of data: 1) interviews with families; 2) interviews with individual members of the care team; and 3) self-reported health resource use.

3.2.2.2. Study population and sampling

A purposive sampling strategy (Polit & Beck, 2012) will be used to recruit children, aged 0-18 years old, matching Cohen et al.'s (2011) definitional framework for medical complexity (Cohen et al., 2011). We will purposively recruit children and families fitting specific characteristics (i.e. demographics, clinical characteristics, level of complexity, health resource use, etc.) based on significant findings from Phase 1. For example, this may include certain clinical presentations or urban/rural residency that may prompt further examination. Families must be primary residents within one of the

provinces of interest (NS, NB, or PEI). Two cases from PEI, NB, and NS will be developed to capture the potentially varying experiences of children and families living inside and outside of the provincial boundaries of the pediatric tertiary care facility. Children and families must be able to speak the English or French language to be eligible to participate. We will not attempt to specifically recruit participants identified within the Phase 1 dataset. However, it is anticipated that potential participants in the qualitative phase will have been captured in the health administrative cohort.

The number of participants is not the focus in case study design, rather it is about gathering multiple forms of data from various perspectives to develop a deeper understanding of a specific case. Two case studies will be developed for each Maritime province, PEI, NS, and NB, resulting in a total of six. This will allow for the examination of differing familial and contextual factors. One primary caregiver will self-identify as the primary contact for the study. If more than one caregiver would like to be interviewed, all caregivers will be interviewed at the same time. The primary caregiver will also be asked to identify a maximum of two key members of the care team that can be approached for an interview.

Multiple recruitment strategies will be used to reach our target population. Recruitment flyers will be posted to relevant units at the pediatric tertiary care facility, community pediatric care sites, and social media platforms. We will also circulate the recruitment poster and study summary via email to key stakeholders involved in the care of children with medical complexity and families to share with their networks. An email and phone number contact for the principal investigator (PI) will be on all recruitment materials. The PI will respond to all inquiries related to study

participation and will provide potential participants with additional study information and an eligibility screening checklist. Once eligibility is confirmed, the PI will forward the consent form for their review. This consent form will be reviewed with participants prior to the interview and signed.

3.2.2.3. Measure

A semi-structured interview guide for families will be developed based on significant findings from Phase 1 and from the 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018). Barnert et al. (2018) provided the most comprehensive understanding of the conceptualization of health for children with medical complexity and their families by creating a conceptual framework outlining 10 Domains of health (Barnert et al., 2018). These domains included: (i) basic needs; (ii) inclusive education; (iii) child social integration; (iv) child health-related quality of life; (v) long term child self-sufficiency; (vi) family social integration; (vii) community system supports; (viii) health care system supports; (ix) high quality patient-centered medical home; and (x) family-centered care (Barnert et al. 2108). This interview guide will also include prompts that are informed by significant findings from Phase 1. As case study research is designed to address how, what, and why questions, findings from Phase 1 will be used to develop prompts and potential questions to create a more comprehensive understanding of observed and/or unobserved patterns of resource use.

Before family interviews occur, the interview guide will be pilot tested through a "think-out- loud" session with a parent researcher who works at the tertiary care facility. Changes will be made as required following this pilot testing.

3.2.2.4. Procedure

All interviews will take place over the phone or by Zoom Video Conferencing system (Zoom, 2019). Family interviews are anticipated to last 45-60 mins. Demographic and socioeconomic information will be collected on families at the beginning of the interview process. This will include number of individuals in the family unit, child's health condition(s), type of medical device/technology, urban/suburban/rural community, child's gender (as identified by the child), participant's gender, child's age, participant's age, participant's race/ethnicity, employment status of the caregiver(s), and access to transportation services. Families will be asked to identify all of the individuals or specialty clinics involved in the care of their child. An additional data source for the development of the case studies will include asking families to track their health resource use over a 3-week period. This timeframe was chosen through consultations with clinicians and researchers, and while we recognize that health resource use can vary greatly amongst individuals with complex chronic conditions, we did not want study procedures to place unnecessary burdens on families. Families will be provided a health resource journal with a draft template to follow. Within this diary, families will be encouraged to track encounters with resources and supports needed to provide care for and support the health of their child. This would include, but is not limited to, ambulatory care clinic visits, inpatient stays, acupuncture, physiotherapy visits, home care visits, emergency department visits, respite care, and dental visits. Families will also be prompted to track the care coordination activities they undertake (i.e. calling different clinics to arrange appointments on the same day). Additionally, we will ask study participants if they believe their 3-week timeframe was representative of their average

health resource use. A $50 gift certificate to either Superstore, Amazon, or Irving Gas will be provided to families in appreciation for their time.

Semi-structured interviews with care team members will be composed of 2-3 questions developed based upon the respective family interview to reflect specifically on the local context and the participating families. Interviews are anticipated to last 10-15 minutes over the phone or Zoom Video Conferencing System (Zoom, 2019). This data will be used to supplement the family-reported experience, providing more context to the identified case. A $10 coffee gift card will be provided for their time.

3.2.2.5. Data Analysis

All interviews will be transcribed verbatim and uploaded to NVivo 11 Qualitative Data Analysis Software (QRS International, 2018). Given the recognized need for theory-informed approaches to case study design (Crowe et al., 2011), all interviews will be coded using a deductive content analysis approach based on the COM-B (Capability, Opportunity, Motivation – Behaviour) theoretical model (Michie et al., 2011). The COM-B is a comprehensive framework created to explore the interactional factors that influence health behaviour (Michie et al., 2011). This will allow us to explore the use of health resources, why they might be the way they are, and what resources are required to meet the health needs of families. Further, this analysis approach could be used in future studies aiming to map study findings to the Behaviour Change Wheel to design a knowledge translation intervention (Michie et al., 2011). The COM-B will provide an initial theory-based coding scheme to deductively code qualitative findings. Two independent coders will code the three domains of capability, opportunity, and

motivation. Following this, an inductive coding analysis approach will occur within each domain to group similar statements. This will reveal the presence of contradictory and common themes throughout the data, while providing a theoretical foundation that can help better understand the phenomenon under investigation. Self-reported health resource data from the family health resource journaling will be examined using descriptive and frequency statistics (mean, median, range, percentage).

Interview and self-reported data for each case will undergo data triangulation to create a greater understanding of child and family experiences. All quantitative and qualitative data will be organized into a matrix table based on themes resulting from the interview and self-reported health resource data, to examine patterns of convergence and divergence within each case study (Crowe et al., 2011; Yin, 2014). Descriptive statistics will be used to describe the variables captured in the self-reported diaries. Each case will be analyzed separately to create an in-depth representation of their individual experiences. Member-checking will occur with the findings from each case by presenting the results back to the family to check that we captured their experiences accurately (Polit & Beck, 2012). After data analysis is completed for each case study, a cross-case analysis will occur to examine common themes and areas of divergence among cases (Yin, 2014). A matrix table will be created to display data from each individual case based on common and emergent themes (Yin, 2014). This matrix will reveal similarities or uniqueness among cases (Yin, 2014).

3.2.2.6. Anticipated Outputs

There are two main outputs from this phase of the research. First, we will have a rich description of the first-hand experiences of children with medical complexity

and their families, as well as their formal and informal health resource use. Second, we will use this family-reported data to compare it with findings from Phase 1 to develop a greater and more comprehensive understanding into the health resource use and health needs of children with medical complexity and their families.

3.2.3. Data Triangulation

The intent of data integration in an explanatory sequential design is to examine the extent in which the follow-up rich qualitative results connect or explain the initial quantitative data (Creswell & Plano Clark, 2018). To do this, we will triangulate Phase 1 and Phase 2 data using a joint display table to visually depict the quantitative and qualitative phases. This joint display will be organized based on a 'statistic-by-theme' framework, linking relevant and related health administrative data with the follow-up case study findings (Creswell & Clark, 2011). Using this data triangulation approach, we will be able to create a greater understanding of the population, health resource use, and health needs of children with medical complexity in the Canadian Maritime provinces.

3.3. RESULTS

Phase 1 and Phase 2 are in-progress. Findings from each phase of research and the integration of the two will be reported in full in 2022.

3.4. DISCUSSION

3.4.1. Principle Considerations

There is a disconnect between current the Canadian health care system and the needs of children with medical complexity and their families (Cohen et al., 2018). By combining both health administrative and family reported data, this study can unveil critical information about children with medical complexity and their families to health

researchers, clinicians, policy makers, administrators, and families themselves. Mixed methods research has been underutilized in the current literature surrounding pediatric complex care, leaving gaps in our understanding of the responsiveness of our health care resources caring for this vulnerable population. As such, this research study is designed to take a novel approach to the study of children with medical complexity and their health resource use, contributing to the advancement of this body of research. While the development of an intervention is beyond the scope of this research, the strong theoretical underpinning, methodology and methods used will ensure its findings can be used in future work to advocate for and inform the design of health policy and programs in the Canadian Maritimes for this population of children and families. As such, this research has the potential to improve the health care delivery, experiences, and outcomes for children with medical complexity and their families.

3.4.2. Limitations

The findings from this research should be considered with the following limitations in mind. A limitation to secondary data analysis is that the researcher can only work with the data originally collected and stored. Hospital data is collected primarily for administrative purposes and not specifically designed for research (Gordis, 2014). This can lead to incomplete or missing records and variability in diagnostic codes (Gordis, 2014). Further, although the inclusion criteria to identify the cohort of children with medical complexity has been used in previous studies (Arthur et al., 2018; Chan, Di Gennaro, et al., 2016; Coller, Kelly, et al., 2018; Ralston et al., 2015), there are limitations to relying solely on diagnostic codes. The use of ICD codes may result in patients with medical complexity not being captured or capturing those that would not fit

the definitional framework. Furthermore, we make the assumption that children with medical complexity will have received care at the pediatric tertiary care facility at least once during their initial or follow-up medical care. As such, our prevalence estimates may be slightly underrepresented given the possibility that some of these children may be seen and managed fully by their local/regional hospitals. We are also limited by the lack of sociodemographic variables, such as race and ethnicity, available in our health administrative datasets. We, however, strongly believe these are critical intersectional factors in the lives of these families and requires exploration in future work. Furthermore, not all community hospitals report to NACRS at the highest level resulting in potentially incomplete reporting related to emergency department encounters. It is also important to note that due to constraints across provincial data linkage, we chose to use Nova Scotia and their provincial health administrative databases as the exemplary province to explore health resource use for children with medical complexity. We recognize that health resource use as indicated by health administrative data may differ in Prince Edward Island and New Brunswick.

Although purposive sampling will be used for Phase 2 to explore results found during Phase 1, participants' opinions or experiences may not be shared by other families or care team members. This study will also be limited to the experiences of children from one pediatric tertiary care center serving children and families in three small provinces in Eastern Canada, which operates within a universal health care model. Other health centers may differ in the structure, programs, and care provision for children with medical complexity; thus, results may not be reflective of other families and sites.

3.5. CONCLUSION

Improvements in medical treatments and technologies will likely result in an increased population of children with complex conditions. It is critical that we begin to develop a greater understanding of the health resource use of this vulnerable population to more efficiently and effectively meet their health care needs. Results from this research will be an important step forward in designing patient-oriented health policies and programs to improve the experiences, health resource use, and health outcomes of children with medical complexity and their families.

Chapter 4: QUANTITATIVE PHASE ONE

The work presented in Chapter 4 is presented in manuscript format and prepared for submission to *Journal of Pediatric Health Care*. Authorship is as follows: Breneol, S., Curran, J.A, Montelpare, W., Stewart, S., Vine, J., Martin-Misener, R., Macdonald, M. Title: Children with Medical Complexity and their Health Resource use in the Canadian Maritimes: A Retrospective Cohort Study

Statement of manuscript contribution: S.B conceptualized this work in collaboration with JAC, WM, JV, SS, RMM, and MM. SB submitted all data access applications, analyzed the data, and drafted the manuscript. All authors contributed to revising the manuscript.

Disclaimer: Portions of the data used in this report were made available by Health Data Nova Scotia of Dalhousie University. Although this research analysis is based on data obtained from the Nova Scotia Department of Health and Wellness, the observations and opinions expressed are those of the authors and do not represent those of either Health Data Nova Scotia or the Department of Health and Wellness.

4.1. INTRODUCTION

Improved survival rates and management of individuals with complex chronic conditions has resulted from progressive developments in health knowledge, treatment, and technologies (Cohen et al., 2011). These advancements have contributed to an epidemiological shift in the pediatric population, with one report from the United States estimating that in the last 60 years there has been a 400% increase in children living with a chronic condition (Perrin et al., 2014). Amongst this cohort are children and youth with

the most complex medical, developmental, and mental health needs, who are often described as 'children with medical complexity' (Cohen et al., 2018; Cohen et al., 2011). As opposed to the adult population, where medical complexity is predominantly associated with a finite number of conditions, children and youth with medical complexity are a diverse cohort with varied pediatric conditions (Berry et al., 2015). Despite this heterogeneity, children and youth with medical complexity are distinguished by four predominant characteristics: 1) diagnosed or presumed complex chronic condition(s); 2) high health resource use; 3) family-identified needs; and 4) functional limitations (Cohen et al., 2011).

The body of literature surrounding pediatric complex care has been steadily rising since the early 2000s (Cohen et al., 2018). Previous work has suggested that while children with medical complexity may account for less than 1% of the pediatric population, they require a disproportionate quantity of health resources to support their care needs (Berry, Hall, et al., 2013; Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). Evidence also suggests they are at greater risk for recurrent hospital admissions, discontinuity in care coordination, multiple unmet health care needs, and disturbed family functioning (Berry et al., 2017; Kuo et al., 2011; Kuo et al., 2014; O'Mahony et al., 2013; Simon et al., 2010). These findings highlight the need for more concentrated efforts to identify resource gaps and develop tailored strategies to improve the health and well-being of children and youth with medical complexity and their families.

Much of our current knowledge base about the population of children with medical complexity is from studies conducted in the United States (US), with limited

exploration in the Canadian context. While geographically close, there are contextual differences between the two countries, with one of the most predominant being that Canada has universal health insurance coverage (Tikkanen et al., 2020). To date, there have only been two reports published in Canada exploring the prevalence and health resource use of children and youth with medical complexity (Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). Cohen et al. (2012) were one of the first teams to publish a Canadian epidemiological study on this population. However, this work was focused in one Canadian province and was published over a decade ago (Cohen, Berry, et al., 2012). A second report from the Canadian Institutes of Health Information (CIHI) was recently released providing the first pan-Canadian report on the prevalence and health service use of children and youth with medical complexity (Canadian Institute for Health Information, 2020). While this report is an important step towards generating critical knowledge about this pediatric cohort in Canada, further provincial and regional work is warranted to better understand variation in health resource use, needs, and resource availability. This evidence is critical to informing health planning, policy development, program expansion and practice change.

The Canadian Maritimes, composed of Nova Scotia (NS), Prince Edward Island (PEI), and New Brunswick (NB), provides a unique jurisdictional context for this type of research. There is only one pediatric tertiary care facility, located in NS, responsible for the care of children, youth, and women in all three provinces. A deeper understanding of this population is needed to inform the development of relevant and evidence-informed recommendations for health policy, practice, and research. With the limited number of published works using Canadian-based health administrative data to explore this

vulnerable population, we sought to address the following research questions: 1) What is the prevalence of medical complexity among children in the Canadian Maritimes and what are their clinical characteristics?; and 2) What is the health resource use according to the health administrative data for children with medical complexity in Nova Scotia?

This work is the first phase of a larger mixed-methods study seeking to develop a greater understanding of the population of children with medical complexity in the Canadian Maritimes (Breneol, Curran, et al., 2022). The qualitative and data-integration phases will be reported in subsequent publications.

4.2. METHODS

4.2.1. Study design and data sources

This work involved a secondary data analysis of routinely collected health administrative data. Data sources included: the pediatric tertiary care facility's discharge data, the National Ambulatory Care Reporting System (NACRS), Physician Billings (MED), The CIHI Discharge Abstract Database (CIHI-DAD), and Vital Statistics – Death (VTIAL). A complete description of study methods can be found elsewhere (Breneol, Curran, et al., 2022). Ethics approval was obtained from the IWK Research Ethics Board (#1026835).

4.2.2. Study Population and Identification

All children between 0-18 years of age who were discharged from the pediatric tertiary care facility between April 1st 2004 and March 31st 2014 were considered for inclusion in the study cohort. This data source was chosen to identify our Maritime cohort as pediatric tertiary care centres are often the main care site for children with medical complexity (Berry et al., 2015; Simon et al., 2010). As existing literature

suggests that this population is relatively small in numbers, a 10-year timespan was chosen to ensure a suitable cohort size to power statistical analyses and provide a sufficient time-span to explore up to 5 years of health resource use data (up to March 31st 2019). The Pediatric Medical Complexity Algorithm (PMCA) 3.0 leverages the use of International Classification of Diseases (ICD) Codes to classify children based on their level of complexity and has a very good sensitivity (86%) for identifying children with complex chronic conditions in health administrative data (Simon et al., 2018). The more conservative version of this algorithm was applied to the tertiary care facility's discharge dataset. All children who were classified by this algorithm as having a complex chronic condition in the discharge data were included in the final cohort. Index date was defined as the first discharge date with a complex chronic condition as indicated by the PMCA. The PMCA further categorizes complex chronic conditions as progressive conditions [defined as "deteriorating health and an increased risk of shorter life expectancy in adulthood" (Simon et al., 2014, p. 1649)], malignancies, and body system involvement flags (i.e. cardiac, metabolic, genetic). Body system flags were grouped into the involvement of 1 body system, 2 body systems, and >2 body systems. This stratification will help illuminate the clinical characteristics of this cohort. It is important to note that while malignant and progressive conditions are mutually exclusive categories in the PMCA, the categorizations of these conditions and body system flags are not mutually exclusive. For example, an individual may be classified as having a progressive condition, while also having 1 or more body system flags. Further, an individual may have a malignant condition and no associated body system flag. Please see Appendix E for examples of clinical diagnoses included in the PMCA.

Deviations from the original protocol included the inability to determine the use of technological devices (i.e. tracheostomy, central venous devices) (Breneol, Curran, et al., 2022). We were unable to identify this concept as the PMCA omitted this characteristic flag as it did not improve the algorithm's ability to identify children with complex chronic conditions (Simon et al., 2014). Further, we omitted the time-to-event analysis due to scope/resource limitations and this will be explored in future work.

4.2.3. Study procedures

Health card numbers for all children/youth meeting our inclusion criteria were retrieved from the pediatric tertiary care facility's discharge data. This health card number was then encrypted by a health system partner and sent to the provincial data repository organization to examine health resource use. Encrypted health card numbers were then used to link the NACRS, MED, CIHI-DAD, and VITAL datasets. Using age, sex, and urban/rural location, a 3:1 matched control cohort was identified using the provincial MED dataset. This control cohort was used to provide a description of health resource use for children without medical complexity and to control for potential confounders. Health resource use was followed from an individual's index date for up to 5 years, or until 18 years of age.

4.2.4. Measures

Variables related to child characteristics for the Maritime cohort included age (categorized as <1; 1-4; 5-9; 10-13; 14-18 years old), sex assigned at birth, urban/rural home community, province of residency, and clinical presentation (categorized as progressive, malignancy, 1 body system, 2 body systems, and >2 body systems). Urban/rural residence was determined by the second character of the postal code, with '0'

indicating a rural area and all else being urban (Health Data Nova Scotia, n.d.). Age was categorized in alignment with previous Canadian work exploring this population (Cohen, Berry, et al., 2012). Given limitations in cross-province data linkages, health resource use was only examined in Nova Scotia. This province was chosen as it has the largest provincial pediatric population and is the location of the only pediatric tertiary care facility in the Maritimes (Statistics Canada, 2017). Variables related to health resource use spanned across both tertiary and community care sites and included hospital admissions, outpatient visits, home care services, emergency department visits, length of stay, and care provider specialties.

4.2.5. Data Analysis

To address our first research question, the prevalence of children with medical complexity in the Maritimes was determined by dividing the number of cases of children with medical complexity in our cohort by the number of children 0-18 years old reported by the Statistics Canada Census Data for Nova Scotia (2016) (Statistics Canada, 2017). Prevalence was then stratified by age, sex, urban/rural home community, province of residency, and clinical presentation.

To address our second research question, health resource use was examined and reported using descriptive statistics. This included number of distinct provider specialties, emergency department visits, inpatient admissions length of stay, outpatient visits, home care services, and admissions via ambulance. Rates of health resource use were further stratified based on the previously identified demographics. Odds ratios were calculated to determine the extent of variation in health resource use between children with medical complexity and children without medical complexity. We also sought to explore

associations between child characteristics and health resource use. A negative binomial regression was used to model our count variables and was chosen given the overdispersion of the data. Primary outcomes of interest included counts of hospital admissions, emergency department visits, primary care (community-based) visits, and hospital-based clinic visits. Predictors of interest included age, urban/rural residency, sex, and medical complexity status (yes/no). STATA version 9.3 (STATA, 2018) was used for all data analysis. Any missing, outlier, and implausible data points were examined thoroughly. No missing data were identified, but implausible data were examined closely to determine potential causes (i.e. true outliers, clerical errors). In the instances where values were implausible and logical typos could not be identified, analysis was restricted to individuals who had complete and plausible data.

4.3. RESULTS

A total of 91,485 records were included in the pediatric tertiary care facility's discharge data during our defined study period. Following the application of the PMCA, 3058 children/youth were identified as having a complex chronic condition. Further stratified by province, there were 1976 individuals from NS, 859 from NB, and 223 from PEI (See Table 4-1)

Table 4-1. Children with Medical Complexity in the Maritimes Demographic Data (2004-2014)

	Total		Progressive Condition		Malignancy		1 Body System		2 Body Systems		>2 Body Systems		Mental Health	
	N	%	N	%	N	%	N	%	N	%	N	%	N	%
Total	3058	100	2513	82	533	17	1790	59	722	24	248	8.1	375	12
Province														
NS	1976	65	1661	66	279	52	1204	67	468	65	146	59	314	84
NB	859	28	666	24	213	40	464	26	199	28	78	31	51	14
PEI	223	7.3	186	7.4	41	7.7	122	6.8	55	7.6	24	9.7	10	2.7
Age														
<1 years	980	32	918	37	39	7.3	553	31	247	38	129	52	12	3.2
1-4 years	559	18	407	16	159	30	304	16	134	19	44	18	53	14
5-9 years	468	15	341	14	140	26	300	17	74	10	16	6.5	24	6.4
10-13 years	429	14	339	13	86	16	241	13	109	15	27	11	72	19
14-18 years	622	20	508	20	109	20	392	22	131	18	32	13	214	57
Sex														
Female	1487	49	1220	49	261	49	888	50	348	48	107	43	244	65
Male	1571	51	1293	51	272	51	902	50	374	52	141	57	131	35
Residency														
Urban	2407	79	1955	78	439	82	1413	79	556	77	194	78	294	78
Rural	651	21	558	22	94	18	377	21	116	23	54	22	81	22

4.3.1. Prevalence and Clinical Characteristics

Between 2004-2014, the crude rate of children and youth with medical complexity in the Maritimes was 882 per 100,000 children and youth (See Table 4-2). When stratified by province, the crude rate of medical complexity was 1133 per 100,000 children and youth in NS, 600 per 100,000 in NB, and 756 per 100,000 in PEI. Infants under the age of 1 represented approximately one third of children with medical complexity (32%). Youth between the ages of 14-18 years comprised the next largest age cohort, representing an estimated 1 in 5 children and youth with medical complexity (20%). The remaining age groups were relatively evenly distributed, with those between the ages of 1-4 years representing 18%, 5-9 years representing 15%, and 10-13 years representing 14%.

