Towards a Clinical Definition of Children with Medical Complexity: A Delphi Process

by

Kyle Millar

A Thesis submitted to the Faculty of Graduate Studies of

The University of Manitoba

in partial fulfilment of the requirements of the degree of

MASTER OF SCIENCE

Department of Community Health Sciences

University of Manitoba

Winnipeg

Table of Contents

1. ABSTRACT

INTRODUCTION: Children with medical complexity (CMC) are a subpopulation of children with chronic disease who live with extreme medical and social impairment. Identification of these children remains vague and controversial but in 2011, Cohen *et al.* formulated a conceptual definition composed of four required domains: *Family Needs, Healthcare Use, Functional Limitation,* and *Diagnostic Conditions*. Unfortunately, there has not yet been agreement on the specific criteria to fulfill each of the four domains. A consistent definition is an important requirement for clinicians, researchers, and policy makers to describe this growing population, understand their needs, develop evidenced-based policy and research management strategies and models of care.

OBJECTIVE: The first objective of this research was to search the literature for variables associated with CMC and that would theoretically fulfill one of the domains. The second objective was to convert these variables into items which had the potential to fulfill their respective Cohen *et al*. 2011 domain, as determined by an expert panel in a Delphi study. The overall goal was to create a standardized definitional tool for CMC.

METHODS: A scoping review of the CMC literature was completed and variables that could theoretically fulfill one of the four domains were extracted. These variables were converted to items which were collated into a survey delivered to an expert panel. Researchers and clinicians with CMC expertise were gathered from a variety of professional societies to undergo a four-iteration Delphi procedure.

RESULTS: From the literature, 1411 studies were considered and 132 of these informed 55 items which were presented to the expert panel. Eighty-two experts were recruited from literature

authorship and professional societies to complete the Delphi study. With an average response rate across iterations of 70%, these experts reached consensus for inclusion on 48 items and reached consensus for exclusion of 1 item, for a total consensus rate of 89%.

CONCLUSION: The consensus items were collated into a definitional tool for CMC with 25 items presented to fulfill the *Family Needs* domain, 10 items in the *Healthcare Use* domain, 2 items in the *Functional Limitations* domain and 1 item in the *Diagnostic Conditions* domain. These results represent the first step to create a standardized definition of CMC.

2. ACKNOWLEDGEMENTS

I will be forever grateful to the people that allowed me to take on an ambitious project and complete it successfully. I required a lot of guidance as there were many components of this research that I was doing for the first time. The guidance the following people provided allowed me to grow so much as a researcher and allowed this project, and my MSc, to come to a successful completion.

1. Dr. Allan Garland was my MSc supervisor but was also very generous as my research and career mentor. He taught me how to do research, be a researcher, be an academic, and be a successful clinician. He was so generous with his time.

2. Dr. Eyal Cohen was an expert in the field of pediatric complex care and guided me through the research complexities of studying these amazing children and the clinical complexities of caring for these children and their resilient families. I will always be grateful that he found so much time to open up the world of academic pediatric complex care to me.

3. Dr. Gina Rempel has taught me so much over the past 5 years and has laid the foundation for any future work with Children with Medical Complexity in Manitoba. She taught me to be a complex care pediatrician and is an encyclopedia of all literature ever published on complex feeding and on Children with Medical Complexity. She guided the clinical relevance of my research and constantly reminded me to consider the clinical and social context of any research with these children.

4. Dr. Celia Rodd taught me the solid foundations of pediatric research. She organized my thoughts and my actions. She is so generous to share her years of experience in pedaitric research and was never reluctant to make time for a new learner like me.

5. Dr. Kathyrn Sibley was a critical voice reminding me of the principles of quality research, specifically around qualitative and Delphi methodology. Research must always have parental input, I will never forget.

6. The Centre for Healthcare Innovation allowed me to deliver my REDCap survey, particularly Mr. Abin Chandrakumar who was as dedicated to the project as anyone.

7. The Michael Nesbitt Family Complex Care Fellowship funded all expenses of the MSc project as well as countless projects in clinical Complex Care. Nothing would be accomplished without this generous endowment, vision, and commitment.

3. LIST OF TABLES

4. LIST OF FIGURES

5. BACKGROUND

5.1. CHILDREN WITH MEDICAL COMPLEXITY

Children with medical complexity (CMC) are children with chronic disease but with the most pronounced medical impairment, functional limitations, and family needs. Although these problems result from a wide variety of specific medical conditions, CMCs share similar limitations independent of diagnosis including heightened community care needs, higher dependence on polypharmacy and technology-augmented care, greater health care system utilization, and higher need for care coordination.[1]

Population prevalence estimates have been extremely variable due to inconsistent definitions between studies and differing populations under study. Four previous studies, all of which were cross-sectional, reported point prevalence varying nearly 5-fold, from 0.20-0.95% of children.[2-5] Using differing definitions of CMC, these studies examined: 1) the in-hospital, age-standardized rates of CMC across Canada, 2) the in-hospital rates of CMC in Ontario, Canada, 3) the crude rate in the North Carolina, USA Medicaid population, and 4) the crude rate in a Washington state, USA health insurance network.[2-5] Though these studies did not specifically explore secular trends, there are three reasons to suspect increasing prevalence of CMC. First, since the 1960's there has been a 400% rise in prevalence of children with chronic disease – a broader term that includes both CMC as well as other children.[6, 7] Second, CMCs are having an increasing impact on the pediatric inpatient system demonstrated by increasing proportion of hospital days, admissions, readmissions, preventable adverse events and inpatient charges.[8-13] Third, there has been increased survival of many important groups of

children including premature infants, children born with congenital anomalies, and oncology survivors; groups at high risk of being medically complex due to the severity of their medical and functional deficits. [14-18]

There is increased attention recently on these children as their care puts a disproportionately heavy stress on health systems and caregivers. Despite composing <1% of the pediatric population, CMC account for a large amount of healthcare expenditure.[2-5] During 2005-2007 these children utilized 33% of all the provincial pediatric health care expenditures in Ontario.[2] Among children in Canada from 2015-2016, CMC account for 37% of hospital admissions, 54% of inpatient days and 57% of hospital costs.[5] During 2015-2016 hospital costs were estimated to be $866 million in the two years after diagnosis.[5] The increased healthcare system utilization and hospitalizations also place demands on caregivers and is one of many stresses caregivers face to meet the increased needs of these children. Caregivers of CMCs experience elevated levels of psychosocial stress, health deterioration and financial pressures due to the care required of their children.[19-25]