The distribution of children according to sex assigned at birth was relatively even, with 51% male and 49% female. This remained relatively consistent across demographic stratification, except for mental health conditions (65% females). Overall, children and youth with medical complexity in the Maritimes lived in urban areas (78%). There was, however, a higher percentage of children and youth living in urban areas in NS (73%) in comparison to PEI (47%). Due to the way that NB assigns postal codes we were unable to identify urban and rural residencies in that province. We estimated the geographical distribution of the NB cohort using extrapolated (mean) findings from NS and PEI (See Table 4-3).

Most children and youth with medical complexity were categorized as having a progressive chronic condition (82%). Furthermore, just over half of these children and youth had a single body system involved (58%), 23% had two body systems involved,

and 8.1% had greater than two body systems involved. Approximately 16% of the identified cohort were flagged as having a malignancy. About half of individuals with more than two body systems contributing to medical complexity were under the age of 1 at their index date. When stratified by body system involvement, the PMCA flagged neurologic (28%), cardiac (21%), and mental health (12%) as the top three body system flags. Mental health related conditions were noted to be highest amongst youth between the ages of 14-18 (57%) (See Tables 4-2 and 4-4).

Table 4-2. Rate of Children with Medical Complexity per 100,000 Children and Youth

Jurisdiction	Total		Progressive		Malignancy		1 Body System		2 Body Systems		>2 Body Systems	
	N	Rate	N	Rate	N	Rate	N	Rate	N	Rate	N	Rate
Maritimes (Total)	3058	882	2513	725	533	154	1790	516	722	208	248	72
NS	1975	1133	1661	953	279	160	1204	691	468	269	146	84
NB	859	600	666	466	213	149	464	325	199	139	78	55
PEI	223	756	186	631	41	139	122	414	55	186	24	81

Table 4-3. Urban/Rural Community Distribution of Children with Medical Complexity by Province

Location	Total		NS		NB		PEI	
	N	%	N	%	N	%	N	%
Urban	2407	79	1444	73	N/A	63*	105	47
Rural	651	21	532	27	N/A	37*	118	53

*Extrapolated findings from PEI and NS distribution

Table 4-4. Counts and Proportion of Children with Medical Complexity by Body System

Clinical Presentation		*Maritimes (Total) (n=3058)*	*NS (n=1975)*	*NB (n=859)*	*PEI (n=223)*
Progressive	N	2513	1661	666	186
	%	82	84	78	83
Malignancy	N	533	279	213	41
	%	18	14	25	18
Cardiac	N	645	375	209	61
	%	21	19	24	27
Craniofacial	N	31	19	7	5
	%	1	0.94	0.81	2.2
Dermatologic	N	14	11	N/A*	N/A*
	%	0.46	0.56	N/A*	N/A*
Endocrinology	N	67	51	12-15*	<5*
	%	2.2	2.6	1.8	N/A*
Gastrointestinal	N	145	86	49	10
	%	4.7	4.4	5.7	4.5
Genetic	N	270	167	82	21
	%	8.8	8.5	9.6	9.4
Genitourinary	N	131	83	34	14
	%	4.3	4	4	6.3
Hematologic	N	206	119	70	17
	%	6.7	6	8.2	7.6
Immunologic	N	251	161	73	17
	%	8.2	8.2	8.5	7.6
Mental Health	N	375	314	51	10
	%	12	16	5.9	4.5

Clinical Presentation		*Maritimes (Total) (n=3058)*	*NS (n=1975)*	*NB (n=859)*	*PEI (n=223)*
Metabolic	N	220	161	41	18
	%	7.2	8.2	4.8	0.45
Musculoskeletal	N	262	150	88	24
	%	8.6	7.6	10	11
Neurologic	N	869	553	241	75
	%	28	28	28	34
Ophthalmologic	N	33	20	9-12	<5*
	%	1.1	1	N/A*	N/A*
Otologic	N	26	19	3-6	<5*
	%	0.85	0.96	0.7	N/A*
Pulmonary/Respiratory	N	339	236	84	19
	%	11	12	9.8	8.5
Renal	N	193	116	61	16
	%	6.3	5.9	7.1	7.2

*Small cell sizes are not reported to protect patient confidentiality and anonymity

4.3.2. Health Resource Use

Following the data linkage procedures, 1943 NS individuals were eligible (had a valid and linkable NS health card number) to be included in the final case cohort of children with medical complexity to examine their health resource use. A subsequent 5,829 matched controls (3:1 match) were identified through the MED database. See Tables 4-5 - 4-8 for full results.

4.3.3. Number of Specialty Care Providers

Children with medical complexity (n= 1943) had a median of 6 distinct specialists involved in their care. This was in comparison to children without medical complexity who saw a median of 2 types of provider specialties over the follow-up period.

4.3.4. Hospital Discharges

During the 5-year follow-up post index discharge, almost all children with medical complexity 99% (1929/1943) had a subsequent hospital admission. This was in comparison to only 17% (997/5829) of the control cohort. The odds of children with medical complexity having an inpatient admission were 668 times greater in comparison to children without medical complexity [CI: 391.51, 1130.03]. Children with medical complexity had a median of 2 hospital discharges, with a range of 0-55 and an average length of stay of 4 days. Rates stayed relatively similar across clinical demographics, except for individuals with a malignancy diagnosis (median = 6) or having more than 2 body systems involved (median = 4), where the median number of discharges appeared to slightly rise.

4.3.5. Emergency Department Visits

Children with medical complexity had a median of 2 emergency department visits during the follow-up period (range = 0-62), whereas their matched counterparts had a median of 0 (range = 0-34). Further, 74% of the case group and 49% of the control group had a recorded emergency department visit. The odds of having an emergency department visit were 2.97 times higher among children with medical complexity in comparison to children without medical complexity [CI: 2.64, 3.35].

4.3.6. Outpatient Visits

Approximately 97% of children with medical complexity had a primary care visit and 74% had a hospital-based clinic visit. Within the control group, the proportion was 87% and 34%, respectively. Across the sample of children with medical complexity, we observed a median of 34 primary care visits and 3 hospital clinic visits. In comparison, the control group had a median of 10 primary care visits and 0 hospital clinic visits. We further observed the odds of having a primary care visit to be 4.26 times higher amongst children with medical complexity than the control group [CI: 2.46, 3.39], and the odds of having a hospital clinic encounter to be 8.3 times higher [7.39, 9.39].

4.3.7. Home Care Services

Overall, only 3% of our sample of children with medical complexity had a physician or nurse practitioner home care visit. The control group had less than 1%. Despite this small number, the odds of children with medical complexity having a home care visit from a nurse practitioner/physician were 6.64 times higher than their matched controls [4.14, 10.59].

Table 4-5. Descriptive Statistics of Health Resource Use for Children with Medical Complexity Over a 5-Year Period

	Percentiles						
Cases	Min	25.00%	50.00%	75.00%	Max	Mean	SD
Distinct Provider Specialties	0	4	6	8	18	5.89	3.04
Emergency Department Visits	0	0	2	5	62	3.85	5.64
Hospital Discharges	0	1	2	5	55	4.09	5.29
Length of Stay (Days)	0	2	4	9	1011	11.36	30.028
Outpatient Visits (Total)	0	20	39	65	331	47.78	39.41
Hospital Clinic Visits	0	0	3	9	118	6.81	10.35
Primary Care Visits	0	18	34	55	272	40.97	32.78
Home Care Visits	0	0	0	0	35	0.1	1.01
Pediatrician Visits	0	2	10	24	234	17.31	22.55
Admit Via Ambulance	0	0	0	0	30	0.48	1.41
Controls	Min	25.00%	50.00%	75.00%	Max	Mean	SD
Distinct Provider Specialties	0	1	2	3	17	2.17	1.66
Emergency Department Visits	0	0	0	2	34	1.37	2.39
Hospital Discharges	0	0	0	0	12	0.23	5.29
Length of Stay (Days)	0	1	2	4	275	4.19	11.08
Outpatient Visits (Total)	0	4	11	21	120	14.33	14.18
Hospital Clinic Visits	0	0	0	1	29	1.21	2.8
Primary Care Visits	0	3	10	19	109	13.12	13.19
Home Care Visits	0	0	0	0	13	0.01	0.21
Pediatrician Visits	0	0	0	0	48	1.13	3.38
Admit Via Ambulance	0	0	0	0	7	0.02	0.19

Table 4-6. Distribution of Provider Specialty Across All Encounters for Children with Medical Complexity

Provider Billing Specialty	Percent
Pediatrics	50
General Practitioner	24
Anaesthetist	6.4
General Surgery	2.4
Hematology	2.3
Ophthalmology	2.2
Orthopaedic Surgery	2.2
Diagnostic Radiology	2.1
Otolaryngology	1.8
Neurosurgery	1.1
Urology	0.89
Neurology	0.56
Optometry	0.48
Dermatology	0.41
Psychiatry	0.38
Plastic Surgery	0.36
Radiation Oncology	0.33
Internal Medicine	0.28
Obstetrics & Gynecology	0.24
Emergency Medicine	0.23

Table 4-7. Health Resource Use Odds Ratios Between Children with Medical Complexity (Cases) and Children without Medical Complexity (Controls). All differences are significant with a p-value <0.001

	Percentiles			
	Cases (n=1943)	Controls (n=5829)	OR	95% CI
Hospital Discharges	1929	997	667.78	391.51, 1130.03
Intensive Care Admissions	276	33	28.94	20.09, 42.1
Neonatal Intensive Care Admissions	268	53	17.39	13.07, 23.34
Emergency Department Visits	1435	2840	2.97	2.64, 3.35
Outpatient Visits (Total)	1883	5109	4.42	3.39, 5.81
Hospital Outpatient Visits	1435	1985	8.3	7.39, 9.39
Community Office Visits	1877	5070	4.26	3.32, 5.47
Home Care Visits	54	25	6.64	4.14, 10.59
Primary Care Visits	1764	4970	2.89	2.46, 3.39
Pediatrician Visits	1537	1376	12.25	10.91, 13.87
Admit Via Ambulance	482	107	17.64	2.65, 3.09

Table 4-8. Health Resource Use (Median Rate) Among Children and Youth with Medical Complexity, By Type of Medical Complexity, in the 5-Years Post Index Discharge

Health System Use (Median)	Total Cohort (n)	Progressive (n)	Malignancy (n)	1 Body System (n)	2 Body Systems (n)	2> Body Systems (n)
Inpatient Discharges (Median)	2 (n=1943)	2 (n=1634)	6 (n=273)	2 (n=1183)	3 (n=459)	4 (n=144)
Sex						
Female	2 (n=957)	2 (n=812)	5 (n=128)	2 (n=596)	2 (n=231)	4 (n=57)
Male	2 (n=986)	2 (n=822)	6 (n=145)	2 (n=587)	3 (n=228)	4 (n=87)
Age						
>1 years	2 (n=620)	2 (n=586)	2 (n=23)	2 (n=350)	2 (n=175)	4 (n=82)
1-4 years	3 (n=339)	3 (n=255)	7 (n=84)	2.5 (n=192)	4 (n=83)	6 (n=25)
5-9 years	2 (n=269)	2 (n=203)	6 (n=67)	2 (n=181)	3 (n=45)	12 (n=5)
10-13 years	2 (n=263)	2 (n=213)	5 (n=41)	2 (n=158)	2.5 (n=64)	3 (n=11)
14-18 years	2 (n=452)	2 (n=377)	5 (n=58)	1 (n=302)	2 (n=92)	2 (n=21)
Residency						
Urban	2 (n=1422)	2 (n=1193)	5 (n=199)	2 (n=881)	2 (n=326)	3 (n=104)
Rural	2 (n=521)	2 (n=441)	7 (n=74)	2 (n=302)	3 (n=133)	5 (n=40)
Emergency Department Visits (Median)	2 (n=1943)	2 (n=1634)	2 (n=273)	2 (n=1183)	2 (n=459)	3 (n=144)
Sex						
Female	2 (n=957)	2 (n=812)	2 (n=145)	2 (n=569)	2 (n=231)	3 (n=57)
Male	2 (n=986)	2 (n=822)	2 (n=128)	2 (n=587)	2 (n=228)	3 (n=87)
Age						
<1years	3 (n=620)	3 (n=586)	3 (n=23)	3 (n=350)	2 (n=175)	2 (n=82)
1-4 years	4 (n=339)	3 (n=255)	3 (n=84)	3 (n=192)	4 (n=83)	5 (n=25)
5-9 years	2 (n=269)	1 (n=203)	2 (n=67)	1 (n=181)	2 (n=45)	13 (n=5)

Table 4-8. Health Resource Use (Median Rate) Among Children and Youth with Medical Complexity, By Type of Medical Complexity, in the 5-Years Post Index Discharge

Health System Use (Median)	Total Cohort (n)	Progressive (n)	Malignancy (n)	1 Body System (n)	2 Body Systems (n)	2> Body Systems (n)
10-13 years	1 (n=263)	1 (n=213)	1 (n=41)	1 (n=158)	1 (n=64)	3 (n=11)
14-18 years	1 (n=452)	1 (n=377)	1 (n=58)	1 (n=302)	1 (n=92)	1 (n=21)
Residency						
Urban	2 (n=1422)	1 (n=1193)	2 (n=199)	2 (n=881)	2 (n=326)	4 (n=104)
Rural	1 (n=521)	2 (n=441)	1 (n=74)	2 (n=302)	1 (n=133)	1 (n=40)
Community Outpatient Visits (Median)	34 (n=1943)	32 (n=1634)	61 (n=273)	29 (n=1183)	37 (n=459)	48 (n=144)
Sex						
Female	33 (n=957)	29 (n=812)	59 (n=128)	26 (n=596)	39 (n=231)	50 (n=57)
Male	35 (n=986)	34 (n=822)	67 (n=145)	33 (n=587)	36 (n=228)	42 (n=87)
Age						
<1years	43 (n=620)	42 (n=586)	49 (n=23)	38 (n=350)	45 (n=175)	54 (n=82)
1-4 years	47 (n=339)	43 (n=255)	83 (n=84)	44 (n=192)	42 (n=83)	43 (n=25)
5-9 years	34 (n=269)	30 (n=203)	74 (n=67)	30 (n=181)	33 (n=45)	81 (n=5)
10-13 years	29 (n=263)	26 (n=213)	54 (n=41)	25 (n=158)	29.5 (n=64)	43 (n=11)
14-18 years	17 (n=452)	16 (n=377)	39.5 (n=58)	15.5 (n=302)	18.5 (n=92)	21 (n=21)
Residency						
Urban	35 (n=1422)	32 (n=1193)	63 (n=199)	29 (n=881)	39 (n=329)	47 (n=104)
Rural	33 (n=521)	31 (n=441)	58.5 (n=74)	27 (n=302)	35 (n=133)	40.5 (n=40)
Hospital Outpatient Visits (Median)	3 (n=1943)	2 (n=1634)	12 (n=273)	2 (n=1183)	4 (n=459)	6 (n=114)
Sex						
Female	3 (n=957)	2 (n=812)	12 (n=12)	2 (n=596)	4 (n=231)	7 (n=57)

Table 4-8. Health Resource Use (Median Rate) Among Children and Youth with Medical Complexity, By Type of Medical Complexity, in the 5-Years Post Index Discharge

Health System Use (Median)	Total Cohort (n)	Progressive (n)	Malignancy (n)	1 Body System (n)	2 Body Systems (n)	2> Body Systems (n)
Male	3 (n=986)	3 (n=822)	13 (n=145)	2 (n=587)	4 (n=228)	5 (n=87)
Age						
<1years	3 (n=620)	3 (n=586)	7 (n=23)	2 (n=350)	4 (n=175)	4 (n=82)
1-4 years	6 (n=339)	4 (n=255)	21.5 (n=84)	3 (n=192)	9 (n=83)	14 (n=25)
5-9 years	3 (n=269)	3 (n=203)	19 (n=67)	3 (n=181)	4 (n=45)	20 (n=5)
10-13 years	3 (n=263)	2 (n=213)	10 (n=41)	2 (n=158)	3 (n=64)	7 (n=11)
14-18 years	1 (n=452)	1 (n=377)	5 (n=58)	1 (n=302)	1.5 (n=92)	6 (n=21)
Residency						
Urban	2 (n=1422)	2 (n=1193)	11 (n=199)	2 (n=881)	3 (n=326)	5 (n=104)
Rural	5 (n=521)	4 (n=441)	14 (n=74)	4 (n=302)	5 (n=133)	8 (n=40)

4.3.8. Association Between Child Characteristics and Health Resource Use

Following a review for implausible values between index and death dates to account for length of follow-up in our models, two control subjects were dropped from the regression analysis for a total of 7770 individuals across case and control cohorts (See Table 4-9). Cases were significantly more likely to have a higher incidence rate ratio (IRR) than controls, from an IRR of 23.5 (95% CI: [21.6, 25.6]) for hospitalizations to an IRR of 3.1 (95% CI: [2.9, 3.4]) for emergency department visits.

Across all outcome predictors explored in this study, being above the age of 1 was associated with a lower IRR of health resource use in comparison to those under 1 year of age. All models showed a similar U-shaped IRR pattern in age, with children under 1 having the highest IRR, then decreasing for the following three age groups (1-4, 5-9, 10-13), before increasing again in the 14-18 age group.

Urban and rural communities also appeared to play a significant role in the IRR of hospital discharges (IRR=1.2, 95% CI: [1.1, 1.3]), emergency department visits (IRR=0.84, 95% CI: [0.78-0.90), hospital clinic visits (IRR=2.32, 95% CI: [2.11-2.54]), and primary care visits (IRR=0.89, 95% CI: [0.85-0.94]). Notably, the rate of hospital clinic visits among those living in a rural area was 2.32 times higher than the rate of those living in an urban area [CI: 2.11-2.54].

There was no major effect of sex in any of the models. For emergency department (IRR=1.1, 95% CI: [1.03, 1.18]) and primary care (IRR=1.08, 95% CI: [1.03, 1.13]) visits there was significant effect of sex, but the effect size was minor.

Table 4-9: Negative Binominal Regression of Four Health Resource Outcome Variables Among Children with Medical Complexity (n=1943) and Children without Medical Complexity (n=5827) in Nova Scotia

	Outcome Variable: Hospital Discharges			
	IRR	Std. Error	P>|z|	95% CI
Age				
1-4 years	0.6015	0.0361	<0.001	0.5348-0.6765
5-9 years	0.4661	0.0312	<0.001	0.4088-0.5314
10-13 years	0.5592	0.0370	<0.001	0.4911-0.6366
14-18 years	0.8083	0.0478	<0.001	0.7199-0.9076
Rural	1.222	0.0546	<0.001	1.1195-1.3339
Female	0.9670	0.0391	0.407	0.8932-1.0468
Case Group	23.541	1.0075	<0.001	21.6472-25.6011
# of Obs*	7770			
Pseudo R2	0.2245			
	Outcome Variable: Emergency Department Visits			
	IRR	Std. Error	P>|z|	95% CI
Age				
1-4 years	0.7605	0.0375	<0.001	0.6905-0.8376
5-9 years	0.4936	0.0275	<0.001	0.4426-0.5504
10-13 years	0.5868	0.0325	<0.001	0.5263-0.6541
14-18 years	0.8007	0.0392	<0.001	0.7274-0.8814
Rural	0.8369	0.0328	<0.001	0.7750-0.9037
Female	1.1056	0.0385	0.004	1.0327-1.1837
Case Group	3.1374	0.1191	<0.001	2.9125-3.3798
# of Obs*	7770			
Pseudo R2	0.0383			
	Outcome Variable: Hospital Clinic Visits			
	IRR	Std. Error	P>|z|	95% CI
Age				
1-4 years	1.0550	0.0659	0.392	0.9334-1.1923
5-9 years	0.8970	0.0607	0.108	0.7855-1.0242
10-13 years	0.8959	0.0615	0.110	0.7830-1.0250

	Outcome Variable: Hospital Discharges			
	IRR	Std. Error	P>|z|	95% CI
14-18 years	0.9085	0.0559	0.119	0.8053-1.0249
Rural	2.3182	0.1096	<0.001	2.1130-2.5434
Female	0.9371	0.0406	0.134	0.8607-1.0202
Case Group	6.8171	0.3244	<0.001	6.2100-7.4836
# of Obs*	7770			
Pseudo R2	0.0626			
	Outcome Variable: Primary Care Visits (Community-Based)			
	IRR	Std. Error	P>|z|	95% CI
Age				
1-4 years	0.7497	0.0242	<0.001	0.7038-0.7987
5-9 years	0.5808	0.0204	<0.001	0.5422-0.6221
10-13 years	0.6022	0.0213	<0.001	0.5618-0.6455
14-18 years	0.7484	0.0230	<0.001	0.7045-0.7945
Rural	0.8924	0.0221	<0.001	0.8500-0.9367
Female	1.0805	0.0241	0.001	1.0343-1.1288
Case Group	3.4895	0.0874	<0.001	3.3223-3.6651
# of Obs*	7770			
Pseudo R2	0.0418			

4.4. DISCUSSION

This study suggests that children with medical complexity in the Maritimes represent less than 1% of the pediatric population but use a disproportionate amount of health resources in comparison to children without medical complexity. This work further contributes to the pediatric complex care literature by providing an understanding into health resource use amongst children with medical complexity over a 5-year period in comparison to children without medical complexity. We observed a higher rate of encounters for children with medical complexity across primary, hospital, and emergency care settings. The results of this study have various implications for pediatric complex care practice, policy, and research.

Over 80% of our identified cohort of children with medical complexity had a progressive chronic condition, with approximately one third being under the age of 1 year. These conditions are associated with a decline in health status over time "and an increased risk of shorter life expectancy in adulthood" (Simon et al., 2014, p. 1649). Recent research has also indicated the improved survival rates and treatments for various pediatric complex chronic conditions (Fraser et al., 2021; Perrin et al., 2014). These findings hold importance when considering resource allocation and policy development from birth to adult care. Particularly, the transition between pediatric to adult care can become challenging given the number of providers involved in the care of these children (up to 18 different provider specialties) and the evident need for comprehensive care coordination (Canadian Association of Pediatric Health Centres, 2016). There is also no direct equivalent to a pediatrician in the adult sector, which may result in fragmented care across providers. Primary care practitioners play an important role in facilitating care

across the continuum; however, there is a paucity of literature exploring the role of primary care in supporting these children and their families during this transition in care (Kerr et al., 2017). This being said, the facilitation and development of trusting relationships between providers and families in both adult and pediatric sectors has been shown to support a seamless transfer of care (Doucet et al., 2022; Weissberg-Benchell & Shapiro, 2017). A recent scoping review examining programs to support the transition from pediatric to adult care also suggested the need for improved awareness and training for pediatric providers in this process (Doucet et al., 2022). More evaluation and implementation research is required to identify and monitor the impact of key intervention components to support children with medical complexity during the transition from pediatric to adult care (Doucet et al., 2022; Pape & Ernst, 2022).

Children and youth with mental and behavioural health related diagnoses are an important, yet under-represented subgroup of children with medical complexity (Cohen et al., 2018). In this study, we identified mental health as the third most frequent body system flag indicated by the PMCA, comprising 12% of our case cohort. When further stratifying this cohort, our findings suggest that mental health conditions were most prominent amongst female youth between ages 14-18 (65%). Previous research has indicated increasing rates of hospitalizations for pediatric mental health disorders, with the highest rate of admissions among older children/youth (Bardach et al., 2014). Research from Brown et al. (2021) correspondingly noted increasing lengths of stay amongst children with medical complexity with a mental health condition. Gender differences can also be observed in health-seeking behaviour, with women-identifying youth seeking support for mental health concerns more often than men (Haavik et al.,

2019; Thompson et al., 2016). Given these findings, inpatient pediatric mental health is a national priority for quality improvement initiatives in the US (Bardach et al., 2014). While previous researchers have identified the need to explore the role of mental health conditions within the population of children with medical complexity (Canadian Institute for Health Information, 2020; Cohen et al., 2018), the present study is one of the first in Canada to explore this distribution. These findings highlight the need for clinicians and policy makers to be aware of this unique subpopulation and remain inclusive in eligibility criteria for complex care resources. Further, much research is still left to be done to explore the needs of children who specifically have complex mental and behavioural health related diagnoses.

Prior to this work, our team completed a scoping review exploring how health administrative data are informing practice, policy, and research recommendations for children with medical complexity and their families (Breneol et al., n.d.). Data extracted from included articles revealed a dearth of information related to potential variations in health resource use for those living in urban versus rural areas. This is an important consideration given the resource challenges to support pediatric health care in rural settings, some of which are modifiable (e.g., staffing) and some of which are not (e.g., distance from tertiary centres) (Graves et al., 2019). Our findings suggest that the community in which children and youth reside is an important factor in health resource use. Specifically, those living in rural communities were 2.32 times more likely to visit a hospital clinic and 1.22 times more likely to experience a hospital discharge than children from urban areas. Conversely, children from rural areas were less likely to have an emergency department encounter (IRR = 0.84) or a primary care visit (IRR = 0.89) than

those living in urban areas. While we cannot be certain of the factors influencing these observed patterns, potential explanations include: 1) Children in rural areas may attend hospital clinics for their care needs due to a potential lack of capacity amongst rural community providers; 2) rural community hospitals may have mechanisms in place for children to bypass the emergency department and proceed directly to relevant hospital units; 3) Rural communities may lack the resources to support the management of certain illness presentations on an outpatient basis; and 4) Some community hospitals provide respite care on in-patient units if resources allow. It is also important to note that not all community hospitals report to NACRS at the highest level, meaning that some emergency department visits may have gone uncaptured and is another potential factor influencing these observations. Inconsistent findings were noted within the literature in regards to urban-rural differences in pediatric health resource use and costs, with some studies suggesting higher costs (Graves et al., 2019) and others lower (McManus et al., 2016). Interestingly, however, one study did note that children from rural areas who experienced a hospitalization were readmitted more often, even after controlling for medical complexity (Peltz et al., 2016). This may suggest various underlying factors, including potential socioeconomic differences and/or a lack of home care and community-based supports. Additional research is needed to identify and examine potential underlying factors influencing urban-rural differences in health resource use. In the meantime, our findings are useful to program and policy makers, highlighting the need to explore and optimize resource availability and quality regardless of a child's home community.

The majority of children with medical complexity are discharged from hospital back to their homes under the care of their primary caregiver (i.e., biological parent, guardian, foster parent). As one expert stated, "the majority of pediatric health care is not delivered in a children's hospital or pediatric clinics, but rather in the child's home environment" (Gay, 2020, p. 1). The median length of stay in hospital for children with medical complexity (4 days) was double that compared to children without medical complexity (2 days) and had a range of 0-1011 days, in contrast to 0-275 days for the control group. Further, only 3% of our sample of children with medical complexity had a home care visit from a physician or nurse practitioner. Unfortunately, current data collection structures for pediatric home care services in the Maritimes do not provide a comprehensive picture of utilization, lacking information about publicly funded home care nursing, informal resources, and private services (Canadian Institutes of Health Information, 2020). Despite this, it is still crucial that we draw particular attention to this service provision. One study from the US identified a lack of home care nursing as being the top contributor to delayed discharges for children with medical complexity, leading to rising hospital stays and costs (Maynard et al., 2019). Home care is an irrefutable need for children with medical complexity and their families (Barone et al., 2020; Foster et al., 2019; Gay, 2020; Maynard et al., 2019). However, we know from expert clinicians, families, and researchers that home care provision for this population is not optimal, with challenges often related to staffing shortages and a lack of pediatric-specific capacity (Breneol, King, et al., 2022; Foster et al., 2019). Not only could the expansion of pediatric home care be one potential strategy to reduce the length of costly hospital admissions (Maynard et al., 2019), it could also provide families with crucial in-home

support as they navigate the demands of caregiving. Researchers need to direct their attention to exploring potential models of home care delivery to optimize family health and well-being. Decision makers and clinicians should also advocate for the strengthening of home health care provision, recognizing the potential positive benefits for both families and the health system.

This work provided a population-based overview of children with medical complexity that brought forth potential strategic directions for health initiatives. However, this work on its own is limited in its ability to comment on the full extent of resources being used to support the health and well-being of children with medical complexity and their families. While there is a recognized need for more comprehensive health datasets, current repositories do not contain enough substantive information on the range of health resources being used outside of the hospital setting, particularly related to community care, school-based services, and out-of-province therapies. Given that most of these children are cared for in their home communities, it is crucial that we expand our understanding into the true extent and type of resources supporting the overall health of this vulnerable population (Canadian Institute for Health Information, 2020). It is also critical to recognize that human experience cannot be reduced to singular data points.

Qualitative research methods (i.e. semi-structured interviews) may address some of the gaps in current health administrative datasets by generating and analysing data gathered by speaking directly to families. This type of qualitative work could help to illustrate the broad range of resources being used by families, as well as highlight their care needs. By combining health administrative data with richly descriptive qualitative findings, we can gain a much more detailed and contextual understanding of the patterns

of health resource use and health needs of children with medical complexity and their families. This current study is part of a larger explanatory sequential mixed methods project to generate such knowledge.

4.5. LIMITATIONS

Study findings should be considered in light of several potential limitations. Health administrative data are primarily collected for administrative purposes and as such researchers can only work with the data originally collected and stored (Gordis, 2014). Although the PMCA has been used in previous studies to identify children with medical complexity (Arthur et al., 2018; Coller, Klitzner, et al., 2018; Ralston et al., 2015), there are limitations to relying solely on diagnostic codes. Given the wide variation in clinical presentations and social/health factors that influence medical complexity, we may not have captured all potential children/youth that fit our conceptual definition. Current provincial datasets do not contain sociodemographic variables such as race and/or ethnicity. We, however, strongly advocate for the collection and storage of these critical intersectional factors and this should be a focus for future work. It is also important to note that there is only one pediatric tertiary care facility in the Maritimes and we worked under the assumption that children with medical complexity would have received care at this hospital at least once during their initial or follow-up care. With this said, our prevalence estimates may be underrepresented given the possibility that some of these children may be seen at their regional hospitals or referred to facilities in provinces outside of the Maritimes. This is of particular importance when interpreting the NB and PEI crude prevalence rates. For example, children living in more western/northern communities in NB may be referred to Quebec, and/or children living in PEI may not

experience an inpatient admission to the pediatric tertiary care facility. Families with children with medical complexity may also choose to move closer to the tertiary care facility given their medical needs, which may be a potential reason for the higher crude prevalence rate in NS.

4.6. CONCLUSION

Children with medical complexity are an important population in need of innovative health policy, practice, and research initiatives. While they are a small proportion of the pediatric population, we observed a higher likelihood of outpatient, hospital, and emergency care encounters for children with medical complexity in comparison to children without medical complexity. Potential strategic directions to improve the care for children with medical complexity may include focusing on comprehensive care coordination and supportive transitions in care. Future research is needed to better understand variation in resources use across urban and rural settings.

Chapter 5: Phase One Post-Script

The following section will provide more detailed information into the health administrative databases, variables, and analysis to examine the prevalence and health resource use in this retrospective study.

5.1. HEALTH ADMINISTRATIVE DATABASES

Five health care databases were accessed for this study: The pediatric tertiary care facility's discharge data, MSI Physician Billings (MED), National Ambulatory Care Reporting System (NACRS), Canadian Institute for Health Information Discharge Abstract Databases (CIHI-DAD), and VITAL Statistics – Death (VITAL).

The pediatric tertiary care facility's discharge data houses clinical and administrative information on within institutional encounters. This database was used for the identification of the study cohort and their associated demographics.

The MED captures information on physician and nurse practitioner billings. This database permits the exploration of provider specialty information, hospital unit where the care occurred, and when care occurred (Health Data Nova Scotia, 2019).

The NACRS database houses data for ambulatory care encounters for all individuals, excluding military personnel (Health Data Nova Scotia, 2019). This includes day surgeries, hospital outpatient clinics, community-based clinics, and emergency department visits. Please see Appendix D for a detailed document on the level of information hospitals in Nova Scotia report to NACRS.

The CIHI-DAD contains clinical and demographic information on all hospital discharges, deaths, and transfers (Health Data Nova Scotia, 2019). It is mandatory for all hospitals to report this information to the CIHI-DAD. As such, we were able to capture variables related to hospital discharges and length of stay occurring in hospitals across Nova Scotia (i.e. community and tertiary care hospitals) (Health Data Nova Scotia, 2019).

The VITAL database was used to identify the date of death (if occurred) amongst our cohort and was used to account for length of health resource use follow-up in our regression analyses.

5.2. THE PEDIATRIC MEDICAL COMPLEXITY ALGORITHM

The PMCA has two versions to identify children with medical complexity in health administrative data: the least and more conservative versions (Simon et al., 2014). The least conservative option classifies a child/youth as having a complex chronic condition if they have 1 provider claim for a progressive condition, malignancy or per body system for 2 different systems during the study period. The more conservative version requires 1 claim for a progressive condition or malignancy and 2 claims per body system for 2 different systems. Authors of the PMCA recommend using the least conservative option when accessing more comprehensive datasets that span hospital and community care sites, whereas they suggest the more conservative option for discharge data with 3 years of data. However, given the relatively small pediatric population in the Maritimes, we chose to use the 10-year time span to achieve a sample population large enough to stratify our cohort based on clinical characteristics and to power statistical analyses. The more conservative option was chosen in consultations with experts in

biostatistics and health administrative data as the best suited option for this study. This was due to the longer study period and the threshold of 2 claims per body system for 2 different body systems to be classified as having a complex chronic condition. (Simon et al., 2014, 2018). See Appendix E for examples of clinical conditions within each system.

5.3. VARIABLES OF INTEREST

Please see Appendix C for a full list of retrieved variables, data sources, and reasons to achieve the study aim.

Age. Age was determined through birth year and month extracted from the pediatric tertiary care facility's discharge data. To safeguard privacy and confidentiality, the day of the month was not retrieved. Infants under 1 year of age were categorized as such.

Sex. Sex was retrieved by the pediatric tertiary care facility's discharge data. Sex was defined as a male/female sex assigned at birth. Gender demographics are not included in current health administrative datasets.

Technological assistance. Cohen et al.'s (2011) definitional framework characterizes this cohort as often relying on technological assistance. However, we were unable to determine technological assistance using the PMCA. The PMCA removed technological assistance as one of their complexity indicators as it did not improve the algorithm's sensitivity or specificity (Simon et al., 2014).

Geographical location. The general geographical location of the child's home community was obtained from pediatric tertiary care facility's discharge data via postal code. A rural location was approximated based on the presence of a '0' in the second position within a postal code (this indicates a rural mail delivery route in Canada) (Health

Data Nova Scotia, n.d.). All other numbers were categorized as urban (Health Data Nova Scotia, n.d.).

Body system involvement. Details regarding the clinical characteristics of the population was determined through the PMCA algorithm which stratified the sample according to 17 primary body systems: craniofacial, dermatologic, cardiac, endocrinologic, gastrointestinal, genetic, genitourinary, hematologic, immunologic, mental health, metabolic, musculoskeletal, neurologic, ophthalmologic, otologic, pulmonary/respiratory, and renal. The PMCA also indicates if the condition is progressive and/or malignant (Simon et al., 2018). This information was provided by the pediatric tertiary care facility's discharge data.

Care team characteristics. Information on the type of providers caring for this cohort was obtained from the MED databases. Health care providers' specialties were obtained to determine the number and type of care team members providing inpatient and outpatient care to this cohort. We were unable to identify the involvement allied health providers or other professionals that could be involved in the care of children with medical complexity.

Hospital Discharges. Hospital discharges and length of stay were examined through the CIHI DAD. This included any discharge after their initial index date (determined as the first discharge the PMCA flagged as medical complexity).

Outpatient hospital use. Outpatient hospital use was examined using MED. Total outpatient visits were defined as any encounter with a location code of "OFFC" (office) or a hospital unit code of "OTPT" (outpatient). This was inclusive of nurse practitioner

visits. This was chosen as a proxy to represent any health visits happening within an outpatient setting. Hospital-based outpatient visits were defined as any encounter with a hospital unit code of "OTPT" (outpatient) and community-based primary care visits were identified by a location of code of "OFFC". Similarly, pediatrician office visits were defined as a location code of "OFFC" (office) and a provider billing specialty of "PEDI" (Pediatrician) (Health Data Nova Scotia, 2019),

Home care services. Home care services were derived by a location code indicating "HOME" (home) in the MED dataset. This denotes a provider billing for an encounter happening within the home setting and does not represent a complete picture of home care services.

Emergency department visits. Emergency department visits were examined through the NACRS and MED databases. Emergency department visits were derived using two datasets, MED and NACRS. The NACRS dataset only has data dating back to 2011. Prior to 2011, we explored the possibility that the CIHI DAD may have collected emergency department data, however they only captured day surgeries. As such, we chose to identify any encounters in the MED dataset indicating a location code of "EMERG" (Emergency), as well as any emergency department records in NARCS. If duplicate service dates were found between datasets, it was only counted as 1 emergency department encounter to avoid overestimation. These two indicators contributed to the final count of emergency department visits.

Ambulance Transfer. Information regarding where children are being transferred via ambulance to and from hospital encounters were gathered through the NARCS and CIHI DAD.

5.4. PREVALENCE RATE CALCULATIONS

As a forementioned, the prevalence of children with medical complexity was determined by dividing the total number cases by the number of children 0-18 years old reported by Statistics Canada Census Data for Nova Scotia in 2016. For transparency in calculations, the denominators for the pediatric population were as follows: Maritime = 346,755; Nova Scotia = 174,280; New Brunswick = 142,990; and Prince Edward Island = 29,485 (Statistics Canada, 2017).

5.5. MIXED METHODS: POST QUANTITATIVE SCRIPT

This study employed an explanatory mixed methods research design (Creswell & Plano Clark, 2018; Doyle et al., 2016; O'Cathain et al., 2008). In alignment with this methodology, the quantitative results (Phase 1) contributed to data collection measures in the subsequent qualitative study (Phase 2). Phase 1 results contributed the purposive sampling strategy and semi-structured interview guide used to build the family case studies in Phase 2. The following section provides greater detail into the sampling strategy, recruitment strategies, and interview guide development used to expand our understanding of health resource use and care needs for children with medical complexity and their families in the Maritimes.

5.6. SAMPLING STRATEGY: CHILDREN WITH MEDICAL COMPLEXITY AND THEIR FAMILIES

Phase 1 results suggested that children with medical complexity are indeed heterogenous in their clinical presentation. As such, we sought to identify caregivers of children with range of pediatric conditions to better understand potential similarities and variations in resource use and needs. Second, the PMCA identified cardiac, neurologic,

and mental health conditions as the top three body systems flags. Specifically related to neurologic and mental health conditions, we aimed to recruit caregivers of children with chronic conditions presenting with one of these clinical presentations. This would allow for a deeper exploration of resource use and needs amongst children who may have disorders impacting their cognition, communication, and/or behaviour. Third, given we were unable to determine geographical demographics (urban/rural) for the NB cohort, we aimed to recruit one NB family living in a rural community and one NB family living in an urban/suburban area. It is important to note that while we aimed to recruit these specific characteristics, we did recognize that this may not always be possible given the overall small population of children with medical complexity in the Maritimes.

5.7. RECRUITMENT

We employed multiple recruitment strategies to reach potential participants. Taking Phase 1 findings into consideration, we placed recruitment posters in hospital-based clinical areas relevant to children and youth with medical complexity in NS and PEI. Further, based on experiences from a previous NS-based study exploring respite care for families of children with medical complexity (Breneol, King, et al., 2022), we posted our recruitment flyer on social media platforms. Lastly, we emailed study information to key stakeholders in the care of children with medical complexity to share amongst their networks. Potential interested participants contacted the PI via email to receive more information. The PI answered any questions potential participants may have had and completed the screening/eligibility check list (See Appendix F). Once eligibility was confirmed, a copy of the consent form was provided to participants for their review. A

date and time for the interview was then set at a time best suited to potential participants. The recruitment process of care team members are described in the subsequent chapter.

5.8. INTERVIEW GUIDE

We developed a semi-structured interview guide using the 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018). We then included prompts based on the presence and absence of significant findings in Phase 1. For example, we included questions and prompts related to hospital-based services, number of care team members, and resource availability. It is, however, also important to consider patterns of health resource use that could not be explored using the health administrative data in Phase 1. The lack of community-based care resources and home care services was a significant limitation in mapping resource use within this population. This further supported the use of the 10 Domains of Health for Children with Medical Complexity as a data collection framework. This allowed for the exploration of the varied resources needed to support the health of the child and their family (i.e. accessible housing, respite care, private services). The semi-structured interview guide was reviewed and tested by a parent research partner who has a child with medical complexity to improve the relevancy of questions, identify any unclear terminology, and suggest areas for improvement in both content and process. Minor revisions were made, and a final semi-structured interview guide was created.

Chapter 6: QUALITATIVE PHASE TWO

The work presented in Chapter 6 is presented in manuscript format and prepared for submission to the journal *Child and Family Social Work*. Authorship is as follows: Breneol, S., Curran, J.A, Johnson, C., Macdonald, M., Martin-Misener, R., Vine, J., Montelpare, W., Stewart, S. Title: The Health Resource Use and Needs of Children with Medical Complexity and their Families: A Multiple Case Study Design

Statement of manuscript contribution: S.B conceptualized this work in collaboration with JAC, JV, WM, SS, RMM, and MM. SB collected and analyzed all data and drafted the manuscript. CJ doubled coded all transcripts. All authors contributed to revising the manuscript.

6.1. INTRODUCTION

Children with medical complexity are a growing population in hospital and community settings (Berry, Hall, et al., 2013; Foster et al., 2019). In response, there has been increasing efforts from researchers, policy makers, and clinicians to improve our understanding of this pediatric population and identify responsive strategies to attend to their care needs (Canadian Association of Pediatric Health Centres, 2016, 2018; Cohen et al., 2018). While diagnoses can be wide ranging, children and youth with medical complexity are a unique cohort united by four main characteristics: (1) presumed complex chronic condition(s); (2) high health resource use; (3) high family-identified needs; and (4) functional limitations (Cohen et al., 2011). Although these children may be relatively small in numbers, they require a vast amount of resources to attend to their complex care needs (Berry, Hall, et al., 2013; Charlton et al., 2017; Cohen, Berry, et al., 2012).

Much of the care for children with medical complexity is provided in their home by their families (i.e. biological/adoptive parents, guardians) (Foster et al., 2019; Gay, 2020). These children often have multifaceted care plans and/or medical equipment that require intensive supervision and management from their caregivers (Barone et al., 2020; Barton et al., 2021; Gay, 2020). Previous research using health administrative and national-survey data has sought to map the health resource use of this growing population (Berry, Hall, et al., 2013; Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012; Kuo et al., 2011). However, resources under investigation primarily focus on hospital-based health services such as inpatient admissions, emergency department use, and ambulatory clinic visits (Berry, Hall, et al., 2013; Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). While this work provides crucial information into the health system use of this population, these data sources often lack the ability to capture the range of resources needed to support and optimize the health of children with medical complexity and their families living in the community (e.g., equipment funding, home care services, and respite care). Further, previous research has shown that children with medical complexity and their families report having multiple unmet care needs, financial challenges, and increased stress levels (Charlton et al., 2017; Kuo et al., 2011; Kuo et al., 2014). These findings call for greater exploration into the range and extent of resources families access and need to optimally care for their children at home.

Barnert at al. (2018) developed a conceptual framework challenging the prevailing understanding of health for children with medical complexity and their families. Titled the 10 Domains of Health for Children with Medical Complexity, this

framework integrated findings from an evidence synthesis, participant observations, and interviews with families, clinicians, researchers, and policy makers (Barnert et al., 2018) (Figure 6-1). Unfortunately, a number of the domains outlined in this conceptual framework continue to be neglected in current pediatric complex care research literature (Barnert et al., 2018). In alignment with the World Health Organization's definition for health, this framework serves as a reminder that "health is a state of complete physical, mental, and social well-being and not merely the absence of disease or infirmity" (World Health Organization, 2022, p. 1). Exploring the resources needed to support the health of children with medical complexity and their families is an important first step in developing family-oriented policy and practice recommendations to improve care. Barnert et al.'s (2018) conceptual framework serves as an important structure to examine existing and needed resources across a range of health domains, moving beyond the predominant focus on biomedical services.