5.2. THE NEED FOR A DEFINITION OF CHILDREN WITH MEDICAL COMPLEXITY

As mentioned, studying the epidemiology of these children is difficult as a standardized definition has not yet been established; CMC prevalence is likely increasing; and the impact of these children on caregivers and the healthcare system is disproportionately high. Additionally, the needs of these children and their families are not being met by traditional models of healthcare delivery as these models were never designed to care for children with this degree of medical and social need, care coordination, technical expertise required from the care team,

and levels of resource need.[26-29] All of these factors account for why care providers and policy makers are recognizing that this population requires alternative methods of healthcare delivery.[26-29] Both the American Academy of Pediatrics and the Canadian Pediatric Society have called for the standardization and investigation of the optimal models of care and management strategies for CMC.[30-33] However, evidenced-based progress in caring for these children has been hampered by the lack of "a uniform definition that is clear, reproducible, and comparable across studies."[24, 31, 34-50]

The absence of a standardized definition of CMC hampers progress in multiple ways on the population level, health systems level and individual level. The identification of CMCs on the individual and health systems level requires a standardized clinical definition. Although efforts have been made to create a standardized, clinical, definitional framework for these children (Section 5.4), practical recognition of these children remains a subjective exercise, drawing on a care providers experiences and held perceptions.[36, 51] Identification of CMC on the population level has so far been attempted with administrative data definitions and population-based surveys, but these definitions have never been validated against a reference-standard clinical definition.[51] For these reasons, a call to develop a standard definition has been made by multiple thought leaders in the field over the past two decades.[31, 34-36] The need for this call can be illustrated with examples from the population-level, systems-level and individual-level study of CMCs.

5.2.1. POPULATION LEVEL

On the population level, the difficulty in measuring prevalence rates of CMC exemplifies the problem of inadequate definitions. Calculating the population prevalence of these children, a fundamental descriptor required for research and policy, has been hampered by varying definitions employed in these studies.[2-5] In four studies, prevalence estimates range over 4-fold, from 0.22 to 0.95 children per 100 000 children. Although these differences may reflect the populations under study, the studies used widely different definitions of CMCs. Two studies used the Complex Chronic Conditions administrative definition (Section 5.3.2.1), but with slightly different methods to stratify into subclasses within the definition.[2, 5] Another used the Clinical Risk Grouping administrative definitions (Section 5.3.2.3), and the last study used an administrative listing of assistive technology placement codes (Section 5.3.3.1).[2-5] Of course, these studies employed administrative data definitions which often, by nature, differ due to the availability and quality of data in the administrative data sources. It must be noted that administrative data definitions have advantages as the datasets are readily available, they do not require the substantial effort to prospectively collet data from patients, caregivers or clinicians, and they are increasingly being used in population health. However, efforts to validate administrative data definitions are not possible without a reference-standard clinical definition of CMC.

5.2.2. HEALTH SYSTEMS LEVEL

The needs of CMC are not currently met by existing hospital-based and community structures, necessitating research into the optimal care delivery models for CMC.[31, 52, 53] There are three main care models currently employed in the pediatric complex care field: 1)

primary care centred models, 2) complex care centred models, and 3) episode-based models.[37, 50, 54] However among structured complex care clinical programs, there is considerable variability even when the same model is being used.[37, 50, 54] The variability both between and within clinic models illustrates the lack of evidence and consensus on best practices to manage these children.[37]

First, *primary care centred models*, or medical home models, refer to models where primary care and care coordination is provided by the primary care provider (PCP) who may be a general practitioner, nurse practitioner, general pediatrician, complex care pediatrician or another pediatric subspecialist. The PCP is often supported by a multi-disciplinary team which may be composed of nursing, social work, allied health, or coordinator support. However, these clinics are often separate from the subspecialty care needed by CMCs and the PCPs often find it difficult to provide the specialized care needed. In our current climate, this clinic model seems to be preferred by health and policy-making organizations to provide care coordination.[37, 50, 54-64]

Second, in *complex care centred models,* specialty care and care coordination are provided by complex care pediatricians or nurse practitioners in consultation to, or comanaged with, primary care providers. These clinics are often integrated within tertiary care centres, as much CMC care is delivered in that setting. A tertiary care locus also allows proximity to the child's other specialized care providers and related resources.[5, 8, 11, 38, 65-69] Unfortunately, these clinics are often removed from the patient's local community and the providers usually do not provide primary care to the patient and their family.[37, 54]

Third, *episode-based models* refer to programs in which care coordination and medical management are delivered during specific episodes, discrete in time and location, during times of highest need and caregiver stress. Often these teams are involved during acute illnesses in the inpatient or transitional care home settings. There is usually less longitudinal and preventative care of these children with this model.[37, 54, 70]

There are many published evaluations of the efficacy of different complex care clinical models.[54, 61, 71] These evaluations vary in the nature of the intervention, research design, and outcome measures, but many have suggested benefits of complex care models. Improved outcomes included those related to effectiveness of care (e.g. disease specific outcomes and healthcare use), efficiency of care (e.g. healthcare use outcomes and costs), patient or family-centredness of care, patient safety (e.g. frequency of acute illnesses), and timeliness of care (e.g. timely access to care).[38, 50, 58, 60-62, 66, 68-71] Although outcomes seem to be improved by these models, the complex care interventions themselves are quite divergent with some nicely fitting into one of the three model designs and other interventions being a hybrid of multiple designs.[54, 61, 71] There have been no head-to-head comparisons between these three models to guide optimum care delivery.[37] Furthermore, the study populations are varied, including various disease-specific populations (e.g. cystic fibrosis), and various heterogeneous populations (e.g. children born premature).

In the absence of a standard definition of CMC, the identification of an optimal performing care model and determining the generalizability of this care model is problematic.[37, 50] In fact, without a standardized CMC definition it is unclear which children

currently participating in complex care clinics would benefit from a complex care program in the first place.

5.2.3. INDIVIDUAL LEVEL

Determining standards of care on the patient level has been a challenge with the CMC population. Evidence is lacking regarding the inpatient and outpatient care of CMC, even commonly experienced acute and chronic conditions. An example is the management of pneumonia secondary to aspiration. It is relatively common, accounting for 9.7% of all admissions for pneumonia in children with neurologic impairment and 10.0% of all admissions for pneumonia in the general pediatric population.[72, 73] Compared to all children with aspiration pneumonia, CMCs with aspiration pneumonia have longer hospital lengths of stay, greater likelihood of needing ICU care, higher hospital costs, and more frequent hospital readmissions.[72, 73] Despite its prevalence, there are few trials to guide therapy for aspiration pneumonitis.[74-77]

Extrapolation of this limited evidence to the entire CMC population is difficult as each study used a different definition of CMC. These studies were performed on populations defined by a shared condition (e.g. cerebral palsy) or a by a more narrowly defined clinical entity (e.g. neurologic impairment). CMC as a group have variable underlying clinical conditions as they are primarily defined by shared needs. To optimize and generalize the treatment of common conditions, like aspiration pneumonia, in CMCs, a clinical definition of these children is necessary.[65]

5.3. PREVIOUS DEFINITIONS OF CHILDREN WITH MEDICAL COMPLEXITY

Before the pediatric complex field arrived at the current clinical definitional framework (Section 5.4), there was a lineage of definitions of various, related populations. Initially, efforts were taken to identify children with chronic conditions. The concept of "Children with Special Healthcare Needs" (CSHCN) (Section 5.3.1) was the initial effort to define a broad population of children with chronic conditions. With time it was recognized that a subset of CSHCN exhibited extreme fragility and care needs, and researchers began formulating definitions for this population, that eventually led to the concept of CMC. Large, uncoordinated efforts led to various administrative data, clinical and research definitions for CMC. These definitions focused on listing diagnostic conditions perceived to be complex (Section 5.3.2), using measures of functional limitation (Section 5.3.3) increased healthcare system utilization (Section 5.3.4), and/or family or social needs (Section 5.3.5).[51] Each of these definitional strategies have a place in the literature depending on the question being asked and the population under study. However, each has limitations when attempting to capture the children seen in Complex Care clinics. It was apparent the four definitional strategies described components of medical complexity; they would later inform the domains of the currently accepted clinical definitional framework for CMC (Section 5.4).

5.3.1. CHILDREN WITH SPECIAL HEALTHCARE NEEDS

Early on, multiple groups attempted to define a group of "children with special healthcare needs."[78, 79] The CSHCN concept originated from the Maternal and Child Health Bureau in the United States, which proposed a broad definition of children with chronic disease as "those who have or are at increased risk for a chronic physical, developmental, behavioral, or

emotional condition and who also require health and related services of a type or amount beyond that required by children generally."[78] However this definition did not delineate children based on their social complexity, or differentiate the degrees of functional limitation or resource needs seen among them.[80-82] Such delineations are important to differentiate between children with chronic disease who are also medically complex (Section 5.4) from children with acute, time-limited conditions, or children with chronic disease but lesser complexity.[35] Differentiation of these groups is important as resource needed and outcomes will differ greatly. This led to future efforts to define these complex children termed CMC.

5.3.2. DEFINITIONS UTILIZING UNDERLYING MEDICAL CONDITIONS

Discussed in this section are four systems of grouping diagnostic codes from pediatric administrative data sources to identify children with medical conditions considered complex. Though not all of these were designed for the specific purpose of identifying CMCs, they have all been used for that purpose. These four systems are the most prevalent in the literature, but numerous other groups have attempted to define a medically complex population based on a limited number of handpicked diagnostic codes customized to their study needs or data limitations.[58, 66, 83-86]

5.3.2.1. COMPLEX CHRONIC CONDITIONS (CCC)

Fuedtner *et al.* produced a list of International Classification of Diseases (ICD) codes of complex chronic conditions (CCC) that was first published in 2000, with a further update in 2014.[87, 88] Intended to be applied to administrative data, a condition was selected for inclusion in the list of CCCs if it, "*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." This list has become commonly used when describing CMCs with adminstrative data. Compared to the general pediatric population and to CSHCN, children with these conditions have been shown to have a higher proportion of childhood deaths[87], increased inpatient costs[8], increased admissions,[8] increased readmission rates[12, 89, 90], increased inpatient length of stay,[8] increased inpatient technology assistance procedures,[8] higher emergency department use,[91] and higher preventable admissions.[92]

This definition makes the consequential assumption that all children with a shared CCC are homogenous, which is limited for several reasons. First, children with a shared CCC have variability in their needs, functional limitations, complexity, severity, and fragility. Second, many of the children clinically idenitifed as having CMC do not have a diagnosis conforming to a specific ICD code. Last, this definition excludes children with: a) minor chronic conditions not included in the CCC list may actually be considered complex due to other unconsidered pyschosocial or functional reasons or b) children with multiple non-complex chronic conditions whose additive effect leads to a high degree of complexity.

5.3.2.2. PEDIATRIC MEDICAL COMPLEXITY ALGORITHM

In 2014, the Pediatric Medical Complexity Algorithm (PMCA) was developed by researchers at the Seattle Children's Hospital in an attempt to define levels of medical complexity for children.[93, 94] Using administrative datasets, this algorithm utilizes a child's history of ICD code assignment to classify children into 3 levels of complexity: 1) complex,

chronic disease (CCD), 2) non-complex, chronic disease (NC-CD), and 3) without chronic disease (without CD). This algorithm performs best with a minimum of 2 years of retrospective data, which may add a cumbersome time-lag to diagnosis. Unlike CCC, validation in the form of association with relevant outcomes is lacking. However, when compared to a clinical standard population we do know that the sensitivity of the algorithm for children with CCD is 84-89%, 41-45% for NC-CD, and 80-96% for those without CD.[93, 95] Specificity for identifying these conditions was good, ranging 85-92% for the three classficiations. However, this framework is based on grouping ICD diagnostic codes and shares the same pitfalls as the CCC (Section 5.3.2.1) when using it to clinically identify CMC.

5.3.2.3. CLINICAL RISK GROUPINGS

Clinical Risk Groupings (CRGs) is another ICD-based diagnostic algorithm designed for use with administrative datasets in adults and children.[96] A proprietary system using both inpatient and outpatient administrative data developed by 3M Health Information Systems,[96] it identifies patients within the hierarchy of CRGs when incorporating a minimum of 3-year retrospective history of ICD diagnoses and procedure codes for children. However, the CRG methodology can function on any amount of data with varying degrees of performance. The system categorizes children into nine mutually exclusive CRG categories. This spectrum is anchored by the categories: (1) healthy children with an acute condition and, (9) unhealthy children with a catastrophic condition. This system shows increased flexibility compared to other diagnostic algorithms in that the number of comorbidities and involved organ systems is considered when assigning risk groups, enabling children with the same primary condition to be assigned to differing categories.