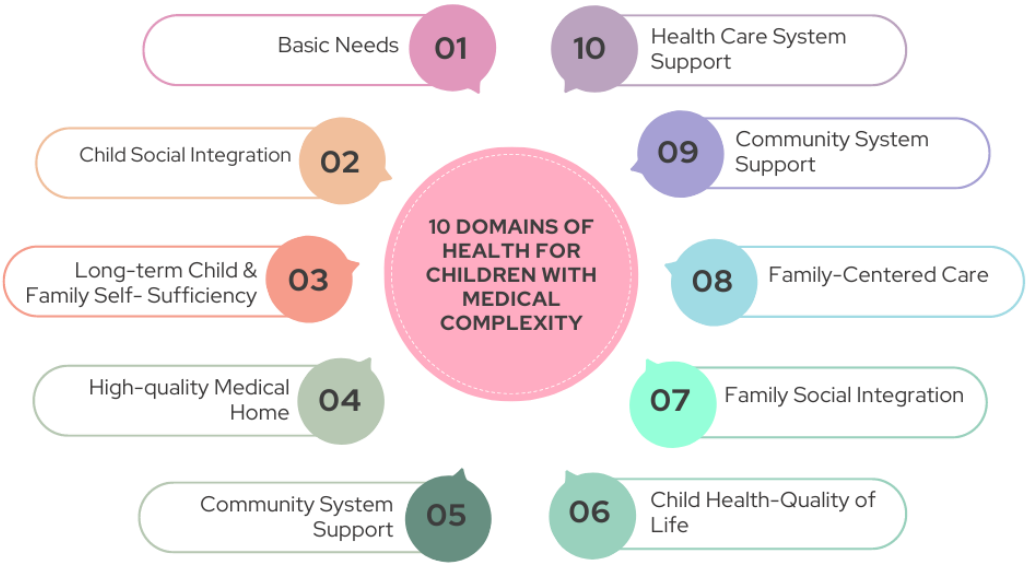

Figure 6-1. The 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018)

6.2. RESEARCH QUESTION

This research aimed to address the following research question: What are the family-reported experiences, health resource use, and needs of children with medical complexity and their families?

6.3. METHODS

This work is part of a larger mixed methods study being conducted to explore and map the prevalence, health resource use and needs of children with medical complexity and their families in the Canadian Maritimes (Breneol, Curran, et al., 2022). Full study methods can be found in our protocol (Breneol, Curran, et al., 2022). In the present study, a multiple case study design was used to examine the health resource use and needs of children with medical complexity and their families. Case study designs promote in-depth examination of complex phenomena in a 'real world' setting, considering surrounding contextual and interpersonal factors (Yin, 2017). Further, multiple case studies help identify common patterns, characteristics, and themes (Yin, 2017). This design was chosen to elicit information regarding: 1) 'what' resources families use to support their health; 2) 'how' they use these resources; 2) 'what' resource gaps are being experienced; and 3) 'why' these patterns of resource use may be evolving. Each case was informed by multiple perspectives, gathered from family caregiver(s), care team members, via semi-structured interviews and family resource journaling. This work was approved by the IWK Health and Health PEI Research Ethics Boards (#1026835 and #1024934).

6.3.1. Setting

The Canadian Maritimes is composed of three provinces, Nova Scotia (NS), New Brunswick (NB), and Prince Edward Island (PEI) and operates within a universal health care model (The Commonwealth Fund, 2022) . The primary site of this research was the pediatric tertiary care facility, located in NS, which provides specialty care for children and youth across all three provinces. This setting was chosen given that children's hospitals are often the main care site for children and youth with complex chronic conditions (Berry, Hall, et al., 2013; Bogetz et al., 2015). There are approximately 350,000 children and youth across the Maritimes (Government of Canada, 2018).

6.3.2. Population and Sampling

The focus of case study designs is not on the number of participants, rather it is on the gathering of multiple data sources to obtain a robust description of the case under examination (Yin, 2017). Children with medical complexity were conceptualized using Cohen et al.'s definitional framework for children with medical complexity and from findings from the first phase of this research (see protocol for more information) (Breneol, Curran, et al., 2022). We purposively sought to recruit children and families to build cases fitting a range of characteristics, such as rural and urban residence, level of complexity, and physical and neurodevelopmental limitations. We aimed to develop two cases from each Maritime province to examine possible variation in family experiences for those living inside and outside the provincial boundaries of the pediatric tertiary care facility. Families were eligible to participate if their child had a diagnosed or suspected complex chronic condition, if they were primary residents in one of the three provinces,

under the care of the pediatric tertiary care facility, and if they spoke English or French (See Appendix F for eligibility screening criteria). One primary caregiver self-identified as the contact for the study and as the individual seeking to take part in the interview. Participating caregivers were also asked to identify two members of the care team that could be sent an invitation via email for a short interview.

6.3.3. Recruitment and Data Collection

Multiple recruitment strategies were used to reach potential participants including distributing posters to pediatric hospital units, social media platforms, and through key stakeholders involved in pediatric complex care. S.B. responded to all participant queries, completed screening for eligibility, and provided detailed study information to eligible participants prior to facilitating written consent. Care team members that replied to the email invitation to participate in a short interview also underwent the informed consent process.

The caregiver interview guide was developed by S.B and J.A.C. and guided by the 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018). This semi-structured guide also included prompts that were informed by significant findings from the first phase of this work to create a better understanding into health resource use. This interview guide was pilot tested with a parent-research partner and minor changes were made to increase clarity and relevancy of included questions (See Appendix G).

All interviews took place over Zoom Video Conferencing or the phone and lasted 45-60 minutes. Demographic information was collected prior to the beginning of the interview and included the number of individuals in their family unit, child's condition,

child's age, community type, use of medical equipment, child's gender, caregiver's gender, caregiver's race/ethnicity, access to transportation, and caregiver's employment status. To expand perspectives, caregivers were provided with a journal template and asked to track their use of health resources over a 3 week-period (See Appendix H). Participants received a $50 gift card as an appreciation for their time.

Interviews with care team members took place over Zoom Video Conferencing and lasted between 15-30 minutes. The interview guide was composed of 2 open-ended questions and prompts were used based on the respective family interview (See Appendix I). A $10 coffee card was provided for their time.

6.3.4. Data Analysis

All interviews and self-reported journals were transcribed verbatim and uploaded to NVivo 12 Qualitative Data Analysis Software. In alignment with case study methods, a theory informed data analysis approach was undertaken using conceptual and theoretical frameworks (Yin, 2014). All data sources were read multiple times to become familiar with the data. First, each case was developed separately to create a representation of the unique family experience. Next all resources reported as being used by families were deductively coded using the 10 Domains of Health as the coding framework (Barnert et al., 2018). Data contained within the self-reported journal were also summarized using descriptive statistics. Interview and journal data were triangulated by organizing a matrix table based on the 10 Domains of Health.

Next, all interviews were coded using a deductive analysis approach based on the Capability, Opportunity, Motivation – Behaviour Model (COM-B) (Michie et al., 2014). The COM-B is a comprehensive synthesis of behaviour change theories and provides a

strong theoretical foundation to explore the plethora of factors that can influence health behaviours. This framework was chosen to help illuminate the 'what', 'how', and 'why' questions (Michie et al., 2011). Following this deductive analysis into the COM-B framework, a subsequent inductive coding analysis approach was undertaken to group similar themes within each case. After the within case analysis was complete, each case was treated as a unit of analysis to complete a cross-case comparison to explore common or dissimilar themes. A matrix table was created to visually depict emerging commonality or uniqueness across cases. Member-checking with participants was also completed to provide families' the opportunity to confirm, omit, or revise the synthesized data. S.B performed all data analysis and C.J double coded transcripts. S.B met frequently with the senior investigator to discuss emerging study findings.

Various strategies were undertaken to protect the confidentiality of participants. This included: removing all family names; omitting references to specific home provinces; replacing names of providers/programs/organizations with generic terms; reporting child diagnoses as an overarching condition group; reporting some demographic data in aggregate form; and assigning a randomly generated participant identification number for participant quotes.

6.3.5. Researcher's Position & Self-Reflection

The first author is 12 years in remission from pediatric cancer. Given these life experiences and her position as a Registered Nurse, she acknowledges the potential for bias, as well as unique insights, in this work. She has maintained a reflexive journal throughout this research as a strategy to separate her reflections with those shared in this work.

6.4. RESULTS

While we aimed to develop six case studies, the small population of children with medical complexity (<1% of the Maritime pediatric population) and the impacts of the COVID-19 pandemic presented challenges to recruit potential participants. In total, five case studies were developed, reflecting experiences in the 3 Maritime provinces. Ten emails were sent to care team members across the 5 cases and 5 consented to participating. The following section provides a high-level summary of each case and their use of health resources (Table 6-1 and 6-2). All caregiver participants self-identified as white and women. One caregiver was fully employed, one was on maternity leave, two had casual work, and one did not work outside the home.

6.4.1. Case Summaries

6.4.1.1. Case A.

This case depicts the family experiences of a 9-year-old girl living in a rural community with her parents and siblings. At just over a year old, her family began noticing that she was not developing "quite the same" as her peers. Her condition has eluded doctors, but they suspect it is a rare genetic condition and will continue to learn more with time. She has undergone day surgeries but has not needed a hospital admission. Her condition presents mobility limitations that are supported by using a wheelchair, cruiser, and walker. According to her mother, they have a very supportive care team at their community hospital, rehabilitation centre, and pediatric tertiary care facility. Her mother reported being the primary person organizing her care and is supported by one of the clinic receptionists to help coordinate appointments.

Over the course of three-weeks of health resource tracking, this family recorded: coordinating health appointments through phone and email (3.5 hours); having two appointments to support her medical needs with speech and physiotherapy (1.5 hours); participating in online educational classes (45 minutes); as well as spending approximately an hour locating and organizing a new respite worker.

6.4.1.2. Case B.

This case illustrates the family experiences of a 12-year-old boy living in a suburban community with his mother and sibling. At age 5, a formal diagnosis of a neurodevelopmental disorder was made at the pediatric tertiary care facility. His disorder presents a range of sensory and behavioural needs, lower cognitive level, and a very limited diet. Due to his care needs, he requires regular supervision. Various teams have been involved in his care including specialists at the tertiary pediatric care facility (e.g., psychiatry, dentistry, ear nose and throat), school-based therapy (e.g., occupational therapy, psychology), early intervention therapy, and community-based supports (e.g., nurse practitioner, psychiatry). His mother reported being the primary person responsible for managing his care but indicated a need for more support in this area.

Journaling reported two professional development days at school, leaving his mother without childcare options and having to adjust her working hours. She also received various text messages from his teacher about his behaviour (30 mins) and had a 1-hour Individual Program Planning meeting with the school. They also recorded having one dentist appointment (every 3 months) and nurse practitioner appointment (every 6 months).

6.4.1.3. Case C.

This case reflects the health resource use and needs of a 3-year-old boy living with his parents and younger sibling. At 3 months old, he was first admitted to hospital for approximately 10 days to run a series of diagnostic tests. At 6 months old, he was diagnosed with a rare form of muscular dystrophy. His mother describes them as "frequent flyers" to the hospital, being admitted approximately 10 more times since their initial discharge. His condition impacts his muscular system, resulting in muscle weakness and an inability to walk and feed orally. He uses a standing frame and a small wheelchair to help with ambulation. He has a gastrostomy tube (G-tube) for his nutrition needs, as well as a cough assist and suction machine to help his airways remain clear. His mother reports a positive relationship with their care team at their community hospital and hands-on physiotherapy and occupational therapy. They are also cared for by specialists at the Maritime's pediatric tertiary care facility (e.g, genetics, feeding team). His mother reports being a "very large part of his care" and feels supported by their local pediatrician and social worker who primarily manage and organize his care.

Over the three-weeks, the child had two sessions with physiotherapy, occupational, and speech therapy, an eye specialist appointment (time not reported), and spent two and a half days admitted to hospital. They spent 45 minutes, 6 times a day, to complete tube feeding and 5 minutes, 3 times a day, to use the cough assist. They also reported coordinating future doctors' appointments.

6.4.1.4. Case D.

This case represents a 13-year-old girl living in a suburban community with her mother and siblings. Shortly after birth, she was transported by helicopter to the pediatric

tertiary care facility where she spent the next 10 months in hospital. She was diagnosed with a rare skeletal genetic disorder characterized by hydrocephalus, syndactyly in the hands and feet, constricted airways, and developmental delays. She has a ventriculoperitoneal (VP) shunt and has recently had her tracheostomy of 12 years reversed, marking her 44th surgery. She now uses a continuous positive airway pressure (CPAP) machine at home. Though initially hesitant to engage in respite care discussions brought forth by her care team, an inpatient nurse introduced the mother to a local private respite family who specialized in caring for children with complex needs. This respite family has since become an incredible resource to this family, helping to support their housing (e.g., renting one of their rental properties), medical travel, and caregiving needs (e.g., respite breaks). Her care is primarily managed by specialists at the pediatric tertiary care facility (e.g., plastics, neurology), with a local pediatrician supporting their basic prescription needs and a larger out-of-province pediatric facility providing specialized surgical needs.

The health resource journaling outlined: an appointment for CPAP settings (1.5 hours); two unexpected audiology visits at their local hospital (3 hours); and a Zoom call to discuss an upcoming surgery (30 mins). There was also communication with care team members to organize three appointments at the pediatric care facility, arrange vaccines at school, and review CPAP needs.

6.4.1.5. Case E.

This case represents the family experiences with a 4-year-old child living with cerebral palsy. Born at 24 weeks gestation, she spent over 5 months in the neonatal intensive care unit where she experienced various medical complications. Following this

admission, she has not required any subsequent hospitalizations. Her mother attested this to being very careful to reduce potential exposures to viruses in the community. She uses several assistive devices, including a hypervibe, stander and walker. In total, they reported approximately 15 different specialists and care teams involved in their child's care including members from the Maritime's tertiary care facility (e.g., rehabilitation therapy, vision clinic), out-of-province health facilities, specialized educational authority, and community care (e.g., pediatrician).

This family reported: having 7 appointments for educational needs (8 hours total); attending 4 appointments to support her medical needs with an additional 2 hours of daily at home therapy (47.5 hours total); spending 30 mins daily on care coordination activities (10.5 hours total); traveling to pick up medical equipment (1 hour); and devoting approximately 3 hours/week organizing fundraising efforts to access a specialized care out-of-province (9 hours total).

Table 6-1. Summary of Case Characteristics

Characteristics	Case A	Case B	Case C	Case D	Case E
Diagnosis and/or Illness presentation	-Rare Genetic Condition -Global Developmental Delay -Speech Delays -Epilepsy	-Two Neuro Developmental Disorders -Sensory Needs -Behavioural Needs -Limited Diet -Lower Cognitive Function	-Muscular Dystrophy -Functional Limitations -Gastrostomy Tube -Unable to Walk or Feed Orally	-Rare Skeletal Genetic Disorder -Hydrocephalus -Syndactyly in the Hand and Feet -Tracheostomy for ~12 years (Recently Reversed) - Developmental Delays -VP Shunt	-Cerebral Palsy -Cortical Visual Impairment -Global Developmental Delay -Functional Limitations -Supination of Feet
Child/Youth Age	-9 years old	-12 years old	-3 years old	-13 years	-4 years old
Child/Youth Gender	-Girl	-Boy	-Boy	-Girl	-Girl
Caregiver Interviewed	-Mother	-Mother	-Mother	-Mother	-Mother
Number of Caregivers in the Household	-2	-1	-2	-1	-2
Community Composition	-Rural	-Suburban	-Rural	-Suburban	-Suburban
Number of Care Team Members/Teams	-9	-12	-11	-17	-17
Number of Care Team Providers Interviewed	-0	-1	-1	-2	-1

Table 6-2: Mapping of Resources Supporting Health of Child and Family Based on the 10 Domains of Health for Children with Medical Complexity (Caregiver Interviews + Care Team Members + Journaling)

Domain	Case A	Case B	Case C	Case D	Case E
Basic Needs	-Disability Tax Credit -Provincial Funding Supports	-Disability Tax Credit - Registered Disability Savings Plan	-Family Support -Disability Tax Credit -Provincial Funding Support	-Disability Tax Credit -Provincial Funding Supports -Affordable Housing -Social Assistance -Gas Cards	-Provincial Funding Support
Inclusive Education	-Online Home-Schooling Community -Library	-Individualized Learning Plan -Text Messaging with Learning Centre Teacher -Teaching Assistant -Accessible School Bus -School Meetings to Facilitate Grade Transitions	-Special Needs Assistant -Day Care	-Modified School Desk -Modified School Chair -Wheelchair for Use in Emergencies -Accessible School Bus -Educational Assistant(s) -School Installed Elevator to Improve Accessibility	-Specialized Educational Worker -Pre-primary Care Team Meetings -Developmental Interventionist -Occupational Therapy for Seating -Online Child Educational Class
Child Social Integration	-Grocery Store -Library Activities -Summer Camps -Facebook Community and Support Groups	-None noted	-Day Care	-Parent to Meet all Educational Assistants	-Planning for pre-primary
Current Child Health-Related Quality of Life	-Family & Friends	-Summer Respite Program (local non-profit organization)	-Day Care -Family & Friends	-School -Accessible Playground in	-None Coded

Domain	Case A	Case B	Case C	Case D	Case E
		-Arts Program (Community-based organization) -Local Informal Supports from Community Members		Backyard Built by Non-Profit	
Family Social Integration	-Creating Advertisements for Respite Worker -Flexible Working Hours	-Online Family-Respite Provider Matching Service -Co-parenting support	-Option for Hospital-Based Respite -Family Support	-Caregiver Therapy and Counselling -Private Respite Family -School-Based Support Program for Siblings	-Recreational Therapy (Hippocamp and Life Jackets for Beach)
Community System Supports	-Support Group for Children with Complex Needs in the Maritimes -Community-based Equipment Loan Program	-Community-based organizations -Local Informal Respite from Community Neighbours -Provincial	-Provincial Funding Support (Medical Equipment) -Speech therapy, occupational therapy, and physiotherapy (delivered in day care depending on pandemic restrictions)	-Provincial Funding Support / Social Worker (Medical Travel Expenses) -Private Respite Family -Online Family Support Groups	-Fundraising for private services
Long-Term Child and Self Sufficiency	-Care Team at Rehabilitation Facility -Other Parents	-Personal Network of Health Providers -Personal Educational Background -Child Psychiatrist -Transition to Adulthood Navigator	-Pediatrician -Genetics	-Social Worker -Private Respite Family with Pediatric Expertise -Online Family Support Groups	-Personal Educational Background -Friends -Social Media and Support Groups for Children with Complex Needs

Domain	Case A	Case B	Case C	Case D	Case E
		in Upper Grade School -Registered Disability Savings Plan			
Health Care System Supports	-Speech Therapy -Rehabilitation Facility -Community Hospital -Social Worker -Feeding Department -Physiotherapy -Occupational Therapy -Stander Trial at Community Recreational Centre -Provincial Funding Supports (reimbursement for medical appointments, equipment costs) -Occupational Therapy -Non-Profit Support (Equipment costs) -Private Physiotherapy (with private health insurance coverage) -Pediatric Tertiary Care Facility	-Dentistry -Nurse Practitioner -Speech Pathology (School-Based) -Pediatrician -Psychologist (School-Based) -Tertiary Care Facility -Ear Nose and Throat -Occupational Therapy -Early Intervention -Physiotherapy -Pediatric Psychiatrist -Social Worker -Mental Health Clinician	-Cough Assist -Standing Frame -Suction Machine -Physiotherapy -Occupation Therapy -Speech Therapy -Tube Feedings -Community Hospital -Genealogy -Rehabilitation therapy -Feeding Team -Respirology -Pediatrician (Community Hospital) -G-Tube Nurse -Social Worker (Community Hospital) -Private Insurance -Eye Specialists -Maritime Pediatric Tertiary Care Facility	-Sleep Therapy Company -Community Hospital -Large Pediatric Tertiary Care Centre (Out of Maritime Provinces) -Zoom Call with Specialized Team (Out of Maritime Provinces) -Vaccines Delivered at School -Pediatrician -Maritime Pediatric Tertiary Care Facility -Plastic Surgeon -Ear Nose and Throat -Rehabilitation Facility -Neurosurgery -Optometry -General Surgery -Genetics -Dietetics	-Specialized Physical Therapy -Muscle Stim and Hypervibe -Stander and Braces -Surgery Consultation (Out of Maritimes) -Local Tertiary Care Facility -Neonatal Intensive Care Unit -Rehabilitation Services -Equipment Loan Program -Speech Therapy -Eye Clinic -Feeding Clinic -Provincial Funding Supports -Developmental Interventionist -Pediatrician -Home-Based Therapy (completed by caregiver)

Domain	Case A	Case B	Case C	Case D	Case E
	-Rehabilitation Team			-Occupational Therapy -Speech Therapy -Physiotherapy -Modified Tricycle -Home Respiratory Care Assessment -Audiology	-Visual Impairment Specialist -Social Development -Psychology -Podiatry and Orthoptics -Augmentative Therapy -Social Worker
High-Quality Patient-Centered Medical Home	-Parent led care coordination activities (emails, phone calls, etc.) -Social worker -Community Navigation Program (No Longer Operational)	-Speech Pathologist (Informal Social Networks) -Parent-Led Care Coordination Activities	-Parent led care coordination activities -Social Worker (Community Hospital)	-Parent-led care coordination activities (i.e. emails, phone calls, etc.) -Private Respite Family	- Specialized Educational Authority Supports -Parent led care coordination activities
Family-Centered Care	-Strong Relationship with Care Team	-Personal Health-Related Education Background	-Strong Pediatrician-Provider relationship	-Collaborative and Child-First Care Team	-Positive Collaboration with Care Teams

6.4.2. Cross-Case Analysis

The following section describes the theoretically driven cross-case analysis organized by the relevant COM-B components and emerging themes (See Table 6-3).

6.4.2.1. Physical Opportunity

6.4.2.1.1. Use of Financial Resources.

All cases used financial resources to help support costs associated with raising a child with medical complexity. The federal Disability Tax Credit was a commonly used resource. Provincial government funding grants were used to supplement the out-of-pocket costs associated with basic needs (e.g., diapers), medical travel (e.g., taxi, mileage, meal, and accommodation coverage), equipment (e.g., g-tube pump) and respite wages. Only one out of the five cases reported these funding programs to be easily accessible, stating "as soon as we knew he wasn't going to walk, we were put in touch with them for his feeding supplies." [P8] Another case was not eligible due to income eligibility.