The increasing severity of the CRG categories has been shown to be associated with increased healthcare encounters, hospitalizations, total hospital days, and healthcare expenditures.[10, 97] There has been a single study testing the performance of the CRG system in identifying CMCs. A clinical expert created a reference-standard population by individually reviewing 700 children and placing them in three groups that corresponded to the groups of the PMCA.[98] The nine CRG categories were then collapsed into three which corresponded to the PMCA categories. Those collapsed CRG groupings accurately categorized the 700 children into the three expert's groups with a specificity of 84-95% and sensitivity of 75-98% for CCD and NC-CD. The sensitivity for NC-CD was poor, ranging 31-58%.[98] Although this algorithm can assign children along a spectrum of nine CRGs, and shows respectable validity, it remains unclear where along this spectrum one should place the threshold for considering an individual to be a CMC.[51] Additionally, this framework is based on grouping ICD diagnostic codes and shares the same pitfalls as the CCC and PMCA (Section 5.3.2.1, Section 5.3.2.2) when using it to clinically identify CMC.

5.3.2.4. CHRONIC CONDITION INDICATOR

The Chronic Condition Indicator (CCI) is the least common of the four presented diagnosis-based definitions for CMC. It is an open-sourced algorithm meant for adminstrative datasets. It categorizes children using ICD codes into one of 18 major clinical categories, with each of the 18 further dichotomized into chronic or non-chronic condition groupings.[99] Unlike the previous algorithms, this was originally designed to be used for adults, but it has been adapted for the pediatric population and has shown usefullness in this population.[100, 101] Its major weakness is an inability to discriminate between complex and non-complex

conditions.[51] Attempting to circumvent this limitation, some authors have counted the number of chronic clinical categories assigned to a child as a measure of complexity.[100] Other authors incorporate other classification systems into the CCI system to subclassify chronic clinical conditions by complexity.[51] Despite its flaws, the number of assigned major clinical categories positively correlates with health resource use.[100] However, this framework is based on grouping ICD diagnostic codes and shares the same pitfalls as the three previous frameworks (Section 5.3.2.1, Section 5.3.2.2, Section 5.3.2.3) when using it to clinically identify CMC.

5.3.3. DEFINITIONS UTILIZING FUNCTIONAL LIMITATIONS

Defining medical complexity solely by clinical conditions is incomplete. The heterogeneity of complexity between children that share the same condition is substantial. Concurrent functional limitation, social consequences, and healthcare system needs all can influence the amount of complexity associated with a given clinical condition. Specifically, the degree of functional limitation experienced by a child is an important descriptor for children with chronic disease as it is an important consideration for clinical management and resource allocation. For example, children with greater functional limitation need more assigned supports through the medical, school, and homecare systems. This has direct bearing on resource allocation and policy planning. Additionally, different functional limitations experienced by children with the same clinical condition impose differing risk for other clinical conditions. For example, some children with cerebral palsy will experience a feeding dysfunction which may then lead to failure-to-thrive and/or aspiration-induced chronic lung disease. As such when caring for a child it is important to consider their state of functional

limitations because the severity vastly alters a child's experience, care needs, and risk of other conditions. For such an impactful factor in a child's healthcare it is important to consider it when defining complexity. A discussion of methods to identify functional limitations is presented in this section.

5.3.3.1. PRESENCE OF TECHNOLOGY ASSISTANCE

Level of functioning in children is a difficult concept to define and collect in adminstrative data and often in clinical data as well. A practical method to assess some aspects of functioning is to collect information regarding assistance on medical technology, including devices to aid mobility, feeding, and breathing. Such technologies, used to support vital life functions and activities of daily living, provide indirect information on moderate-to-severe dysfunction. Greater insight may be gained into the functional status and degree of health care need with this consequence-based method than with diagnosis-based definitions alone. Indeed, one-third of children with medical complexity defined using the complex chronic conditions (Section 5.3.2.1) system are assisted by medical technology, and as will be shown, such assistance is associated with detrimental healthcare consequences.[31]

Many, but not all, such devices are captured in administrative datasets using procedure coding for technology placement. Such coding systems include ICD procedure codes, procedure terminology codes, durable medical equipment codes, and Canadian Classification of Interventions codes. Studies using procedure coding systems to define medical complexity in population datasets have demonstrated that need for medical technology is associated with higher Medicaid expenditure, death, and hospital readmission.[3, 12] Other studies have used

technology assistance codes as a stratifier within diagnostic code algorithms. These approaches demonstrate that within diagnostic categories, technology assistance is associated with greater mortality, home care use, number of involved physicians, hospital use, emergency department visits, and healthcare expenditure.[2, 31] Multiple other studies have used specific billing codes for medical technology placement tailored to their research needs to identify children with variations of complexity and fragility. Examples of such codes include procedural codes for tracheostomy placement and cerebrospinal fluid shunt placement. [102, 103]

Unfortunately, there are limitations to this method of identifying CMC. Firstly, like diagnosis-based definitions, there is a spectrum of fragility and complexity that exist within any given level of technology assistance. For example, possessing a gastrostomy tube may be suggestive of a child with severe complexity and fragility, however it may also be seen in a child with high functioning, low needs, and relatively low complexity. Second, the presence of technology assistance only identifies a narrow segment of the greater functional limitation concept. The World Health Organization's International Classification of Functioning has a detailed taxonomy of functional limitation that includes limitations in body structure and function, activities and participation (section 9.3.3.1).[36, 95] Unfortunately, the presence of technology assistance only identifies children with more severe limitations in body structure and function, and does not identify children with limitations in activities and participation, nor children with limitations in body structure and function without the need to technology assistance.

5.3.3.2. SURVEY-BASED DATA COLLECTION

The predominant survey collecting functional limitation data is the National Survey for Children with Special Healthcare Needs (NS-CSHCN). It is an on-going, nation-wide survey in the U.S.A. administered by the Department of Maternal and Child Health (https://mchb.hrsa.gov/). It is meant to identify and describe CSHCN.[104] The NS-CSHCN uses a standardized, validated, open-source survey script to identify these children and ask about consequences experienced by CSCHN and their families. Prior to administering the greater survey, the identification CSHCN is completed using five-question CSHCN screener (CS) which includes questions to caregivers about [105]: 1) need for prescription medications, 2) need for services, 3) need for specialized therapies, 4) presence of functional limitations, and 5) presence of emotional, behavioural or developmental differences. An affirmative answer to any of the five questions fulfills the definition. Therefore the presence of a functional limitation is all that is required to identify a CSHCN. Beyond the screener, the greater survey collects information on limitations in breathing, swallowing, eyesight, hearing, mobility, communication, and self-care are identified, with severity of these assessed through a 3-point Likert scale.

Authors have utilized this screener to identify CMC using solely functional limitations as criteria, independent of diagnosis. These authors have concluded that compared to other CSHCN, CSHCN with functional limitations have a greater number of health conditions, are less likely to have adequate insurance, have higher family impact and medical costs.[80] Additionally, other authors have used the screener to identify CMC using the presence of functional limitations paired with other realms of the screener, such as increased healthcare needs. These authors have demostrated CMC, identified in this way, have increased healthcare

utilization and higher healthcare costs compared to the remaining CSHCN.[106] Unfortunately, defining CMC in this way has not been validated for use in clinical enrollment and is not in common research use, limiting our understanding of its real-world relevance. Additionally, it requires individual contact by telephone, or internet-based methodologies, and using this method on the population level is impractical and resource-intensive.

5.3.4. DEFINITIONS UTILIZING HEALTHCARE SYSTEM UTILIZATION

The use of increased healthcare utilization is an attractive method of defining CMC. It includes variables that are easy to collect as most adminstrative datasets record instances of resources use, such as hospitalizations or outpatient clinic appearances. When using healthcare utilization as a method to identify CMC, the rate of hospitalizations is the most common way of doing so.[12] It can be argued that healthcare use, like hospitalization rates, are a downstream event indicative of disease severity, inadequate management of needs, or severity of functional limitations.[12, 51] Thus increased healthcare use could be viewed as a proxy of these three domains of complexity. Regarding hospitalizations specifically, CMC, identified in a number of different ways, often demonstrate greater rates of hospitalization compared to the general pediatic population. [8, 12, 101, 107] However, many studies demonstrate that a majority of CMC, identfied by a variety of ways, do not present to hospital at all in a given year thus limiting its sensitivity as a idenfier of CMC.[8, 12, 101, 107]

5.3.5. DEFINITIONS UTILIZING OVERALL SOCIETAL AND FAMILY NEEDS

While the presence of a clinical condition, functional limitation and increased healthcare use are important contributors to defining CMC, many caregivers and providers value the

presence of increased need as a fundamental defining feature of complexity.[51] The needs take many forms including the requirement of treatments, attendance at a high number of appointments, arrangement of specialized transport, need for financial support, and the need for caregivers to deliver treatments, therapy, and care coordination. These needs may be experienced across locations including home, school, or residential living facilities. Increased needs are vitally important when defining these children because the identification of children with these needs is important in policy planning and resource allocation.

The care and social needs of these children are vast, varied, differing in intensity and individual-specific. The NS-CSHCN collects information on child need and demands on family including information on the child's needs from the healthcare and educational system, needs for care coordination, and needs for respite care.[104] Additionally, many authors have defined a large array of needs and used these needs in population definitions, independent variables, covariates, and outcome variables (Appendix 1 – "Family Needs" variables). Regarding population definitions, some authors have used narrowed aspects of this "needs" concept, such as residence type or unmet care needs, to define these children. Populations that are at least partially defined using "needs" variables have been shown to have high rates of healthcare system contact and medical technology placement.[24]

5.4. CONCEPTUAL DEFINITION OF COHEN *ET. AL*. 2011

The previous work on defining children with chronic conditions, and the many methods of defining CMC using ICD lists, measures of functional limitation, measures of healthcare utilization and measures of social need informed the evolution towards a more holistic

diagnostic framework for CMC. With regards to a clinical definition, there is general agreement that, in addition to medical diagnosis, these children must meet some combination of medical and social criteria delineating them as complex.[31, 32, 34-36] A systematic review by *van der Lee et al. 2007* demonstrated that 64 previous attempts to define children with chronic health conditions shared features including: 1) the presence of a chronic condition, 2) the presence of limitations in activities of daily living due to the condition, and 3) an increased need for medical care or related services. [108] Building on this, Cohen *et al. 2011* noted that previous attempts at defining CMC encompassed similar attributes but no definition included all of them. [36, 80, 109-115] Therefore, those authors proposed a conceptual, clinical definition in which a child must possess at least one component in each of four domains to be considered a CMC: a) increased caregiver, medical, therapy and education needs, b) presence of a chronic condition, c) presence of a functional limitation, and d) increased health care utilization (Figure 1).[36] Since its introduction, this framework has been broadly adopted internationally and in Canada it is the basis of a pan-Canadian guideline for the management of medically complex children proposed by Children's Healthcare Canada (formerly, the Canadian Association of Pediatric Health Centres).[31, 35, 36, 116, 117]

Despite the general acceptance of this conceptual definition, standardization of these four domains have yet to be performed. As a result, there is substantial variability around what constitutes a chronic condition, increased healthcare usage, family need and functional limitation. Healthcare providers, policymakers, and researchers have been left in the position of individually interpreting the meaning of these domains.[118] Thus, the numerous definitions used by complex care programs across North America limits generalizability of clinical

interventions aimed at them, and confuses entrance criteria for current and future programming and policy.[24, 31, 39-49] Cohen 2011 highlights that "a uniform definition that is clear, reproducible, and comparable across studies" is crucially needed by clinicians, researchers and policy makers for activities such as the creation of inclusion criteria for clinical intervention, international standard of care recommendations and policy interventions to ensure equitable resource allocation.[34-36]

5.5. CONSIDERATIONS REGARDING THE COHEN 2011 CONCEPTUAL DEFINITION

Three considerations must be acknowledged with regards to the creation of a standardized clinical definition based on the Cohen *et al*. 2011 framework. First is the under-representation of social determinants of health (SDH) in the conceptual definition.[35] SDH are recognized as critical contributors to health outcomes (Appendix: Figure 2).[119] Although the conceptual definition lacks direct reference to SDH, they are indirectly represented in the 4 domains, considering SDH influences healthcare expenditures, functional limitations, and family needs.[119] Family needs (defined as "substantial family-identified service needs" or "significant burden on family") have significant overlap with SDH.[35] Thus research on this topic should consider topics related to SDH when creating a clinical definition.

Second, any CMC definition must consider that complexity in childhood is a dynamic state. Social and medical complexities change with age, growth, evolution of the caregivers' state, and community programming.[31, 35, 36] Thus, research on this topic must consider temporal influence on items that compromise the clinical definition.

Third, a definitional framework that allows multiple ways to fulfill each domain will likely identify children with considerable heterogeneity. Outcomes, needs and clinical programming

would be expected to vary greatly within the broad definition, and it may be beneficial to subclassify these children based on these characteristics.[31, 34, 35] However, the need for subclassification does not negate the need for an agreed-upon broad definition of CMC. The importance of this initial broad definition is two-fold: (i) these children share some similar overarching needs and characteristics, (ii) the initial definition will allow a starting point for subclassification, with future work needed to identify CMC subcategories.