While families were grateful for financial support for medical travel, reimbursement rates have not been updated in several years, requiring families to cover the remaining balance. Other participants described various barriers to accessing provincial funding programs. One family shared that after several unreturned phone calls, they learned that there was no one currently in the position to process applications. They had been waiting for nearly a year for reimbursement for respite wages. In another case, accessing the provincial funding program was "a big struggle" and "took over a year" to be approved. They were told their initial application was lost and they described "endless

amounts of phone calls where [they] were hung up". They also learned that many families are denied from this program and often "don't push it any further because the mental load of even doing it the first time, can nearly break you in half." [P7]

6.4.2.1.2. Use of Care Coordination, Navigation and Communication Resources.

In most cases, mothers primarily organized and coordinated their child's care. Participants often reported being well supported with care team members such as pediatricians, nurse practitioners, social workers, and educational support workers to help coordinate and navigate resources. One case also described a collaborative care approach to their yearly appointments, where the family stays in one room with specialists rotating throughout the day. While some care team members may currently go above their dedicated role to support care coordination, "it feels like at this point it's luck of the draw, if they get somebody like that. As opposed to a real purposeful assigned role." [P2]

Most families reported having trusted and collaborative relationships with their care team members. However, one family felt frequently dismissed and unsupported when bringing forth questions or ideas for therapy to one of their care teams. While the family was being told by providers the importance of early intervention, there were often major delays in accessing the necessary therapy and equipment and were only being seen a couple times a year.

Communication amongst care team members can also be challenging, especially in cases receiving care in multiple provinces. The small community of specialists in the Maritimes makes it easier to contact the appropriate person with questions; however, it is often left to the caregivers to provide "the most vital or up-to-date information". [P12]

6.4.2.1.3. Use of Child Educational/School Resources.

Services such as physiotherapy, occupational therapy, speech therapy and psychology were reported to be delivered in the school/day care setting, increasing accessibility by reducing the amount of travel and time necessary for caregivers. In one case, a physiotherapist would come into the school to teach educational assistants (EAs) how to complete therapy. However, frequent turnover in EAs lead to a lack of hands-on therapy.

Children used a range of resources to participate in school/day care, including EAs, accessible buses, grade-transition planning meetings, and learning centres. While families did not have a choice of EAs, one mother highlighted that all caregivers have a right to know who is helping their child, particularly with personal care. One family had several meetings with the special education authority to prepare for their child for school. Another family chose to home school their children after the school closures began due to the COVID-19 pandemic. This turned into a positive experience for all that has continued regardless of pandemic restrictions.

While most families reported a supportive school environment, this was not the case for all. In one case, the child was suspended from school and "was kicked off the bus because of his behaviour". [P1] While he has been doing well on the new bus, he gets picked up later and dismissed earlier, creating shorter working hours for his mother. Being suspended was an intense experience, and they feel as though the school does not understand his needs. The care team member in this case highlighted that this action was inappropriate "since he's not in control of what he's doing, it's not intentional". [P3]

They also highlighted that it can be difficult to get the appropriate resources in place for a child with behavioural challenges.

6.4.2.1.4. Use of Hospital & Facility-Based Resources.

Participants in all cases reported having various care teams at the pediatric care centre. Some families were also supported by a specialized rehabilitation centre or an out-of-Maritimes tertiary care centre. Further, families were supported by providers at their community hospitals. Based on a previous experience where a life-threatening diagnosis was missed at their local emergency department, one family shared that they now choose to go immediately to the pediatric care centre for any emergent needs.

6.4.2.1.5. Use of Private Services.

Participants in three cases reported using private services. While unavailable now, one family previously accessed physiotherapy through their private health insurance. They also tried to access a local occupational therapist, but cost was a barrier. Another case had a private arrangement with respite providers that have now become a core part of the child's family. One mother reported seeking private services in and outside the region for their child's physical, visual, and ambulation needs.

6.4.2.1.6. Use of Community-Based Services.

Community-based nurse practitioners, child psychiatrists, and pediatricians were reported to support care delivery and prescription needs. Care team members also highlighted that local pediatricians, or primary care providers with a willingness to learn, may be best suited to meet the needs of children with medical complexity in the community. In one case, a community recreational centre provided space free-of-charge for physiotherapists on a 3–4-month rotational basis.

6.4.2.1.7. Use of Child & Family Social Integration Resources.

While families reported wanting their child to participate in summer and day camps, a lack of respite/support workers to accompany their children created a barrier to participation. Some families borrowed adaptive equipment from provincial programs and the tertiary care center to support social outdoor activities, such as going to the beach. Before the COVID-19 pandemic, one case attended the library multiple times a week to support their child's social nature and the grocery store was also seen as a positive social opportunity.

6.4.2.2. Psychological Capability

6.4.2.2.1. Parental & Institutional Knowledge Sharing.

Families turned to key people in their care team, such as social workers, pediatricians and specialists to gain the knowledge to care for their child's needs. They detailed their persistence in asking questions and seeking out additional information sources to increase their knowledge. There was, however, a reported disconnect between the resources that are being actively shared with families, and what is available for families to access. As one caregiver stated: "I feel like they are just told to say 'what would you like' and not say like 'here is everything we can offer you". [P4] This approach was challenging given that families often did not know what support they could be asking for, leaving some caregivers under-utilizing available resources.

6.4.2.2.2. Family Mental Health & Well-Being.

In some cases, participants highlighted the importance of supporting the health of the whole family, because "mental health is, like if parents are not healthy, then [the child] is not going to be healthy". [P7] One mother now attends therapy and support

groups for caregivers to care for her identity beyond being a mom to a child with complex needs. Her other children also receive therapy through their school to support having “as much of a normal childhood as they can.”[P9]

“Like caregiving, it is 24/7 with her, and you can love it but still be tired from it and that's one thing that I've learned over the years, like it is OK to say that, it is OK to say that it is exhausting to continually care for someone." [P9]

6.4.2.3. Social Opportunity

6.4.2.3.1. Social Networks.

All cases described their social networks as an important informal resource. Many turned to close friends and family to help support their respite, social, and informational needs. One case, however, emphasized that not everyone may have this type of support. Social media and online peer support groups were identified as a key resource to support knowledge sharing amongst families.

6.4.2.4. Reflective Motivation.

6.4.2.4.1. Need for Improved Financial Resources.

Cases identified a need for improved funding specific to pediatric care to support respite, medical travel, specialized equipment and tube feedings. Application processes should also be revised to reduce to administrative burdens and barriers to accessing funding support. Programs that provide funds to hire respite workers are often less than minimum wage and not well-funded for nursing level care. Participants indicated that there is sometimes a “lack of awareness” of the travel that’s involved for families and updating medical travel reimbursement, including mileage, food, and accommodation

stipends, to reflect the current cost-of-living is needed. One family highlighted that while they are now okay financially with funding support, they emphasis that there “are so many families that aren't. And are probably choosing between equipment and food right now.” [P7]

6.4.2.4.2. Need for Improved Care Coordination, Communication & Navigation.

All cases indicated a need for improved care coordination, communication, and navigation. Some families indicated that having a dedicated person to help coordinate and navigate their child’s care would be beneficial. Families also indicated a need for more accessible online information about available resources. One family suggested a ‘handbook’ written by families could be a way to reduce the hours spent researching information.

One care team member reported that “if there's a way of sort of checking somewhere to see what's been done in an up-to-date fashion, that would be helpful. If there are any significant changes in [their] care […], if there's a way to be updated if that happens". [P12]

6.4.2.4.3. Need for Improved Access to Respite Care.

Some mothers reported finding a respite worker as a priority need. Having a support person during appointments was also indicated, as they often must split their attention between their child’s needs and the care provider. Pediatric advocates are currently lobbying to formally provide respite in hospital as a “bandaid” solution to address the gap in respite care provision, with the goal to improve community respite in the long-term.

“It’s finding that person that can take her out of the house, to go do things without me, I think that is so important for my daughter and I’s relationship, for relationships with everybody else, like everybody else, and just having some time for myself, or yeah, that's definitely the biggest.” [P4]

If families do have the resources to pay respite wages, they still need to locate a worker with the appropriate qualifications. While certain families have had some success in locating respite workers over the years, it can be challenging to find someone that is qualified, affordable, and available. Some families also noted that it would be great to see day camps with workers that were equipped to care for children with complex needs, as they require one-to-one support and “you want to know that someone’s equipped to handle it completely”. [P7]

6.4.2.4.4. Need for More Accessible Pediatric-Therapy.

Participants in three out of the five cases reported the need for additional and more accessible pediatric-specific therapy. Specifically, the need for more pediatric physical, occupational, and speech therapy was noted.

"I believe my daughter would definitely qualify for more 1-1 physio support, weekly. Like multiple times a week with a physio. I do my best, I listen to everything, and I do a really good job, but I am not a physiotherapist, and even the EAs, they are not physiotherapists. So in an ideal situation, they would be seeing someone at least twice a week." [P4]

Participants in another case highlighted the difficulty in maintaining pediatric coverage in their suburban/rural community. While their community recently hired a new

pediatrician, they still have gaps in pediatric developmental psychiatry and mental help supports. Barriers to accessing pediatric services that were available in some home communities included long wait times, large workloads, travel distances, and cost.

6.4.2.4.5. Need for Inclusive Education for All.

Two families identified inclusive education as one of their top priorities. One family shared that they wished they had someone from the health sector to advocate for them in school. Other children in the school could also play more of a role in supporting children with disabilities to foster social integration and teach kids how to interact with peers with differing abilities (e.g. a peer buddy system).

6.4.2.4.6. Increased Understanding of Children with Medical Complexity & their Families.

Mothers reported the need for increased awareness and a true understanding of children with complex needs and their families. As one mother describes: "Yeah, people with disabilities, their lives matter. They have, you know, that sounds so cliche but like we're not just here to tolerate them and... I always say this, like, I know he's my child and I'm his mother, of course I think this about him, but people that have the opportunity to be around him, it's a privilege, like he can teach you so much […]. It's just, it shouldn't be just getting through and tolerating him". [P1]

Another participant shared that she wishes people would listen to her whole story and understand why she may "come off as difficult." As she states, "there's been too many errors, or like missing pieces, or people just not hearing, yeah day one, […], people just don't hear you, so you advocate your heart out constantly". [P7] Lastly, one family

reported being hesitant to engage in community activities given the easing of COVID-19 related restrictions with seemingly little consideration for children with disabilities (e.g., grocery stores restricting their ‘safe shopping hours’ to the older adult population).

Table 6-3. Cross-Case Analysis of Emerging Themes Based on the COM-B Model

COM-B Category	Theme Name	Cross-Case Comparison	Exemplar Quotes
Physical Opportunity	Use of Financial Resources	A; B; C; D; E	*"Like all of her equipment is so expensive, like when we didn't have, when we were purchasing all of her equipment out of pocket, it was so stressful. Because it's literally like we're buying this piece of equipment or like or something else that you can't do." [P7]* *"I actually heard through my physiotherapist, that she wasn't there anymore and I'd been leaving her messages, like, daily pretty much, and no one was calling me back. Because no one worked there. Even though her voicemail was still there, so I think it was … I think it's been a year, maybe."[P4]*
	Use of Care Coordination, Communication and/or Navigation Resources	A; B; C; D; E	*"So when we go for like a yearly clinic thing, we stay in one room and everyone comes to see us, probably like eight or nine different people throughout the day, and at the end of that day, they all get together without us and talk about everything and then come up with a plan, and bring the plan back to me." [P4]* "*I'm a very large part his care team, and his pediatrician knows that if she has any questions to ask me and I'll know exactly what's happening." [P8]*
	Use of Child Education Resources	A; B; C; D; E	"*Our school, we've been really fortunate, but I don't know if that's the norm for every family. Like, I just, that as a parent you should be like, I understand you can't choose your EA, 'cause that's understandable, like it goes by seniority sometimes, it goes by different things. But I definitely, think that you have a right to know who's around your child especially with help toileting and stuff."[P7]*

COM-B Category	Theme Name	Cross-Case Comparison	Exemplar Quotes
	Use of Hospital and Facility-Based Resources	A; B; C; D; E	*“I think that what's working well is that we can get the best of care in other provinces because we don't have it in our own.”[P11]*
	Use of Community-Based Services	A; B; C; D	“*So I think it's important that children with medical complexity do have, like probably a pediatrician that's best served, best suited, to meet their needs in their home communities, in order to coordinate care. But also family physician, nurse practitioner, with willingness to learn in a smaller community may serve the same role, but I think a community pediatrician is best suited to serve their needs and be able to coordinate all the various aspects of care that children with complex medical needs might require. Especially as they are not all the same.*” [*P6*]
	Use of Private Services	A; D; E	*“It was so much, over $500 for the first appointment, and that was like a half an hour or something like that, it was like OK well..yeah I can’t do that.” [P4]*
	Use of Child and Family Social Integration Resources	A; B; C; D	"S*he loves going to the grocery store and we haven't been since the pandemic. I mean, she hasn't been inside, but like they all know her, and my other kids too, and it's a great experience." [P4]*
Psychological Capability	Parental and Institutional Knowledge Sharing	A; B; C; D; E	*“unless you are offered it you don't necessarily know what to even ask for, is the issue right”[P9]*

COM-B Category	Theme Name	Cross-Case Comparison	Exemplar Quotes
	Family Mental Health and Well-being	D; E	"*So time is long, so like the entire day is very monotonous. It's, it's the same day, nearly every day. Just a different appointment, and then therapy, and then feeding. And we try to make it fun, obviously, but it's exhausting, as a mom and a dad it's exhausting, like yeah and it's exhausting in a way that your brain just feels mushy, because you're not really doing it, You're not really challenging yourself, but it's challenging in the way that it's, it's like Groundhog days. It's like the movie Groundhog Day, like, […] you don't want to say no to appointments because that's the best thing" [P7]*
Social Opportunity	Social Networks	A; B; C; D; E	*"I think I have a lot.. even like other parents that I know that have older kids or adult children that's been like a big source of support and thinking what it might look like in the future." [P4]*
Reflective Motivation	Need for Improved Financial Resource Support	A; C; D; E	**"***Like these the pieces of equipment that we buy, or if you're lucky enough to have insurance, aren't like one time lifetime things, so often a child needs more than one, and they're like $7-8000, and you need like a bunch of them. And a lot of things weren't covered, […]. like there's so many little things that just aren't covered and I think that we need to expand what's acceptable for therapy." [P7]*
	Need for Improved Care Coordination, Communication and Navigation	A; B; C; D; E	"Y*eah like it would be great if there was one person that like kind of like cued us along, like you need to start thinking about this, or you know, there's this program or whatever." [P1]*

COM-B Category	Theme Name	Cross-Case Comparison	Exemplar Quotes
	Need for More Accessible Pediatric-Specific Therapy	A; B; E	"*I should say my biggest issue is with the amount and quality of care that she's received for her development in her motor. Like involving her motor development, like there's just an absolutely nothing I feel like, I've had so many like many mini breakdowns where it's just like, I literally am her physiotherapist, her doctor, her OT, her occupational therapist." [P7]* "I *think it's important for both my daughter and the person to build the relationships and they can understand and be long term and follow through with stuff, also like as a kid to warm up to somebody and like getting them into a routine." [P4]*
	Need for Inclusive Educational for All	B; E	"*I have this like underlining fear, like that and it was kind of validated, that he's not going to be able to go to school. Like…. Even though I know intellectually, well he has a right and in a legal obligation to attend school, but I think when he was suspended and then when he was kicked off the bus, it was it was like I was validated. Like 'See!', like this this could really happen, and then what am I going to do? It's like it was almost like a self-fulfilling prophecy for me when he was suspended, like it's almost it's…. I don't know, like school and health care 'ya, we will be there and will help, you unless it's really bad', but I'm like, when it's really bad, that's when you need to be there the most." [P1]*
	Need for Improved Respite Care and Support Workers	A; B; C; D; E	*"And families have to find and hire their own respite workers, which often ends up being a very underpaid physician, especially for child with complex medical needs." [P6].*
	Increased Awareness and Understanding of Children with	B; D; E	"*I think for any family with social needs, that we just want our kids to have the chance to be happy. And like for [child], that, as much as her medical care is important, it's she's also a little girl. Like she is also a kid. [...]. Like [child] is also a kid that loves her dollies, and likes to play and demands*

COM-B Category	Theme Name	Cross-Case Comparison	Exemplar Quotes
	Medical Complexity and their Families		*grapes whenever she feels like it, and probably should get them, you know." [P9]*

6.5. DISCUSSION

This work describes the landscape of resource use and needs of children with medical complexity and their families. The use of the 10 Domains of Health for Children with Medical Complexity provided a conceptual framework to guide this research and map the range of resources used by families to support their overall health in the community (Barnert et al., 2018). Use of this framework expanded our concept of health beyond the traditional biomedical model to uncover the interwoven resources supporting health across the spectrum. Further, the use of the COM-B model provided an evidence-based theoretical coding framework to uncover what resources families are using, what/who enables resource use, and what gaps exist (Michie et al., 2014). While each case represented a unique family experience with ranging illness presentations and needs, similarities and differences emerged to illuminate potential directions to improve pediatric complex care. Findings from this work may also help to educate health professionals caring for this population on the extensive number of resources families use and need to support their overall health.

Participating mothers reported primarily managing their child's care, with most identifying a need for improved care coordination and navigation of resources. Current national Canadian guidelines on the management of children with medical complexity recommend assigning a 'key worker' to support families to navigate and obtain necessary services (Canadian Association of Pediatric Health Centres, 2018). The recommendations emphasize the importance of service integration across the health, social, community, and school sectors (Canadian Association of Pediatric Health Centres, 2018). In addition, key workers serve as a point of contact for families and members of the care team to ask

questions and foster streamlined communication across settings (Canadian Association of Pediatric Health Centres, 2018). Care coordination roles for this population have been frequently identified in the literature as a promising strategy to support overall care delivery and improve transitions in care (Cady et al., 2020; Cohen, Lacombe-Duncan, et al., 2012; Hillis et al., 2016). We suggest that a key worker role should be explored to attend to the care coordination and navigation needs of children with medical complexity and their families. An implementation and evaluation plan should be co-developed with families to ensure this role is effective in its intentions and avoids adding another area for potential communication gaps (Breneol, King, et al., 2022).

In addition to improving care coordination and navigation supports, findings from our case studies reveal the need for more transparent and accessible information about available resources. Some caregivers reported further challenges when attempting to access supports, including a lack of transparency in application processes and long wait times. Previous regional work exploring respite care for this population also revealed experiences where families received conflicting information about resource eligibility (Breneol, King, et al., 2022). This suggests the need for increased transparency of information from within organization walls and more effective dissemination strategies about available resources, their structure, and application/eligibility processes. While care coordination roles may help to narrow this gap in knowledge exchange, having an online repository of resources for families could empower them to access and advocate for the resources best suited to their needs (Charlton et al., 2017; Glassgow et al., 2018). This could also enable health professionals to improve their understanding of locally available resources. A recently co-developed program by a major Canadian pediatric centre is a

promising model that may attend to these knowledge, coordination, and navigation needs (Krantz et al., 2021). This program consists of a system navigator, supporting health, financial, school, and psychosocial needs (similar responsibilities of a 'key worker'); a parent navigator, attending to educational and social family needs; and a knowledge navigator, assessing community needs, gaps, and available resources (Krantz et al., 2021). The knowledge navigator is also responsible for updating a web-based resource repository (Krantz et al., 2021). In the first 5 years, this program reported positive improvements in connecting families with needed resources. They attributed their success to the carefully developed co-design partnership with families (Krantz et al., 2021). This program provides an innovative practice example directly attending to the needs of this population.

Existing evidence has revealed that approximately half of families of children with medical complexity report experiencing financial challenges and reductions in caregiver employment (Kuo et al., 2011). Canada experienced the largest increase in consumer inflation since 1983, with a marked increase of 8.1% between June 2021-May 2022 (Statistics Canada, 2022). However, it appears that some provincial funding programs have not updated their financial assistance in many years (Department of Community Services, 2012; Government of New Brunswick, 2014). For example, one program's medical travel policy for families outlines meal allowances at a maximum rate of $21 per day per person and $0.24 per kilometer for private vehicles (Finance and Treasury Board, n.d.). However, if an employee of the government is required to travel for work, they receive a total daily meal allowance of between $51.20-$62.55 and $0.40-0.50 per kilometer depending on travel distances (Social Development, 2022). Further,

while government programs like the Registered Disability Savings Plan can help families save for the future, this requires them to have a disposable income to contribute (Connected Care, 2017). Our findings are in alignment with other family voices in the country, suggesting that while current government programs help alleviate some financial strain, they are not yet optimal (Connected Care, 2017). A review of provincial funding programs is recommended to expand eligible expenses, update reimbursement rates, and reduce the administrative burden and barriers to apply. These findings should also signal health professionals to engage in discussions with families about potential financial challenges and advocate for improved policies to support their financial needs (Mooney-Doyle & Lindley, 2019).

Our findings suggest the need for improved pediatric-specific capacity across a range of disciplines, including rehabilitation therapy (physiotherapy, occupational, and speech therapy), mental health services, and respite care. These findings, however, are not unique to our study's context. Previous studies have also highlighted the need for improved accessibility of pediatric services for children with varied complex needs (Abulebda et al., 2021; Foster et al., 2019; Foy & Perrin, 2010; G. Graaf & Snowden, 2021). However, numerous factors can impact service availability and use (Comer & Myers, 2016; Foy & Perrin, 2010; Kuo et al., 2014). For example, while lengthy wait times for rehabilitation services is cited as a national concern, there has been a paucity of work exploring how best to reorganize care (Camden et al., 2010; Wittmeier et al., 2016). Further, there have been discussions around how community pediatricians could help to improve access to pediatric mental health services. However, redefining these roles to include the potential treatment and management of complex mental health and behavioral

needs requires further research and provider-specific education (Paton et al., 2021; Raval & Doupnik, 2017). Adding this responsibility may be further challenging given the difficulties in maintaining pediatrician coverage in some suburban and rural areas (Saltwire Network, 2017; Turner et al., 2020). Nurse practitioners (NPs) may also help to address these gaps by leveraging their unique expertise in holistic care (Samuels et al., 2017). Previous work has identified improved care coordination, health outcomes and family experiences when NPs are a key part of pediatric complex care teams in the community and hospital settings (Gresley-Jones et al., 2015; Mosquera et al., 2021; Samuels et al., 2017). Given that this pediatric population is reportedly increasing in numbers (Berry, Hall, et al., 2013), it is critical that a responsive and equitable system is developed. Future research in each of these service areas is needed to identify effective strategies to attend to the needs of all children and families.

While most caregivers reported having a collaborative care team and being well integrated in their communities and schools, this was not the case for all. A recent systematic review revealed that while there have been increasing efforts to improve social inclusion of children with disabilities in educational settings, they are continuing to face organizational barriers and feelings of exclusion (Woodgate et al., 2020). Children with medical complexity have also been referred to as the "silent victims of the COVID-19 pandemic" (p.171) and faced various restrictions to health resources (Mitchell, 2021). Importantly, this study draws attention to the everyday reality of families seeking resources to support the health of their child. Health professionals should be strongly advocating for the meaningful and full inclusion of children with medical complexity across settings, policies, and programs.