5.6. AN INTRODUCTION TO THE DELPHI METHODOLOGY

5.6.1. WHAT IS THE DELPHI TECHNIQUE?

In all fields of work, including medicine, there are countless questions that lack guidance from evidence. The question may be difficult to study or it may never have been prioritized to study in the past. Nevertheless, decisions must be made in the face of this ignorance. Informally, day-to-day decisions are made using clinician experience or by extrapolating evidence from related topics. Formally, guidance may be provided by collating experts' opinion into clinical guidelines. Like the individual clinician, the expert opinion may be based on extrapolated data or individual experience. Classically, clinical guidelines were generated at a meeting or a conference, attended by experts in the field. More recently questions that are difficult to study have been tackled using consensus building techniques such as the Nominal Group technique or the Delphi technique.

The Delphi technique is a systematic, group communication methodology designed to reach consensus on inherently subjective questions where the current body of literature does not provide adequate guidance.[120-124] Philosophically, the technique relies on the concept

that there can be multiple views of reality, and if different views can be shared and considered then individual views can be altered and a shared understanding can be approached.[120, 121, 125] Therefore, the goal of the technique is to collectively consider multiple expert opinions and stabilize them over time into a group consensus.[120, 126] The Delphi technique requires the following basic foundational elements:[121, 127, 128]

i. Identify an appropriate research problem.

ii. Complete a literature search on the problem.

iii. Develop a questionnaire to present components of the question for group consideration.

iv. Collect an expert panel of questionnaire respondents.

v. Perform iterative survey rounds to reduce or categorize the components of the question.

vi. Determine consensus thresholds to guide reduction and categorization.

vii. Provide feedback to respondents interspersed between rounds to allow reconsideration of opinions.

viii. Analyze and summarize the findings.

The Delphi technique was originally developed by the RAND corporation to describe and discuss the administration and impact of war in the 1950s.[127] Since this time a wide variety of fields have taken up the methodology to answer ambiguous questions, including in medicine. Over the years, the original methodology has been incrementally adjusted to the needs of

individual studies resulting in the evolution and branching of the technique. In fact, there are presently a number of versions of the Delphi technique using names like “modified Delphi”, “policy Delphi”, and “real-time Delphi.”[127] Even the Rand corporation has adjusted their methodology into the RAND appropriateness method which is a variant of both the Delphi technique and the Nominal Group technique. [129, 130] Although most Delphi studies share the same basic foundational elements, there is currently no standardized methodological template for a modern Delphi study.[127, 128, 131]

5.6.2. HOW THE DELPHI TECHNIQUE IS CURRENTLY BEING USED

There are several instances where the Delphi technique is a useful tool to answer complex questions. First, the technique can be useful when the question relies on the melding of subjective statements from experts due to paucity of evidence on the matter.[127, 131] Second, when more experts are involved than can functionally interact in face-to-face meetings then the Delphi technique may be useful.[127] Last, the Delphi is useful if anonymity is critical because disagreements in the field have been historically severe or if the equal consideration of expert opinions is threatened by dominant or persuasive personalities.[127]

In the medical literature, the Delphi technique is most often used for educational curriculum development, research priority development, policy development, analysis of professional competencies, clinical definition creation, and clinical guidelines development when the topics are not fully supported by evidence.[120, 132-135] There are currently only four Delphi studies in the field of pediatric complex care, using divergent methodologies.[136-139] Three of these studies identified consensus outcomes to inform CMC research and quality initiatives, and the fourth identified initiatives to transition CMC to adult care. There have been

several studies that utilized Delphi surveys to reach consensus on pediatric disease definitions.[140-146]

5.6.3. STRENGTHS AND LIMITATIONS OF THE DELPHI TECHNIQUE

The main strength of the Delphi technique is the ability to maximize objectivity in a fundamentally subjective exercise on subjective topics. Specifically, the use of controlled feedback to respondents between survey rounds allows the systematic sharing of group opinions and maximizes objectivity through the individual consideration of the collective interpretation of reality.[120] An additional benefit is the mandate of anonymity which minimizes group pressure for conformity and the threatening influence of highly vocal and influential individuals.[120-122, 126, 128, 147-150] This can be accomplished because a Delphi is almost always a survey-based technique. A third benefit involves the composition of the respondent group. Group composition is optimized if heterogenous opinions are represented among individuals with similar levels of experience. In a Delphi this can be more easily achieved as the survey-based technique allows easier recruitment across wide geographic distribution.[120, 128, 151, 152] Last, the technique is not resource intensive if performing the consensus using electronic survey platforms.[125, 128]

Certain limitations must be acknowledged when considering the Delphi. First, although not a direct critique of this methodology, topics plagued with subjectivity and paucity of evidence will always have risk of personal bias among respondents no matter the rigor of the methodology.[125] This is just a feature of the subjective space the Delphi technique is designed to work within. Second, as with most survey-based methodologies, response bias is a concern. During a Delphi study attrition bias represents an even more significant limitation as

there is a high time and cognitive commitment being asked of the respondents as it is a multi-iteration survey technique asking respondents to carefully consider their opinions on subjective topics.[120, 125, 153] Third, the benefits of anonymity must be balanced against its limitations. When protected by anonymity, all opinions are weighted equally and researchers must ensure each expert participant has similar experience to justify equal weighting.[120, 154, 155] Also, despite protection by anonymity, there is the theoretical possibility of social acceptability bias in which respondents accept consensus rather than share divergent opinion (Section 9.2.3).[120, 122, 149, 156-158] Last, the Delphi technique is a consensus building technique and should be interpreted as such. The results are a collection of expert opinions and should not be interpreted as being more than that.[159, 160]

5.6.4. SURVEY QUESTIONS AND TECHNIQUES

It is important for the researchers to develop a diverse, informed committee to create the survey questions and that the survey be tested for face and content validity during pilot testing. The questions being asked must be designed and tested to ensure they represent the goal of the Delphi, and that a reasonable amount of knowledge is needed to respond fully but with enough room for individual opinions within this expertise.[120, 155] Considering the Delphi technique exists to address subjective, sometimes ambiguous, questions, careful and meticulous instructions must be provided for the survey to prepare respondents for these questions. Questions that are too pointed and specific may lead the respondents to a premature consensus and closure of the study. Questions that are too open, however, may confuse the respondents to the topics that are relevant to the study.[127] Thus it is important to strike a balance, providing structure to the respondents reponses while leaving enough room

for expression of the respondents focused opinions.[127] The goal of the Delphi, the instrument design, and the careful selection of experts all interact, and require careful design in order to answer the subjective research question.[155]

As with any survey-based methodology, response rates and attrition bias must be considered in study design. There are several strategies with evidence to improve response rates.[161] Completion reminders have been shown to increase response rates.[162] These reminders are usually reserved for participants who have not yet completed a survey despite receiving an invitation. Moreover, a statement of scarcity and a statement demonstrating participant contribution both show improvements in response rate.[131] A statement of scarcity reminds respondents of their specific, and rare, skillset that is crucially needed to complete the survey project. Furthermore, a statement demonstrating the importance of the participant contribution is meant to engage participants by showing them the impact of their time. Last, the use of financial incentives to increase survey response rates in the general public is inconsistently supported by evidence.[162] The literature is mixed when examining the use of financial incentives to increase survey response rates in healthcare and medical research professionals.[163-168]

5.6.5. EXPERT RESPONDENTS

Respondents are the group of individuals that are being asked to complete the iterations of the surveys to reach consensus on a subjective topic. The term “expert” does not fully encapsulate the qualities of this group, but this term is ubiquitous in the literature on Delphi and will be used here.[169] There is currently no consensus of what it means to be an

expert in a Delphi study, as there are numerous definitions in the literature.[127] In general, the respondent population should be a group of people who have disciplinary expertise to complement the research question, and are willing to engage in a discussion around the question. They should have the ability to helpfully contribute, and, considering consensus is the ultimate goal, they should be able to adjust their inputs based on group feedback.[120, 149, 160] The composition of the expert group is the most important consideration of a Delphi study because their opinions create the data and their group conclusions direct the context and generalizability of the results.[120, 127, 170, 171]

There are some guidelines that can help direct researchers in selecting experts. First, a respondent group should include both “internal groups” and “external groups” to maximize valid, acceptable results and to maximize credibility with the target audience.[172] External groups are mainly composed of people studying the topic who demonstrate academic and/or clinical expertise in the area. These individuals may produce valid consensus due to their experience, skills and resources, but their consensus may not be acceptable by internal groups.[126] Internal groups which are the “front-line” people utilizing the definition for clinical and policy purposes.[126] These people may produce consensus that is more likely to be accepted by the community of front-line stakeholders, but the results may be less scientifically valid due to a relative lack of expertise in the scientific method. Second, and somewhat related, a heterogeneous group of respondents with a variety of perspectives and backgrounds may produce higher quality results, closer to the shared reality of the field. When composing an expert group researchers should strive for a varied composition.[121, 122, 124] Third, these heterogenous perspectives must be contained within a group that is perceived to have a similar

level of expertise relative to the research question.[120, 155] During the Delphi process, respondents must consider others opinions but weighting individual opinions based on expertise cannot be done because they are anonymous. A standardization of expertise in the group can counter this by negating the need for weighting. Last, and perhaps the most challenging, researchers must recruit experts who are invested in the lengthy process. They must have opinions and knowledge on the topic, but remain impartial to the final findings and are not obstinate to their way of seeing the world.[150]

As demonstrated, the concept of "expert" is poorly defined. In this setting, it is quite difficult to identify the greater population of experts with these qualities from which to randomly sample a study population. Instead, purposive sampling is most often employed to recruit experts, often with snowball recruitment from this originally invited group.[120, 125, 127] For example, invitations for research participation are often sent to the rosters of pertinent professional organizations, authors of pertinent papers after review of the literature, and positional leaders identified through networking among the peers of the researchers.[120, 173-175] It is important that expert respondents are not simply identified and chosen based on their acquaintance to the research team.[122] Identified individuals are then screened for study specific enrollment criteria before enrollment as expert respondents. It is acknowledged that this common recruitment strategy is susceptible to selection bias.

5.6.6. NUMBER OF EXPERT RESPONDENTS

In classic quantitative research, an adequately sized, random sample is fundamental to the power of a study. However, in Delphi research representative samples are less contingent on sample size as more importance is placed on adequately representing the qualities, and

varied opinions, required of an expert on the topic.[121] The goal of a Delphi is to move towards group consensus with a large enough number of respondents that all points of view are represented.[120, 127, 176] There is currently little guidance on what is an adequately-sized respondent group needed for reliable or valid consensus in a Delphi technique.[122]

The number of respondents in the Delphi literature is wide and can be as low as 10 to more than 1000.[125, 177] Published guidance on the sample size is not always practically helpful. For example, it has been suggested that an adequate respondent group size is a sample of 10% of the greater population. This is theorized to be an adequate size to represent all opinions around the research question.[127] Of course, this depends on whether we know the size of the greater population. Other researchers suggest that more is better, ensuring that there is complete representation of opinions and more robust and dependable consensus.[122] This advice, however, downplays the practical difficulties of running a study with a large sample.[176] Additionally, other authors suggest that a minimally sufficient number of respondents should be chosen, and the stability of their consensus should be verified in follow-up studies. Again, this leaves Delphi researchers on the hook for future studies and a "sufficient" number depends on the yet-unknown results of the future studies.

Practically, several authors recommend hard targets for recruitment numbers. Generally, 10-20 respondents are recommended.[120, 147, 178] However, one of these authors provide the proviso that 10-15 respondents are adequate if their background is homogenous but more would be needed if their backgrounds were heterogenous, to a general maximum of 50.[120] Furthermore, two studies presented some evidence to guide the sample size requirements. One study showed that a respondent group size of 23 participants was adequate

to provide stable consensus using bootstrap analysis.[177] The other study demonstrated that a consensus arrived at with a respondent group of 50 was consistent with a group of 150 who completed the same survey.[127] With this paucity of evidence in the literature it is apparent why many Delphi researchers settle on a sample size that is based on convenience rather than methodologic rigour.

5.6.7. NUMBER OF DELPHI SURVEY ITERATIONS

The Delphi technique depends on the delivery of multiple survey iterations with the goal to approach consensus among the respondent group. Section 5.6.11 describes the definition of consensus, but in short it refers to a threshold of agreement among the participants regarding an answer to the question being asked. Again, there is little guidance from the literature on the adequate number of iterations to achieve consensus. The achievement of consensus is dependent on the research question, the qualities and numbers of the respondents, and the contruction of the surveys. Therefore the number of iterations is important for practical planning but may not be the most critical factor in achieving consensus.