6.6. LIMITATIONS

There are limitations to consider with this study. First, this work was limited to the experiences of families in one Canadian region. Other contexts may differ in relation to resource availability and structures. It is also important to note the strength of a case study design is to provide an in-depth and detailed examination of the health resource use and needs of a child with medical complexity and their family; however, we acknowledge that these cases may not be representative of the entire pediatric complex care population, a noted limitation to case study designs (Yin et al. 2017). Second, interviews took place in English or French, potentially excluding an important population of non-English/French speaking families and their experiences. While we initially aimed to create a total of 6 case studies, with up to two interviews from care team members in each, the relatively small pool of potential participants and the COVID-19 pandemic created a challenge to recruitment efforts. While we had promising engagement on social media platforms and a strong connection with members of the Maritime's support group for families, the COVID-19 pandemic likely impacted family availability. Further, all caregiver participants in this study self-identified as white and women. However, previous work has shown ethnic, racial, and gendered disparities in pediatric care and caregiving (Courtney-Long et al., 2017; Kuo et al., 2011; Sharma et al., 2016). The intersections of identities likely impact health resource use and experiences more than our findings reveal. Greater work in this area is needed to design and build equitable and responsive systems.

6.7. CONCLUSIONS

Families used numerous resources across the health, social, community, and education sectors to attend to their care needs. These case studies highlighted various gaps to accessing health resources. Family-reported unmet care needs call for improvements to care for children with medical complexity and their families. Specifically, improvements to care coordination, financial funding supports, application processes, and knowledge sharing represent an important first step in supporting family needs. Further research is needed to identify and explore innovative care models to improve pediatric coverage across a range of disciplines and settings. Finally, health professionals should be advocating for improved health policy, programs, and education to support the health of children with medical complexity and their families.

Chapter 7: TRIANGULATION PHASE THREE

The work presented in Chapter 7 is presented in manuscript format and prepared for submission to the journal *Paediatrics & Child Health*. Authorship is as follows: Breneol, S., Curran, J.A., Martin-Misener, R., Macdonald, M., Vine, J., Montelpare, W., Stewart, S. Title: Children with Medical Complexity in the Canadian Maritimes: An Explanatory Sequential Mixed Methods Study

Statement of manuscript contribution: S.B conceptualized this work in collaboration with JAC, JV, WM, SS, RMM, and MM. SB analyzed the data and drafted the manuscript. All authors contributed to revising the manuscript.

Disclaimer: Portions of the data used in this report were made available by Health Data Nova Scotia of Dalhousie University. Although this research analysis is based on data obtained from the Nova Scotia Department of Health and Wellness, the observations and opinions expressed are those of the authors and do not represent those of either Health Data Nova Scotia or the Department of Health and Wellness.

7.1. INTRODUCTION

Pediatric complex care has been steadily gaining attention in recent years across research, practice, and policy sectors (Cohen et al., 2011, 2018). While there is variability in terminologies and conceptualizations used to describe this population, Cohen et al.'s definitional framework for children with medical complexity suggests that these children share four main characteristics: 1) the presence of a diagnosed or suspected complex chronic condition; 2) high health resource use; 3) functional limitations impacting daily activities; and 4) high family-identified needs (Cohen et al., 2011). In North America, children with medical complexity have been estimated to represent 10% of hospital

admissions, 20% of pediatric patients, 25% - 50% of inpatient stays, and 33% - 50% of health care costs (Berry, Hall, et al., 2013; Cohen, Berry, et al., 2012; Simon et al., 2010). Interpretation of these statistics should also consider that children with medical complexity have been estimated to represent just under 1% of the pediatric population (Aboneh & Chui, 2017; Cohen, Berry, et al., 2012; Kuo et al., 2011). Furthermore, most children are discharged from hospital to be cared for in their homes (Berry, Hall, et al., 2013; Foster et al., 2019). Families demonstrate unwavering resiliency when caring for children with medical complexity, but often contend with varied unmet care needs, such as unmet respite, rehabilitation and financial needs (Charlton et al., 2017; Connected Care, 2017; Kuo et al., 2011; Kuo et al., 2014; Thomson et al., 2016).

Developing a comprehensive understanding into the prevalence, health resource use, and needs of these children and their families can be challenging (Berry et al., 2015). To date, understanding of the population of children with medical complexity in Canada has predominately been informed by literature using national survey and health administrative data from the United States. However, there are important contextual differences between the United States and Canada, particularly related to health care systems. There have been only two reports in Canada detailing the prevalence and health resource use of children with medical complexity (Canadian Institute for Health Information, 2020; Cohen, Berry, et al., 2012). While these are seminal works using Canadian health administrative data, greater regional research is warranted to explore potential contextual variations in resource use, availability and needs. Furthermore, health administrative data lack the ability to capture the true extent of resources families use to support their health, particularly while living in the community (i.e. private

services, community programs, home nursing, equipment funding and respite care). Increasing our understanding of this population is a critical first step in developing responsive family-centered strategies to attend to the needs of children with medical complexity and their families in the Canadian Maritimes.

Previous work in this field has been primarily quantitative or qualitative in design and a-theoretical in nature (Barnert et al., 2018). Mixed methodology can be beneficial when qualitative and quantitative methods alone do not adequately address the research question (Creswell & Plano Clark, 2018). Combining both health administrative and family-reported data may be one strategy to attend to the respective strengths and limitations of singular research designs. Routinely collected health administrative data is a powerful tool to provide important population-based prevalence and health resource use estimates. Whereas qualitative work can help to further identify and map the range of resources used by families to support their health, while also providing an opportunity to explore contextualized experiences using these resources and their care needs.

This paper is focused on the third and final phase of a mixed methods study aimed to gain a greater understanding into the population of children with medical complexity and their families in the Canadian Maritimes (Breneol, Curran, et al., 2022). The work was driven by the overarching research question: How can health administrative and family-reported data be used to gain a comprehensive understanding into the patterns of health resource use and needs of children with medical complexity and their families in the Canadian Maritimes? As such, the objective of this final phase was to explore and describe the ways in which family-reported health resource use, experiences, and needs

converge or diverge with the health resource use identified within routinely collected health administrative data.

7.2. METHODS

We used an explanatory sequential mixed method design to address our research question (quan → QUAL) (Figure 1) (Creswell & Plano Clark, 2018). A pragmatic philosophical approach was used to guide the overall design of this project. This position assumes a 'what works' approach to answering an overarching research question (Creswell & Plano Clark, 2018; Feilzer, 2010).

We conceptualized the population of children with medical complexity based on Cohen et al.'s (2011) definitional framework. The 10 Domains of Health for Children with Medical complexity was also used to guide the conceptualization of health for this unique population (Figure 7-1) (Barnert et al., 2018). This framework was developed from a comprehensive synthesis of evidence from the literature, families, children/youth, clinicians and other key stakeholders (Barnert et al., 2018). More details about the study population and use of conceptual frameworks can be found in the full study protocol (Breneol, Curran, et al., 2022).

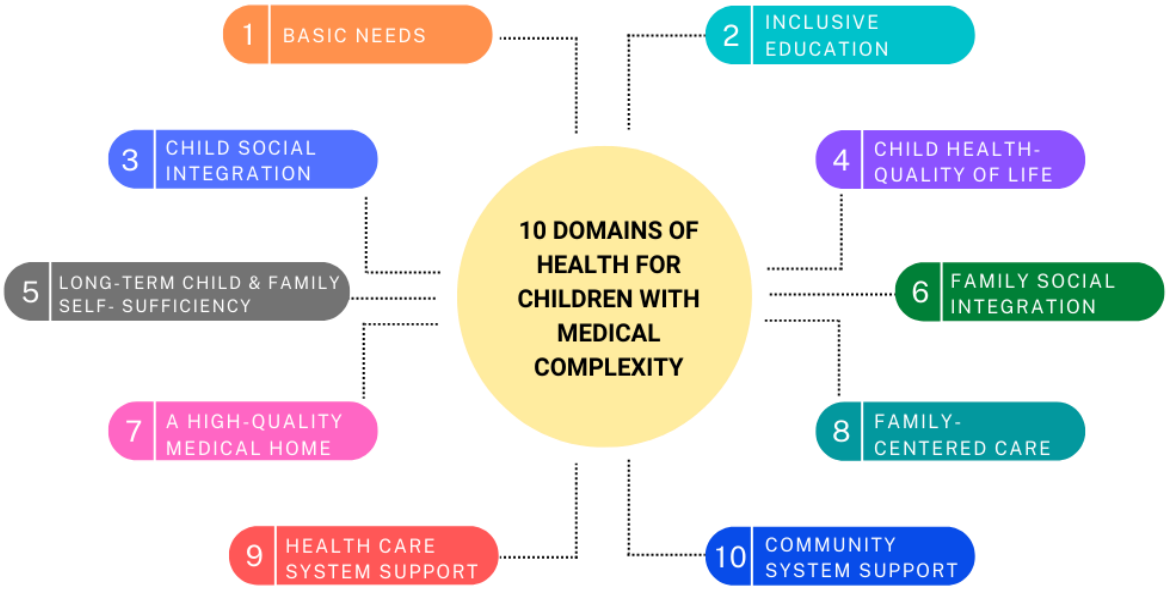

Figure 7-1. 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018)

7.2.1. Setting

This study took place in the Canadian Maritimes, an eastern Canadian region composed of three provinces (Nova Scotia [NS], Prince Edward Island [PEI], and New Brunswick [NB]). There is one pediatric tertiary care centre, located in Nova Scotia, providing specialty care to all children and youth in the Maritimes. The pediatric population across all three provinces is estimated to be around 350,000 (Government of Canada, 2018).

7.2.2. Quantitative and Qualitative Phases

This work began with a secondary analysis of routinely collected health administrative data to map the prevalence, clinical characteristics, and health resource use of Maritime children with medical complexity (Phase 1). Findings from this first quantitative phase also informed recruitment and semi-structured interviews during phase 2. A qualitative phase (Phase 2) was then subsequently conducted by building case

studies illustrating family health resource use, experiences, and needs. These case studies were informed by semi-structured interviews with family caregivers and members of their care team and a 3-week journaling of health resource use. This design was chosen to elicit a deeper understanding of the health resource use and needs of children with medical complexity and their families than either data source on its own. Full study methods and results for each phase can be found elsewhere (Breneol, Curran, et al., 2022). Ethics approval was obtained from the IWK Health and Health PEI Research Ethics Boards (#1026835 and #1024934).

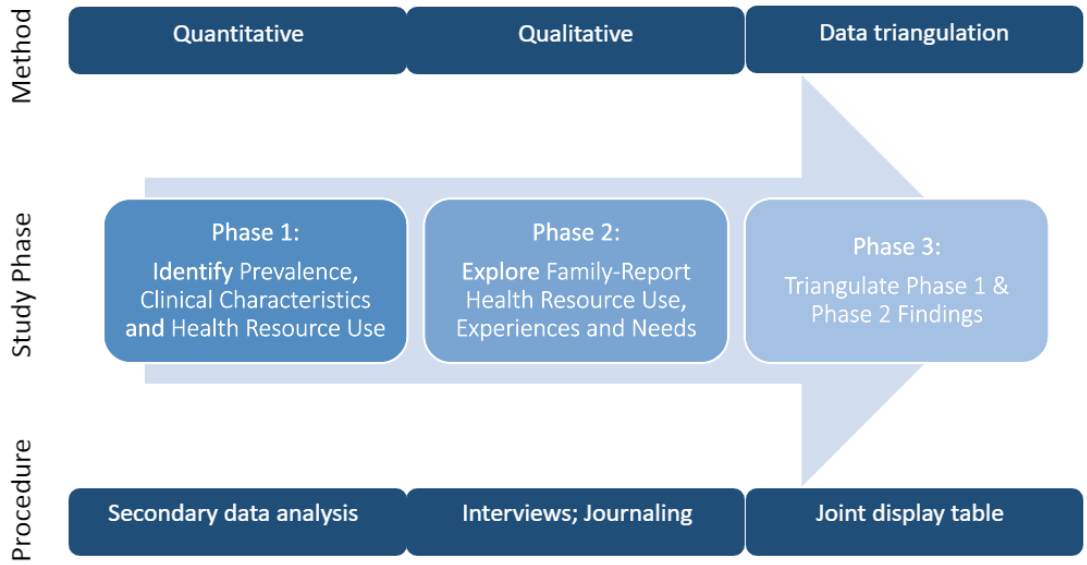

Figure 7-2. Summary of Study Methods

7.2.3. Data Triangulation

In alignment with an explanatory sequential mixed methods design, the third phase of this work sought to examine the extent to which the qualitative case study results converge with or diverge from the initial findings from the health administrative data. To do so, S.B and J.A.C met to review phase 1 and phase 2 findings. Next, we

sought to triangulate findings from phase 1 and 2 using a joint display table (Creswell & Plano Clark, 2018; Guetterman et al., 2015). Joint displays are a central tool in mixed methods research that serve to visually depict both data sources together to create new perceptions and insights (Creswell & Plano Clark, 2018; Guetterman et al., 2015). We used a statistics-by-theme framework to organize the joint display table to provide a visual representation and structure to map relevant findings from the health administrative data with the subsequent qualitative case studies (Creswell & Plano Clark, 2018; Guetterman et al., 2015). We started by sorting key findings from phase 1 into a structured table. We then subsequently mapped the family-reported resource use, experiences, and needs to related concepts identified in phase 2. This resulted in a joint-display table to better understand the similarities and differences between findings produced by health administrative and case study data. This table also highlighted where unique findings were produced from one data source alone.

7.2.4. Trustworthiness of Findings

Guidance on the conduct of mixed methods research stipulates the importance of attending to its overall validity, as well as the validity of each quantitative and qualitative phase on their own (Creswell & Plano Clark, 2018). Strategies outlined by Guba (1981) were employed during the qualitative phase, including maintaining a reflexive journal throughout, frequent debriefing with members of the research team, and keeping detailed methodological notes. Additional strategies to attend to the overall validity of the mixed methods design included considering all significant and non-significant quantitative data, linking phase 1 findings with sampling and interview procedures in phase 2, and carefully

designing each phase to provide a deeper exploration of the topic of interest (Creswell & Plano Clark, 2018).

7.3. RESULTS

The triangulation of the health administrative and case study findings is presented in Table 7-1. There was a total of 11 themes generated, with six stemming solely from phase 2 findings. The statistics-by-theme joint display table highlighted both the strengths and limitations of using health administrative and family-reported data as a single source, versus multiple sources that together, improved our understanding of the health resource use and needs of this population. Health administrative data revealed important population-level measures, indicating that children with medical complexity represent 0.88% of the Maritime pediatric population and are at greater odds of experiencing a hospital discharge [odds ratio: 668; CI: 391.51, 1130.03], emergency department visit [odds ratio: 2.97; CI: 2.64, 3.35]; and primary care visit [odds ratio: CI: 2.46, 3.39]. Case study findings confirmed and built upon phase 1 findings by providing a greater, more contextualized, understanding into the health resource use and needs of this population. Further, this work provided important contextual information for a more nuanced interpretation of health data findings (i.e. respite care delivered in-hospital). Case studies also revealed an unseen network of resources families use to support the health of their children. This demonstrated the interplay between community, hospital, education, social and private-based resources to support the overall health of both child and family. Families take on responsibilities in care coordination and communication and spend a significant amount of time providing care at home, researching potential supports, and applying for resources. Families have also faced several barriers to accessing resources,

including a lack of transparency in eligibility, lack of human resources, and lengthy application processes.

Table 7-1. Statistics-by-Theme Joint Display

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
Care Providers / Care Coordination, Communication and Navigation Resources	A median of 6 distinct provider specialists were in their care team (range 0-18).	Family case studies reported a range of 9-17 care team members/teams. In the absence of a dedicated care coordinator role for children with medical complexity, mothers primarily coordinated their child's care. Mothers were supported by members of their care team and often felt like a core member of the care team.	Children with medical complexity may have more care coordination and communication needs than health data alone may reveal. Qualitative work suggests that gaps in care coordination and communication often falls on families to fill.
Hospital-Based Resources	99% of our sample had a hospital admission after their first discharge with a complex chronic condition. There was a median of 2 discharges (range of 0-55 with an average length of stay of 4 days). Children from rural areas were less likely to have a hospital discharge (IRR=1.2, 95% CI: [1.1, 1.3]) than those in urban areas.	3 out of 5 children had an admission to hospital. Discharges varied from 0 to 44. One case living in a rural community indicated that their local hospital had an option to be admitted to their inpatient unit for respite breaks.	Children with medical complexity may not always be admitted to hospital. This has implications when seeking to identify children with medical complexity both at a population and clinical level. Hospital admissions can greatly vary amongst children with medical complexity depending on their conditions and needs. Health data provides a strong population level-overview.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
			Hospital discharges may also be reflective of admissions for respite care. This context is important when interpreting hospital discharge findings from health data.
	Children with medical complexity had a median of 2 emergency department visits. Children from rural areas were less likely to have an emergency department encounter (IRR=0.84, 95% CI: [0.78-0.90]) than those living in urban areas.	One case reported choosing to travel directly to the tertiary care center's emergency department (urban area) for emergent needs because of a previous encounter where a life-threatening diagnosis was missed at their local (suburban area) emergency department.	Pediatric complex care capacity varies in communities outside of urban areas and may impact emergency department use.
	Children with medical complexity had a median of 3 hospital-clinic visits. Rate of hospital clinic visits among children living in a rural area was 2.32 times higher than the rate of those living in an urban area [CI: 2.11-2.54].	Children with medical complexity were reported to see various providers and specialists at both community, rehabilitation and tertiary care centres. Some cases also receive care outside of their home province and/or country.	Children with medical complexity may have more encounters with hospital care than provincial health data may be suggesting. Families often require specialized care outside of their home communities, creating time, transportation, and cost implications.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
	The odds of children with medical complexity having a neonatal intensive care unit admission were 17.39 times higher than children without medical complexity. The odds of children with medical complexity having an intensive care unit admission were 28.94 times higher than children without medical complexity.	Two cases described lengthy neonatal intensive care admissions. Caregivers described several complications and stressful situations while admitted. No cases described having a pediatric intensive care admission.	Both data sources suggest that children with medical complexity have intensive care admissions. Qualitative data revealed that families may experience various stressful and potentially traumatic situations while admitted to the neonatal intensive care unit. This has important implications for the availability of mental-health resources for families.
Community-Based Resources	Children with medical complexity had a median of 34 primary care visits and 10 pediatrician visits. Children from rural areas were less likely to have primary care visit (IRR = 0.89) than those living in urban areas.	Families were often supported by pediatricians and primary care providers, who were also identified as being best suited to meet the needs of children with medical complexity in the community. One case identified challenges in maintaining pediatrics coverage in their suburban/rural community.	Pediatricians and primary care providers are well positioned to support the care of children with medical complexity living in the community. This care provision has potential implications for community providers (i.e. human resources, education on pediatric complex care).

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
	None Reported	Three cases identified the need for more pediatric therapy, particularly related to hands-on rehabilitation services (physical, occupational, and speech therapy) and mental health resources.	Children with medical complexity require care from a range of disciplines in the community that may go uncaptured by health administrative data. Greater pediatric-specific capacity is needed to attend to the needs of children with medical complexity, particularly related to rehabilitation and mental health therapy.
	3% of the sample had a physician or nurse practitioner home care visit.	Case studies did not report receiving home care services. There was one community-based organization that was reported to do in-home visits for respiratory equipment checks. One care team member highlighted that expanding respite care funding could improve in-home supports for families. Caregivers reported a substantial amount of supervision and therapy being delivered in the home. For example, one caregiver spent 45 minutes 6 times a day, to complete tube feeding, requiring waking every 3-4 hours overnight.	Current provincial health administrative data lacks comprehensive indicators for pediatric home care use. Families deliver a substantial amount of care in the home. The lack of home care supports being used by participating families may be related to a lack of available options.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
Prevalence and Characteristics	Regression modeling for hospital discharges, emergency department visits, primary care encounters, and hospital clinic visits showed a similar U-shaped IRR pattern in age, with children under 1 having the highest IRR, then decreasing for age groups 1-4, 5-9, 10-13, before increasing again in the 14-18 age group.	Case studies with school aged children reported using various resources to support their child attend school and transition between grades.	Both data sources indicate that age may play a factor in type and quantity of resource use.
	No major effect of sex in the models.	No indication of sex impacting health resource use.	Neither data source revealed indications of an association between sex and resource use.
	Children with medical complexity represented just under 1% of the pediatric Maritime population.	One case shared that they could not find how many children with medical complexity live in the Maritimes.	While these children may be small in numbers, they require extensive resources to support their health in the community. Caregivers may appreciate knowing how many other families with children with medical complexity live in their area.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
Awareness & Understanding	Children with medical complexity use a disproportionate amount of health resources in comparison to children without medical complexity. Children with medical complexity had a median of 0 ambulance transfers with a range of 0-30.	Cases identified the need for increased awareness and understanding about children with medical complexity and their families. Caregivers indicated they wished people would take the time to hear their stories and get to know their child, beyond their medical and/or behavioural needs. There is a lack of awareness about the amount of medical travel for children and their families. One case reported being transferred to the pediatric tertiary care centre via helicopter. Not all families have access to private transportation. Provincial funding programs can help the costs associated with medical travel. One case reported fundraising efforts for medical travel.	Greater awareness is needed about the lived experiences and realities of children with medical complexity and their families. Transportation to and from appointments is an important element to consider when caring for children with medical complexity and their families.
Private Services	None Reported	Some cases turned to private therapy for rehabilitation therapy, though cost and travel distances can become a barrier.	Families are turning to private service options to address gaps in available care in the public system. This can create additional time, cost and travel implications for families.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
Financial Resources	None Reported	All families used financial resources to support basic needs (i.e. diapers, disability tax credit), medical travel (i.e. mileage, taxis, meals), equipment (i.e. G-tube supplies, wheelchairs, walkers), and respite wages. Families reported barriers when applying for financial support, including a lack of transparency in program eligibility, lack of human resources, lengthy application processes, and income cut-offs. Cases suggest that financial support does not match current family needs.	Qualitative work highlighted several resources families use to support the costs of raising a child with medical complexity. Regretfully, families face various barriers when seeking to access financial support, including lengthy and stressful application processes. If families do get support, reimbursement rates often do not reflect current cost of living, requiring families to cover expenses out-of-pocket.
School-Based Resources	None Reported	Physiotherapy, occupational therapy, speech therapy, and psychology were delivered in school settings. All children had an Educational Assistant. One case highlighted the difficulty in training educational assistants to provide care/therapy due to high turnover. One case had a strong collaboration with a special education authority to create a care plan for school.	Examining health data alone would have missed therapy delivered in the school setting. This further demonstrates the extensive number of interwoven resources across varied settings that families must learn to navigate. Qualitative data also suggests that improvements should be made to improve inclusivity in schools,

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
		One case wished to have an advocate from the health sectors to support her child's inclusion in school. This case also suggested that getting resources in place for a child with behavioral needs can be challenging.	regardless of a child's condition or needs.
Child and Family Social Integration Resources	None Reported	Some cases reported borrowing adaptive equipment to engage in social activities (i.e. hippocamp at the beach). Community spaces, such as the library and grocery stores were seen as important social experiences. Lack of respite/support workers impacted children's abilities to participate in summer/day camps.	Caregivers value having the resources in place to support their children's social needs. Engaging in these settings often required support from health sector (i.e. borrowing recreational equipment, respite providers).
Respite Care	None Reported	Respite care can take on various roles for families, including providing breaks for caregivers, supporting children attend social activities, and helping caregivers during appointments. Caregivers face challenges in finding pediatric-specific respite workers that are	Respite care is an important health resource some families identify as being their top priority. Greater pediatric-specific respite options are needed to support caregiver health, child socialization and care-related appointments.

Theme	Health Administration Data	Family Case Studies	Mixed Methods Interpretation
		qualified, affordable, and a suitable match. One family have a private arrangement with a respite family for 12+ years.	
Social Networks	None Reported	Friends, family, and peers supported respite, social and information needs. Social media groups were a key resource for information sharing.	Families often created strong social networks in their physical and online communities to support their navigation and information needs.

7.4. DISCUSSION

This work sought to employ a mixed methods approach to gain a more comprehensive understanding into the prevalence, health resource use, and needs of children with medical complexity in the Canadian Maritimes. By examining the findings of both quantitative health administrative and qualitative case study data, this study provides important population-level information that was further contextualized and expanded upon through the integration of lived experiences. While families often confirmed what health administrative data suggested, our case studies revealed a complex network of unseen resources supporting the health of children with medical complexity and their families. Despite the vast number of resources families leveraged to support their life in the community, families of children with medical complexity still reported unmet care needs in areas such as care coordination, financial support, respite care, and certain pediatric therapies. We acknowledge that the number of themes included in this work may appear excessive. However, after consultation with study participants, we chose to distinctly identify each of the 11 themes to make every effort to capture the true complexity of resource use and needs as described by families. This study provides a novel contribution to the pediatric complex care literature by taking an explanatory sequential mixed methods approach to gain a greater understanding of the overall population of children with medical complexity and their families. By doing so, this study highlighted potential strategies to improve pediatric complex care in the Maritimes.

Living at home is considered the optimal place of care for most individuals with complex care needs (Foster et al., 2019; Morin et al., 2016). Our results align with those

from previous work describing the extensive amount of responsibilities families take on to fill gaps in health care structures and care for their children at home (Foster et al., 2022). Families not only have to learn how to navigate the siloed medical, social, community, and education systems but also provide daily at-home therapy, personal care, and care management activities. This is in addition to financial implications related to medical travel, equipment and basic supplies, to name a few. Caregivers of children with medical complexity have been referred to as the "shadow of the health care system" (Thomson et al., 2016, p. 187) and as "essential health care personnel" (Murphy & Darro, 2021, p. 1). Drawing focus to the lived reality of caregivers is crucial in caring for this population, as well as the development of responsive health policies and programs. Clinicians caring for children with medical complexity should ensure their practice is rooted in family-centered care and provide anticipatory guidance and screening about potential financial, time, and caregiving challenges (Mooney-Doyle & Lindley, 2019). This is of particular importance given that research suggests the health of the family is intertwined with that of the child (Barakat et al., 2015; Barnert et al., 2018). In addition, however, clinicians should be advocating for improvements in health policies, programs, and resources to meet their needs more effectively.

Children with medical complexity and their families access support from a range of intertwined resources across different government ministries, authorities, settings, and sectors. Using one Maritime province as an example, in-home respite care services are delivered under the Department of Seniors and Long-Term Care. While our work captured few home care encounters, case studies identified the importance of respite care for families to have a break from caregiving and support their child's social integration.

There are government funding programs to receive support for respite care wages through the Department of Community Services. However, finding qualified workers in the community is often a challenge, especially for those in need of nursing tasks (Breneol, King, et al., 2022; Evidence In-Sight, 2013). To address this gap in community respite care provision, some hospitals admit children to their inpatient unit for respite if staffing allows (Breneol, King, et al., 2022), creating potential cost and human resource implications to the Department of Health and Wellness. This example depicts the cascading impacts of resource availability across sectors.

While care coordination and navigation interventions may help facilitate access to care, our findings also suggest the need to expand the quantity and quality of resources available for pediatric complex care. Particularly, these results suggest the need to start expanding the concept of supportive health resources beyond that delivered by traditional health care structures. Data-driven research using routinely collected health administrative data may be contributing to the over medicalization and hospital-dominant narratives of pediatric complex care. However, triangulation of health data with family case studies illustrated an often-unexplored network of community-based resources supporting their health. This is in direct alignment with emerging work from Foster et al. (2022) suggesting that it will take collaborative and innovative partnerships with families, clinicians, policy makers, and researchers to attend to both the medical and social needs impacting the health of children with medical complexity (Foster et al., 2022). Potential strategies may include expanding funding opportunities to support equipment, specialty food, and caregiving costs, increasing access to mental health resources for the whole family, and increasing pediatric respite care capacity in the community. In other health

settings, innovative strategies such as ‘social prescribing’, appear to be gaining momentum (McMaster Optimal Aging Portal, 2022; Nowak & Mulligan, 2021). For example, providers in Canada can prescribe provincial park passes to increase exposure to nature given its beneficial outcomes on both mental and physical health (CBC News, 2022). Committed collaborations across the health, education, community, social, and private sectors is needed to achieve a responsive system to improve health outcomes and experiences for this important pediatric population. Findings from this work call for children with medical complexity and their families to be a priority population for health reform.

7.5. STRENGTHS AND LIMITATIONS

Respective limitations to each phase of this work can be found elsewhere (Breneol, Curran, et al., 2022). A systematic triangulation process was followed to ensure the appropriate integration and interpretation of the quantitative and qualitative data (Creswell & Plano Clark, 2018) and to produce robust results. We also want to acknowledge that the administrative data did not have the functionality to capture race and ethnicity data in Phase 1 findings. Further, caregivers in our phase 2 sample self-reported as white. The intersections of identities within this population and their experiences and use of health resources must be a focus of future work to build an equitable and responsive health system.

7.6. CONCLUSIONS

This work combined the use of health administrative and family reported data to gain a greater understanding into the prevalence, health resource use, and needs of children with medical complexity and their families. While children with medical

complexity represent less than 1% of the pediatric population in the Canadian Maritimes, they are at greater odds of using health care services in comparison to children without medical complexity. Triangulation of findings suggest that the quantity of health encounters may be even greater than health data alone may capture, such as physiotherapy in schools, out-of-province surgeries, and private respite services. Further, families access health resources across countless sectors and settings to attend to their care needs. It will take committed action and co-developed partnerships with families, clinicians, researchers, and policy makers from a range of sectors and settings to develop innovative resources and models of care to support the health of children with medical complexity and their families.

Chapter 8: CONCLUSION

To date, our understanding of children with medical complexity and their families has primarily been informed by research literature outside of Canada using single study designs. This study demonstrated the utility of taking a mixed methods research approach to explore the prevalence, health resource use, and needs of children with medical complexity and their families in the Canadian Maritimes. Each chapter and manuscript included in this dissertation built upon previous phases and informed subsequent work to achieve a comprehensive understanding into the population of children with medical complexity and their families. Chapter 2 provided an overview of the literature regarding children with medical complexity, their prevalence, use of health resources, and care needs. Manuscript 1 (Chapter 2) presented a scoping review detailing how health administrative data are informing practice, policy, and research recommendations for children with medical complexity. Manuscript 2 (Chapter 3) delivered a full outline of the protocol undertaken to complete this work. Manuscript 3 (Chapter 4) leveraged the use of linked health administrative datasets to reveal that children with medical complexity are estimated to represent less than 1% of the Maritime pediatric population. In addition, compared to children without medical complexity in Nova Scotia, they were at greater odds of having a hospital clinic, primary care, and emergency department encounter. Chapter 5 provided more detailed information on the individual health datasets and the Pediatric Medical Complexity Algorithm used during phase 1 (Simon et al., 2014). This chapter also highlighted how Phase 1 results were used to inform the interview guide and recruitment efforts during Phase 2. Manuscript 4 (Chapter 6) presented 5 case studies illustrating family-reported health resource use and needs using complementary

conceptual and theoretical frameworks. This work revealed a complicated network of resources across hospital, community, education, and private sectors and identified several potential directions to attend to the care needs of this population and their families. Manuscript 5 (Chapter 7) triangulated the health administrative and family case study findings to gain a more contextual and nuanced understanding into the population of children with medical complexity and their families living in the Maritimes. Triangulation of findings suggested the need for innovative collaborations with policy makers, families, clinicians, and researchers across sectors to attend to the health needs of children with medical complexity and their families. Chapter 8 concludes this work by summarizing the study's strengths and limitations as well as its implications for clinicians, policy makers, and researchers.

8.1. STRENGTHS AND LIMITATIONS

This study had several strengths, including the novel focus and integration of health administrative and family-reported data to gain a greater understanding of the population of children with medical complexity and their families. Further, previous literature has also highlighted the predominantly a-theoretical nature of pediatric complex care research (Barnert et al., 2018). As such, this study leveraged the use of three complementary definitional, conceptual and theoretical frameworks to help guide the development and analysis of this work. In consideration of these strengths, this study strongly suggests the need for children with medical complexity and their families to be a priority population for health reform and innovative cross-sector collaborations.

The scoping review exploring the use of health administrative data suggested a paucity of mixed methods approaches to examine this pediatric population and served as

an important foundation to guide this work. The quantitative phase of this dissertation was the first to use discharge data from the Maritime's only pediatric tertiary care centre to identify the prevalence of children with medical complexity. By further linking Nova Scotian residents to several provincially held health administrative datasets, this work provided overarching insights into the number of health care encounters by children with medical complexity and revealed greater odds of using health services in comparison to a matched control group. The qualitative phase further confirmed and expanded upon quantitative findings to reveal an interwoven network of resources across sectors and settings that families access to attend to their care needs living at home. The collection and triangulation of multiple sources of data, including health administrative, semi-structured interviews and self-reported journaling, created a robust evidence base which can inform potential strategic directions to improve pediatric complex care in the Maritimes.

Despite several strengths, findings from this work should also be considered alongside the following limitations (more detail on each limitation can be found in the respective chapter/manuscript). First, given the variability in terminologies and definitions used to describe children living with complex chronic conditions, our scoping and literature review may not have captured all potentially relevant articles. Second, health administrative data is not collected for research purposes. As such, researchers can only work with what is originally collected and stored. Further, there are limitations to relying solely on diagnostic codes to identify this multidimensional population. Third, due to resource limitations and constraints with cross provincial data linking, health resource use could only be extrapolated from Nova Scotia residents. This province was

chosen given it has the largest pediatric population of the three Maritime provinces and is home to the pediatric tertiary care centre. Fourth, although we sought to purposively sample certain family characteristics based on Phase 1 findings, we acknowledge that the opinions or experiences reported in our case studies may not be shared by other families. Other health centers and settings may differ in resource availability and care structure. Further, participating families included in the case studies all self-identified as white and mothers. Much research is still left to be completed to explore the intersectionality of identities within this population and must be a focus of future work.

8.2. IMPLICATIONS FOR PRACTICE, POLICY, AND EDUCATION

This study provides overarching insights into the population of children with medical complexity in the Canadian Maritimes that can help inform the development of responsive changes to health policy, education, and practice. While these children may be relatively small in numbers, their disproportionate use of health resources and reports of varied unmet care needs indicate the need to explore innovative strategies to improve policy and service delivery for pediatric complex care. As discussed throughout the manuscripts included in this dissertation, findings from this work have various implications for pediatric health practice, policy, and education.

While social determinants of health are often discussed in health education (e.g., nursing, medicine, etc), the extent of resources needed to support health across these determinants may be shadowed by the prioritization of addressing medical needs. Findings from this work should be embedded in health curriculum and could help educate and inform care providers (e.g., nurses, physicians, social workers) interacting with these children and their families about their experiences and needs living in the community.

This may prompt discussions with families beyond their medical needs to include considerations such as potential financial challenges and the importance of attending to caregiver mental health. This understanding and consideration may be a critical first step in improving care delivery and experiences for these children and families.

As discussed in Manuscript 3, routinely collected health administrative data highlighted important clinical characteristics and resource use that should be considered when caring for this population and designing health policies. Specifically, study findings illustrated the broad range of clinical characteristics within this pediatric population. For example, mental health conditions were noted amongst the top three body system flags as indicated by the PMCA. This is important information to include and discuss when educating potential providers about this pediatric population. These findings should be considered when designing programs and policies supporting children with medical complexity, paying careful attention to eligibility criteria to ensure appropriate reach and inclusivity.

These findings further demonstrated the need for more comprehensive pediatric home care data. While home is considered the optimal place of care for most individuals with complex needs, there remains a large gap in our understanding about pediatric home care provision (Foster et al., 2019). At the time of this study, there were no available health administrative datasets collecting information on pediatric home care nursing. Further, participating caregivers did not report receiving home care services. This, however, may be partially indicative of the availability of pediatric home care. While existing literature highlights the crucial role of home care nurses in supporting children with medical complexity and their families, there remains a shortage of nurses in these

positions (Foster et al., 2019). Previous regional work with this population has also indicated challenges in locating pediatric-trained home care nurses to provide respite care (Breneol, King, et al., 2022). Advocating for more comprehensive home care data is needed, particularly to our national data information systems. In response to this growing need, CIHI has named children and youth, as well as community/home care, as priority areas for data collection in their 2022-2027 strategic plan (Canadian Institutes of Health Information, 2022). Further, clinicians and policy makers should be advocating for the strengthening of pediatric-specific home care provision to attend to the needs of children with medical complexity and their families in the community.

Leveraging the use of mixed methods research, this work illustrated cascading impacts of resource availability on families and across health sectors and settings. Findings suggest that families often assume additional caregiving responsibilities to address the gaps in our health system. This can be seen in areas such as care coordination (e.g., mothers reported primarily managing their child's care), rehabilitation resources (e.g., paying providers in the private industry, daily at home therapies), and respite care (e.g., locating and hiring a private respite worker). Findings from each phase of this work provided valuable and important evidence to support the advocacy to expand the quantity and quality of resources available to families, beyond that delivered by traditional health care sectors. This will require collaborations across governmental and organizational sectors to review and update relevant health policies to fully reflect the needs of this population and their families.

8.3. RECOMMENDATIONS FOR FUTURE RESEARCH

Findings from this dissertation serve as an important foundation for future research initiatives focused on the population of children with medical complexity, some of which have already begun.

The scoping review identified three predominantly reported algorithms used to identify children with medical complexity in health administrative datasets. While prevalence estimates are relatively consistent across the literature, it is unclear how their constructed cohorts may differ in terms of clinical and demographic characteristics. To address this gap in our understanding, I am a co-applicant on a Canadian Institutes of Health Information-funded project seeking to validate and explore each of these algorithms using Maritime health administrative datasets (PI: Dr. Janet Curran). This work will also bring to light what content is being recorded in their medical records, creating a new understanding into the resources and needs being discussed/documented with families.

In alignment with existing literature, results from this work further suggest the need for improved pediatric-specific capacity across a range of disciplines in the community. Potential variations in resource use and availability amongst urban and rural communities were also noted. Future research is needed to explore new models of care to potentially add, train, expand, and redistribute roles to better support these children and their families living in the community. For example, previous work has identified improved care coordination, health outcomes, and family experiences when nurse practitioners are a part of their care team (Antolick et al., 2020; Cady et al., 2020; Gresley-Jones et al., 2015). Given their holistic approach and advanced skills, nurse

practitioners may be uniquely positioned to attend to the needs of children with medical complexity and their families (Cady et al., 2020).

This work provided a broad understanding into the overall population of children with medical complexity and revealed the intertwining relationships between the 10 Domains of Health for Children with Medical Complexity (Barnert et al., 2018). This study serves as an important starting point and foundation to conduct a deeper exploration into each of these health domains and continue to identify further and more tailored interventions to the needs of children with medical complexity and their families in the Maritimes. Specifically, future research questions should explore the theoretical connections, relationships and intersections between the each of health domains.

8.4. CONCLUDING STATEMENT

This mixed methods study advances our knowledge about the population of children with medical complexity and their families in the Maritimes. By combining the use of health administrative and family-reported data, this work provides a greater understanding into the prevalence, health resource use, and needs of this important pediatric population. Children with medical complexity were estimated to represent just under 1% of the pediatric population; however, they were at greater odds of experiencing outpatient, hospital, and emergency care encounters when compared to children without medical complexity. The use of the 10 Domains of Health for Children with Medical Complexity and the COM-B model provided a theoretical and conceptual foundation to build case studies illustrating the complicated networks of resources families use and need to support their health in the community. Triangulation of findings illuminated the extent of resources families use to support their health that is often gone unseen. The

family-reported unmet care needs in areas such as care coordination, respite care, rehabilitation services, and financial supports sends a strong signal to care providers and policy makers that improvements are needed to enhance pediatric complex care in the Maritimes.

This research fills a gap in the pediatric complex care literature by leveraging the use of both quantitative and qualitative data guided by conceptual and theoretical frameworks. To our knowledge, this is the third study to map the prevalence of children with medical complexity in Canada, and the first to combine both population and family-reported data to gain a more comprehensive picture of their health resource use and needs. Overall, this study provides evidence and theory informed recommendations to improve pediatric complex care education, practice and policy. This work acts as a foundation for future research focused on improving the health experiences, resource use, and outcomes for children with medical complexity and their families in the Maritimes.

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Appendix A: Electronic Database Search Strategy

Database	Search Concept Combinations		Limiters
	"children"	"medical complexity"	
PubMed	(child* OR pediatric* OR paediatric* OR preschool* OR toddler* OR kid OR kids OR adolescen* OR youth OR young* OR teen* OR preteen* OR juvenile* OR boyhood OR boy OR boys OR girl OR girls OR infant* OR infancy OR neonat* OR newborn* OR baby OR babies)	technology dependency OR technology dependent OR technology dependence OR technology assisted OR medically complex OR profound disability OR profound disabilities OR severe disability OR severe disabilities OR multiple disabilities OR severely disabled OR complex care OR medical complexity OR complex needs OR medical fragility OR complex chronic OR multiple chronic	Title/Abstract
CINAHL	(MH "Child+") OR (MH "Pediatrics+") OR (MH "Pediatric Nursing+") OR (MH "Hospitals, Pediatric") OR (MH "Child Health Services+") OR (child* OR pediatric* OR paediatric* OR preschool* OR toddler* OR kid OR kids OR adolescen* OR youth OR young* OR teen* OR preteen* OR juvenile* OR boyhood OR boy OR boys OR	technology dependency OR technology dependent OR technology dependence OR technology assisted OR medically complex OR profound disability OR profound disabilities OR severe disability OR severe disabilities OR multiple disabilities OR severely disabled OR complex care OR medical complexity OR complex needs OR	Title/Abstract

Database	Search Concept Combinations		Limiters
	“children”	“medical complexity”	
	girl OR girls OR infant* OR infancy OR neonat* OR newborn* OR baby OR babies)	medical fragility OR complex chronic OR multiple chronic	
EMBASE	OR 'child*' OR 'paediatric*' OR 'pediatric*' OR 'preschool*' OR 'toddler*' OR 'kid' OR 'kids' OR 'adolescen*' OR 'youth*' OR 'young*' OR 'teen*' OR 'preteen*' OR 'juvenile*' OR 'boyhood' OR 'boy' OR 'boys' OR 'girl' OR 'girls' OR 'infant*' OR 'infancy' OR 'neonat*' OR 'newborn*' OR 'baby' OR 'babies'	technology dependency OR technology dependent OR technology dependence OR technology assisted OR medically complex OR profound disability OR profound disabilities OR severe disability OR severe disabilities OR multiple disabilities OR severely disabled OR complex care OR medical complexity OR complex needs OR medical fragility OR complex chronic OR multiple chronic	Title/Abstract

Appendix B: Scoping Review Data Extraction Form

Concept	Extraction
Author	
Year	
Study Design	
Aim/Objectives	
Country	
Data Sources	
Conceptual/Theoretical Framework/Model	
Conceptual Definition of Medical Complexity	
Method for Identifying Cohort	
Result Summary	
Policy/Practice Recommendations	
Research Recommendations	
Strengths to Using Health Data	
Limitations to Using Health Data	

Appendix C: List of Included Variables

List of Variables by Project Objective

Source Dataset	Variable Name (Label)	Variable Description	Level of Identification	Time Span	Why is this element required in the analysis?	Why is this level of identification required?
Pediatric tertiary care centre data	Patient ID	Unique patient identifier	Individual Level	2004-2017	This will allow for linking between the Study ID and encrypted HCN	For cohort identification and data linking
Pediatric tertiary care centre data	HCN	Encrypted Health Care Number	Individual Level	2004-2017	Following encryption at Medavie, this data source will be sent to HDNS for data linking	For cohort identification and data linking
Pediatric tertiary care centre data	Case ID	Unique identified for each observation	Individual Level	2004-2017	This variable is a number assigned to each line of observation	For data management
Pediatric tertiary care centre data	Birth year	Birth date provided by month and year	Individual Level	2004-2017	To identify differences between health care utilization	To identify health care utilization

					within the identified cohort.	
Pediatric tertiary care centre data	Sex	Assigned sex at birth	Individual Level	2004-2017	To identify differences between health care utilization within the identified cohort	To identify health care utilization
Pediatric tertiary care centre data	Postal Code	Postal Code of patient residence	Individual Level	2004-2017	To identify province of residence and urban/rural residence. This will be derived variable of urban/rural residence and will be sent to HDNS at this level of identification.	To identify the Nova Scotia cohort and urban/rural residence for cohort characteristics
Pediatric tertiary care centre data	Discharge Date	The date the patient was discharged	Individual Level	2004-2017	To calculate age of first discharge and identify an index date to	To identify the start of examining health resource use

						follow health resource use from that date onwards	
	Pediatric tertiary care centre data	Up to 50 Diagnosis Codes	Patient's ICD Codes	Individual Level	2004-2017	To be used with the Pediatric Algorithm for Medical Complexity to identify the study cohort	To identify the study cohort
	MED	Msi (Patient MSI Health Card Number)	Encrypted MSI health care number	To be replaced by study ID	2004-2018	For use by HDNS to link across databases	To allow for across database linking
	MED	Birth date	Birth date provided as month and year	Individual level	2004-2018	To determine age of cohort and differences between health care utilization based on their age.	This will be used to calculate age and ensure it matches the data pulled from the IWK Meditech. Flag for <1year old will be required.

						To be used by HDNS to derive age.	
	MED	Sex	The individual's sex	Individual level	2004-2018	To be used by HDNS to determine the matched control cohort and to report characteristics of the matched cohort	To identify a matched cohort
	Pediatric tertiary care centre data	Major Clinical Diagnostic Category	A derived variable from the ICD-Codes and corresponding major clinical diagnostic category as outlined by the Pediatric Algorithm for Medical Complexity (Simon et al.,	Flag if cardiac, craniofacial, dermatologic, endocrinologic, gastrointestinal, genetic, genitourinary, hematologic, immunologic, mental health, metabolic, musculoskeletal, neurologic, ophthalmologic, otologic, pulmonary/respiratory,	2004-2017	To identify cohort characteristics	To identify cohort characteristics.

			2018) using the IWK Dataset	renal, progressive condition, continuous dependence on technology for >6 months, and malignancies progressive or metastatic that affect life function.			
	MED	Postcode (Postal code)	Postal code of the individual's most recent correspondence address given to the provider.	Flag if urban or rural residence	2004-2018	This will be a derived variable of urban/rural residence to determine the matched control cohort	To identify a matched cohort
	MED	Dxdate (Date of Service)	The date the health service was provided	Full date	2004-2018	To determine service date of health care encounter	To identify health care utilization
	MED	Hospunit (Hospital Treatment Unit)	Hospital unit where treatment was received	If hospital unit is emergency (EMCC), intensive care (INCU), neonatal care (NICU), INPT (Inpatient), OTPT (Outpatient) and all others (OTHER)	2004-2018	To stratify health care utilization by service type	To identify health care utilization

MED	Bspecial (Provider Specialty)	The specialty under which the provider billed for the service encounter. Not necessarily the provider's main specialty	All provider specialties	2004-2018	To stratify health care utilization by provider type	To identify health care utilization
MED	Dspecial	Licensed (main) specialty of the physician. May not be the specialty that was billed for in the service encounter	All provider specialties	2004-2018	To stratify health care utilization by provider type	To identify health care utilization
MED	Location (Treatment Location)	Type of treatment facility in which the service encounter occurred	Flag if visit occurred in hospital (HOSP), Physician Office (OFFC), Patient's Home (HOME), Home Hospital Care (HMHC), Nursing Home (NRHM), and all others (OTHER)	2004-2018	To stratify health care utilization by treatment location	To identify health care utilization
MED	Hosp number	Number that corresponds to specific hospitals	Hospital	2004-2018	To examine health care	To understand where care is

						utilization across the province.	primarily provided
	CIHI DAD	MSI		To be replaced by study ID	2004-2018	For use by HDNS to link across databases.	To identify health care utilization
	CIHI DAD	Discharge date	The date when the patient was formally discharged	The date in which they were discharge	2004-2018	To examine health utilization across the province	To identify health care utilization.
	CIHI DAD	Institution Number	Code assigned to a reporting facility identifying the facility and level of care of the data submitted	Hospital number	2004-2018	To examine the type of health care utilization across the province	To understand where care is primary provided and identify health care utilization. This will illuminate where this cohort (community hospital or large health centre) receive care.

CIHI DAD	Admit via Ambulance	Identified whether a patient arrives at the reporting facility via ambulance and the type of ambulance that was used	To flag yes/no	2004-2018	To determine health care utilization of ambulance use	To identify health care utilization
CIHI DAD	Admission Date	The date and time that the patient was officially registered as an inpatient	Full date	2004-2018	To determine service date of health care encounter	To identify health care utilization
CIHI DAD	Calculated Length of Stay	Derived variable - The difference, in days, between the Admission Date and Discharge Date	Number of days	2004-2018	To determine average length of stay for inpatient utilization	To identify health care utilization
CIHI Dad	Institution from	Identifies another health care facility from which the patient was transferred for further care	What hospital within Nova Scotia the cohort is being transferred from or from out of province hospital	2004-2018	To determine number of transfers between facility	To identify health care utilization. This will illuminate where this cohort (community hospital or large

						health centre) receive care.
CIHI DAD	Institution to	Identified the institution number of another health care facility, another level of care within the reporting facility	What hospital within Nova Scotia the cohort is going to or if going to a hospital out of province	2004-2018	To determine where children are receiving care and patterns in that care	To identify health care utilization. This will illuminate where this cohort (community hospital or large health centre) receive care.
CIHI DAD	From Institution level of care	Identify level of care	Flag if level of care is: Acute Care (1), Chronic Care (3), Nursing Home (4), Psychiatric Facility (5), Home Care (8), Day Surgery (A), Emergency Room (E), Organized Outpatient Department (O), all others as (X)	2004-2018	To determine where children are receiving care and patterns in that care	To identify health care utilization
CIHI DAD	To institution level of care	Identify level of care	Flag if level of care is: Acute Care (1), Chronic Care (3), Nursing Home (4), Psychiatric Facility	2004-2018	To determine where children are receiving	To identify health care utilization

				(5), Home Care (8), Day Surgery (A), Emergency Room (E), Organized Outpatient Department (O), all others as (X)		care and patterns in that care	
	NACRS	Health Care Number	Patient's unique health care coverage number.	To be replaced by study ID	2011-2018	For use by HDNS to link across databases.	To identify health care utilization
	NACRS	Institution Number	Code assigned to a reporting facility identifying the facility and level of care of the data submitted	Hospital number	2011-2018	To examine the type of health care utilization across the province	To understand where care is primary provided and identify health care utilization. This will illuminate where this cohort (community hospital or large health centre) receive care.
	NACRS	Date of Registration/ Visit	Date when the patient is officially registered for emergency or	Full date	2011-2018	To determine health care utilization by service type	To identify health care utilization

			ambulatory care services				
	NACRS	ED Visit Indicator	Indicate whether a visit reported under the emergency MiS functional centre account code is an arrange day surgery or clinic visit taking place in the ED or an ED visit	Flag if ED visit or not	2011-2018	To determine health care utilization by location and service type	To identify health care utilization
	NACRS	Provider Services 1-8	Identifies the service(s) of the health professional(s) responsible for provision of services to the patient during the visit.	The 5 digit code	2011-2018	To identify health care utilization	To stratify health care utilization by provider type
	NACRS	Institution To	Identifies the institution number of another health care facility or	What hospital within Nova Scotia the cohort is going to or if going to a hospital out of province	2011-2018	To determine where children are receiving	To identify health care utilization. This will illuminate where this cohort

			another level of care within the reporting facility where the patient was transferred to for further care.			care and patterns in that care	(community hospital or large health centre) receive care.
	NACRS	Institution From	Identifies another health care facility from which the patient was transferred for further care	What hospital within Nova Scotia the cohort is being transferred from or from out of province hospital	2011-2018	To determine number of transfers between facility	To identify health care utilization. This will illuminate where this cohort (community hospital or large health centre) receive care.

NACRS	Institution from type code	Identifies the type of facility in which the patient was transferred from for further care	Flag if level of care is: Acute Care (1), Chronic Care (3), Nursing Home (4), Psychiatric Facility (5), Home Care (8), Day Surgery (A), Emergency Room (E), Organized Outpatient Department (O), all others as (X)	2011-2018	To determine where children are being transferred from	To identify health care utilization
NACRS	Institution to type code	Identifies the type of facility in which the patient was transferred to for further care	Flag if level of care is: Acute Care (1), Chronic Care (3), Nursing Home (4), Psychiatric Facility (5), Home Care (8), Day Surgery (A), Emergency Room (E), Organized Outpatient Department (O), all others as (X)	2011-2018	To determine where children are being transferred	To identify health care utilization
VITAL	Health Care Number		To be replaced by study ID	2004-2018	For use by HDNS to link across databases.	To identify health care utilization

VITAL	Date of Death	Date in which the individual died	Date of death provided as month and year	Jan 2004-Dec 2018	To identify if a death occurred	To account for in health resource use analysis

Appendix D: National Ambulatory Care Reporting System (NACRS) in Nova Scotia

NACRS uses Management Information Systems (MIS) Functional Centres to distinguish types of ambulatory care. All institution numbers in NACRS for Nova Scotia start with a '2' for province and then '9' indicating ambulatory care followed by the three-digit facility number.

Emergency Department (ED)

- All Emergency Departments (ED) have a functional centre that begins with 71310^^
- Not all facilities collect ED information using NACRS and the data cannot be extrapolated from one facility to another.
- ED information is collected using one of two levels of NACRS – Level 1 or Level 3. The field Submission Level Code in the database tells you which level of NACRS is being used.
- Hospitals began collecting ED information at different times and some have started and stopped before completing a year.
- There is no clinical data (diagnosis or intervention) collected with Level 1
- The province has not mandated collection of ED information using NACRS at this time

Coverage for hospitals:

Aberdeen (29011) – Level 3

April 1, 2013 to April 30, 2013

January 1, 2014 to March 31, 2014

April 1, 2014 to March 31, 2015

Currently collecting for fiscal 2015/16

South Shore Regional (29014) - Level 3

October 1, 2003 to March 31, 2015

Currently collecting for fiscal 2015/16

Colchester Regional (29018) – Level 1

April 1, 2013 to June 30, 2013

Hants Community (29037) – Level 1

April 1, 2011 to March 31, 2015

Currently collecting for fiscal 2015/16

Queens General (29038) – Level 3

October 1, 2003 to March 31, 2015

Currently collection for fiscal 2015/16

Yarmouth Regional (29056) – Level 3

Started collecting for fiscal 2015/16

Cobequid Community Health Centre (29061) – Level 1

April 1, 2011 to March 31, 2015

Currently collecting for fiscal 2015/16

Dartmouth General (29065) – Level 1

April 1, 2011 to March 31, 2015

Currently collecting for fiscal 2015/16

QEII (29085) – Level 1 – April 1, 2011 to March 31, 2015

Currently collecting for fiscal 2015/16

IWK Health Centre (29086) – Level 3 –April 1, 2003 to March 31, 2015

Currently collecting for fiscal 2015/16

Fishermen's Memorial (29114) – Level 3 – October 1, 2003 to March 31, 2015

Currently collecting for fiscal 2015/16

Day Surgery

- Collected through both the Discharge Abstract Database and NACRS in the past
- Collected through NACRS by South Shore Regional, Queens General and Fishermen's since October 2003
- Collected through NACRS by IWK since fiscal 2003/04
- Collected through NACRS by St Martha's starting with fiscal 2010/11
- All other hospitals started collecting day surgery information in NACRS beginning April 1, 2011
- Currently all hospitals collect day surgery information using NACRS
- From October 2003 until August 2009 South Shore Regional, Queen's General and Fishermen's also captured clinic information in NACRS
- For comparative reporting of day surgery between DAD and NACRS prior to August 2009, use intervention locations of '01' for Main OR and '02' for Endoscopy room only. Clinic visits cannot be excluded any other way in the database

- For day surgery reporting after August 2009, anything captured in Functional Centres that don't start with 71310^ are considered day surgery
- Day surgery interventions can be done in any Functional Centre in the Hospital – diagnostic imaging, outpatients, cardiac cath lab, OR, endoscopy room, etc.

Appendix E: Examples of Clinical Diagnoses in the Medical Complexity Category

Examples of Clinical Diagnoses in the Medical Complexity Category

Category	Clinical Examples
Progressive*	Cardiomyopathy; Heart Failure; Cystic Fibrosis; Bronchiectasis; Chronic Liver Disease and Cirrhosis; Spina Bifida; Chromosomal Anomalies; Transplant; HIV
Malignancy	Lymphoid Leukemia; Hodgkin's Disease; Malignant Neoplasms of the Brain; Neuroendocrine Tumors
Cardiac	Diseases of Aortic Valve; Hypertensive Heart Disease; Chronic Rheumatic Pericarditis; Chronic Pulmonary Health Disease; Cardiomyopathy; Cardiac Dysrhythmias; Other Congenital Anomalies of Heart
Craniofacial	Cleft Palate and Cleft Lip
Dermatologic	Chronic Ulcer of Skin; Congenital Anomalies of the Integument
Endocrinology	Diabetes Mellitus; Disorders of Adrenal Gland
Gastrointestinal	Diseases of Esophagus; Gastrojejunal Ulcer; Chronic Liver Disease and Cirrhosis; Other Disorders of Gallbladder; Intestinal Malabsorption; Diseases of Pancreas
Genetic	Chromosomal Anomalies; Other and Unspecific Congenital Anomalies
Genitourinary	Endometriosis; Other Disorders of Female Genital Organs; Other Disorders of Male Organs; Congenital Anomalies of Urinary System
Hematologic	Hereditary Hemolytic Anemias; Aplastic Anemia; Diseases of the White Blood Cells
Immunologic	Rheumatoid Arthritis and other Inflammatory Polyarthropathies; Ankylosing Spondylitis and other Inflammatory Spondyloapthies; HIV; Sarcoidosis
Mental Health	Schizophrenic Disorders; Psychoses with Origin Specific to Childhood; Neurotic Disorders; Personality Disorders; Drug Psychoses; Specific Delays in Development
Metabolic	Kwashuirkor; Vitamin Deficiencies; Disorders of Lipoid Metabolism

Musculoskeletal	Muscular Dystrophies and other Myopathies; Intervertebral Disc Disorders; Curvature of Spine; Traumatic Amputation; Fitting and Adjustment of Prosthetic Device
Neurologic	Cerebral Degenerations usual Manifest in Childhood; Spinocerebellar Disease; Multiple Sclerosis; Other Demyelinating Disease of the Central Nervous System; Hemiplegia; Cerebral Palsy; Epilepsy; Inflammatory and Toxic Neuropathy; Hearing Loss; Spina Bifida;
Ophthalmologic	Retinal Detachments and Defects; Glaucoma; Blindness and Low Vision;
Otologic	Otosclerosis; Vertiginous Syndromes and other Disorders of Vestibular System; Congenital Anomalies of Ear Face and Neck;
Pulmonary/Respiratory	Pulmonary Tuberculosis; Chronic Bronchitis; Asthma; Bronchiectasis; Post-inflammatory Pulmonary Fibrosis
Renal	Nephrotic Syndrome; Chronic Glomerulonephritis; Chronic Renal Failure; Urethral Stricture; Organ or Tissue Replaced by Transplant

*A progressive condition was defined as a condition that is associated with deteriorating health with a decreased life expectancy in adulthood (Simon, et al. 2014)

Appendix F: Screening for Caregiver Participant Eligibility Screening for Caregiver Participant Eligibility

1. Are you the legal guardian (caregiver) of a child (0-18 years old) with one or more diagnosed or suspected complex long-term health condition that has an impact on daily living?
    a. If yes – are you under the care of a team or a specialist at the IWK Health Centre?
    b. If yes – are you a primary resident of PEI, NS, or NB?

If 'yes' to these questions, they are eligible to participate in this study.

Appendix G: Caregiver Semi-Structured Interview Guide

Participant Study ID#: _____
Date Enrolled: _____
Current Date: _____

Introduction

Consent verbally

(*Will be personalized where applicable based on the child and family)*
Thank you for agreeing to speak with me today. This interview should take approximately 45-60 minutes and will be audio taped to ensure that I accurately capture all the key points that you share with me. The audio recordings will then be transcribed into written transcripts for analysis purposes. Any identifying information, for example health care provider or family names that you may mention throughout the course of our conversation will be removed from the interview transcripts. There are no right or wrong answers to any of these questions. We are interested in exploring the health resource use and health needs of children with medical complexity and their families. It may seem that some of the questions are similar, however each question has been asked a specific way to explore different concepts of family health. If you feel a question has already been answered or you wish not to response to any questions, please do not hesitate to say so. It is okay if some of your answers are repeated or if you wish to skip any questions. Please note that if you wish to end the interview at any point before I have asked all of the questions, or if you wish to withdraw from the study at any time during the interview, you are free to do so.

a. To begin, we would like to gather some background information about you and your family. If you prefer not to share this information, you may say 'pass'.
b. How old are you?
c. What is your gender?
d. What is your race/ethnicity?
e. How many members are in your family? (for example, do you have any other children, partner or key support individuals?) "Key supports" could also include individuals outside the family unit?
f. Are you currently employed?
g. How old is/are your child(ren)?
h. What is your child's gender?
i. What is your child's primary condition?
j. Could you describe your child's condition and needs to me such as use of medical equipment and other resources?
k. Where is your home community? (Rural/Urban/Suburban)
l. Do you have access to and/or use public/private transportation services?
m. When was your first hospital admission related to this condition?
n. How many days were you in hospital on this first stay?
o. Have you been readmitted since? If so, could you estimate how many times?

p. Who is a part of your child's health care team?
q. Does your child have a primary professional that manages /organizes your child's medical care?

Basic needs:

a) Would you consider all of your families' basic needs met? This would include needs such as housing, food, clothing, safety and security?
    a. If not, what is currently not being met?
    b. What resources (this could include people, services, and structures) could be in place to more adequately support these needs?
b) If so, are there any resources that you see as being important in supporting these needs?

Inclusive Education:

a) Does your child currently attend school or plan to attend school? Or daycare?
b) What resources are you aware of that are in place to support your child to attend and participate in school?
    a. How accessible are these resources?
    b. How often would you use these resources on average per year?
c) Are there resources that you would like to see in place or wish to access that may not be already available to help support your child to attend and/or participate in school?

Child Social Integration

a) Do you feel that your child is fully integrated within your community? This is related to freedom from bullying, discrimination, or neglect.
b) What resources are in place to support your child to interact within their social community?
    a. How accessible are these resources?
    b. How often would you use these resources on average per year? (if applicable)
c) Are there resource that you would like to see in place or wish to access that may not already be available to you or your community to help support your child in fully integrating within their community?

Child health-related quality of life

a) Are there any resources that you are using that help support your child's quality of life? This is in relation to the physical, emotional, and social aspects related your child's health and development status. (*Prompts to clarify may include: This includes activities that promote feeling loved and valued. This could be access to day programs, camps, etc.)*
    a. How accessible are these resources?
    b. How often would you access these resources in average in a year?
b) Are there resource that you would like to see in place or wish to access that may not already be available to you and your child to help support your child's health related quality of life?

Long term child and self-sufficiency

a) Do you feel that you have the resources, knowledge and support to care for your child child's condition now? In the future?
b) Are there any resources that you are accessing or have accessed to help support your ability to feel knowledgeable and confident in caring for your child and their health care needs? This may be related to information about your child's condition or developing the skills to care for specific needs.
    a. How accessible are/were these resources?
c) Are there resources would you like to see in place or wish to access that may not currently be available to help support your knowledge, skills, and/or confidence in caring for your child and their care needs?

Family social integration

a) Are there any resources that you use which help support you to fully participate in your childs life in order to remain active in the community that you live in? *(Prompt if needed– This may also include your ability to remain employed if you wish to do so, or socializing with others in your community)*
    a. How accessible are/were these resources?
    b. How often would you use these resources on average in a year?

b) Are there resources that you would like to see in place or wish to access that may not currently be available to help support you to fully participate in both your child's life and in your community?

Community system supports

a) What types of resources (social, physical, etc) are you using that help to allow your child to live in your home and home community? *(Prompts may include access to medical equipment, family support etc.)*
    a. How accessible are/were these resources?
    b. How often have or do you use these resources in average in a year?
b) Are there resources that you would like to see in place or wish to access that may not currently be available to help support you and your child to live in your home and home communities?

Health care system supports

a) What resources have you or do you access to support you, your child, and your family to obtain all necessary health care services or medical equipment? *(Prompt if needed – Health services at a hospital, etc, this could in private health insurance that covers dental, physio, etc,)*
    a. How accessible are/were these resources?
    b. How often have or do you use these resources on average in a year?
b) Are there resources that you would like to see in place or wish to access that may not currently be available to help support you, your child and your family to obtain all the necessary health services?

High quality patient-centered medical home

a) What resources have or do you access to support care coordination and/or navigating the health care system? *(Prompt if needed: this could be a social worker, a family care coordinator, or anyone at all that has helped you navigate and coordination health resources)*
    a. How accessible are/were these resources?
    b. How often have or do you access these resources on average in a year?
b) What resources would you like to see in place or wish to access that may not currently be available to help support care coordination and navigation?

Family-centered care

a) What kind of partnership do you feel like you have currently with your health care providers? *(Prompts if needed: Do you feel like you are a member of your care team, do you feel heard by your care providers? Do you feel included and supported in decision-making regarding your child's medical care whenever appropriate and possible)*
b) Do you feel included and supported in decision-making regarding your child's medical care whenever appropriate and possible?
c) Are there any resources that you use that have helped or help to support this care team relationships?
d) What resources would you like to see in place or wish to access that may not currently be available to help effectively partner with your health care team?

Additional Questions

a) Thank you for sharing information about your child with me. After listening to our conversation, what you would consider to be the highest priorities for your family / child needs? For example, you mentioned using ____?
b) Those are all the questions that I have for you today, however, I would like to ask if you if there is anything else that you wish to share with me regarding your experience, resources and supports you've accessed or wish to access, or unmet care needs that you wish to further elaborate on or that we have not yet discussed?

Appendix H: Draft Family Health Resource Journaling Template

Date	Type of Resource	Additional Details
Example: June 19th	-Physiotherapy Home Visit -Home Care Nurse Visit	-Took day off work to meet with the physiotherapist and home care nurse
Example: June 22nd	-Interdisciplinary Clinic Visit	-Two hours round trip drive to the hospital

Appendix I: Care Team Semi-Structured Interview Guide

Thank you for agreeing to speak with me today. This interview should take approximately 15 minutes and will be audio taped to ensure that I accurately capture all the key points that you share with me. The audio recordings will then be transcribed into written transcripts for analysis purposes. Any identifying information, for example other care providers or family names that you may mention throughout the course of our conversation will be removed from the interview transcripts. There are no right or wrong answers to any of these questions. We are interested in exploring the health resource use and health care needs of children with medical complexity and their families. Please note that if you wish to end the interview at any point before I have asked all of the questions or if you wish to withdraw from the study at any time during the interview you are free to do so. Today we are talking about the *insert child's name* family's experience in particular.

a) What are the key resources that you perceive as essential to supporting this family?
b) Are there any resources and/or supports that you wish that you and this family had access to better support this family's needs?

   Potential Prompts: This could include social and education needs, in addition to specific medical treatments

   Potential Prompts: Has there been any major barriers or facilitators to providing care?

Appendix I: Journal Copyright Forms

The manuscript with the title Children with Medical Complexity in the Canadian Maritimes: A Mixed Methods Study Protocol authored by Sydney Breneol, Janet A. Curran, Marilyn Macdonald, William Montelpare, Samuel A. Stewart, Ruth Martin-Misener, Jocelyn Vine ("authors") has been accepted for publication by JMIR Publications ("publisher"). A signed copy of this form must be on file with JMIR Publications before the manuscript can be published.

Manuscript #: 33426
DOI: 10.2196/33426

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