Previous studies suggest, or have used, between 2-5 iterations to achieve consensus, however most report between 2-3. [120, 148, 175, 179] There is no standard however, and many authors suggest continuing the iterations until the pre-defined definition of consensus is reached on all items or there is no further movement towards consensus on the remaining contentious issues.[120, 147, 157, 180, 181] This approach somewhat resembles saturation, common in qualitative research methodology. In a review of Delphi methodology in the literature, it was found that 23% of Delphi studies were ended when an a priori definition of

consensus was reached on all questions or items and 2% ended when there was no further movement towards consensus.[120, 127, 175, 182]

An open-ended number of iterations is problematic for three reasons, though. First, it is unclear whether more iterations improve the accuracy of the group's decision making or whether consensus is reached based on capitulation after an exhaustive number of iterations.[122] Second, we know that progress towards consensus slows as respondents progress through the iterations.[120, 183] If this is interpreted as lack of consensus movement, researchers may decide to halt the study before consensus is reached on some items or questions. [125, 149] Third, more iterations results in greater time, cost, participant fatigue, apathy, and attrition.[130, 179, 184] The lack of guidance on iteration number has resulted in a research environment where this decision is usually based on pragmatic considerations.[179] In fact 71% of Delphi studies were ended when the pre-determined number of iterations was reached, independent of reaching consensus or movement towards consensus.[182]

5.6.8. INFORMING THE FIRST ITERATION

Populating the first iteration survey is a vitally important consideration because the topics introduced in this survey will act as trailheads for all discussion and deliberation moving forward through the iterations.[185] The researchers must ensure that the topics introduced in the first iteration represent all necessary components required to eventually build a comprehensive answer to the question, although those components may not yet be refined through ongoing deliberation. As an analogy, if you perfectly shape 3 legs of a table through multiple iterations of work it will not answer the question of how to keep the table upright. The

fourth leg has to be considered at the start. Construction of the survey must be done carefully though as the construction of the first survey has great potential to introduce the personal bias of the researchers.[127]

There are two predominant strategies in the literature on how to populate the first iteration survey: open-ended questions or pre-populated questions. Open-ended questions in the first iteration are meant to collect respondent opinion which will inform the content of the future iterations.[120, 121, 127, 129, 181] Voting on the respondent-identified topics will then continue in future iterations to discern the degree of agreement or disagreement with each topic.[129] This method has the benefit of tapping into the collective experience of the respondent group to identify a breadth of topics to consider when answering the research question, including topics that may not have been considered by the research group. It can limit the degree of researcher bias that would come with a researcher-designed first iteration survey. This open-ended method has limitations however. First, the open-ended questions are designed by the research team and are still subject to bias. Second, open-ended questions may lead to runaway answers that have little bearing to the original research question. Last, the ability of the respondent group to suggest the broadest array of topics depends on the familiarity of each respondent with the literature. It may be difficult to expect the respondent group is up-to-date with the broad literature required to answer a subjective research question. To circumvent this limitation some researchers have distributed literature synthesis documents to the respondent group prior to the first iteration.[175] This is done to standardize the knowledge base of the respondent group but in doing so may add researcher bias.[175]

The second method to inform the first iteration survey is to have a pre-populated survey designed by the research team. The topics to be voted on by the respondents are constructed from the start. These original topics can be informed in a variety of ways including a systematic or scoping literature review, synthesis of existing guidelines, expansion of a previous conceptual framework, a summary of current clinical practice, or expert opinion which can be elicited in a systematic way such as the nominal group technigue or through focus groups.[130, 175, 186] This method is reserved for research questions that have a body of literature available.[120] Considering this method is more dependent on input by the research team to inform the first-iteration, there is a greater risk for introduction of researcher bias.[187] Also this method misses out on the shared experience of the respondents.[127]

A hybrid solution is becoming more popular in the literature to incorporate an open-ended philosophy into a pre-populated survey.[31, 169, 188] Generally, these first iteration surveys offer pre-populated topics for consideration by the respondent group while also providing space for open ended comments. The future iterations will then move forward with a combination of pre-populated and respondent-suggested topics.

5.6.9. STRUCTURE OF FURTHER ITERATIONS

Whether topics are generated with open-ended questions or through pre-population, the point of the further iterations is to refine the topics or to reach consensus for inclusion or exclusion of the topics in the collective answer to the research question. Researchers must decide on how the topics are presented in the iterations for voting. Most often topics are presented with a Likert scale to measure the level of agreement and the most common scales

are 3, 5, 7, or 9 point.[120, 130, 175] The 9 point scale is the scaled preferred to reach consensus by the RAND/UCLA appropriateness method.[129, 175] Studies have used 6, 7, 10, and 11 point scales as well, but are less common.[175] Other less popular methods of respondent opining is ranking of choices or selection of preferred answers from a pre-populated list.[175]

Through the iterations the presentation of topics can evolve through the inclusion of newly generated topics, the modification of topics that have not yet reached consensus, the removal of topics with high inclusion consensus requiring no further deliberation, or the removal of topics with high exclusion consensus.[130, 175] When starting with open-ended questions in iteration 1, researchers have additional considerations when moving forward. To inform future questions, content analysis using qualitative data analysis techniques are used to analyze the early qualitative findings.[120, 121, 149] As iterations progress, debate continues, agreement and disagreement become apparent, and consensus starts to develop.[120] In some studies, researchers have chosen to use the final iteration as a referendum on the consensus that has occurred so far, asking respondents whether they agree with the consensus as it evolved throughout the study.

5.6.10. STRUCTURED FEEDBACK TO RESPONDENTS

There are two types of feedback that should be planned for in a Delphi study. First, is the type and amount of information distributed to the respondents prior to starting the first iteration and second is the type and amount of information fed back to respondents as the iterations progress. Regarding the former, respondents, at the minimum, must understand the

question being asked and understand the research goal of the Delphi. This should be covered in the consenting process but further clarification is sometimes needed.[155] Some research questions require a review of foundational concepts or a more detailed review of the literature to ensure the repsondents, who may have varying degrees of expertise, achieve a minimal understanding of the topic.[128, 189-193] These summaries are sometimes necessary but care in their construction is prudent considering they have a high risk to introduce bias into opinions of the respondents.[128, 189-193]

Regarding inter-iteration feedback, it is important in Delphi methodology that the respondents reconsider their previously held opinions as iterations progress. Therefore, a strategy of controlled information feedback is necessary as iterations progress to ensure all respondents are privy to the same, standardized information.[194-196] This controlled feedback allows respondents exposure to the maximum number of respondent opinions, to reflect and refine their held opinions, and move towards consensus as the iterations progress.[120, 125, 150, 160] Studies employ a variety of techniques in this regard including presentation of the respondents own previous responses, statistical summaries of group responses, and/or thematic summaries of respondent comments. Structured feedback is important but careful consideration of the methodology is vital as responses are shaped by the provision of information, and distortions in the process of feedback can change these responses thus introducing a researcher's personal bias.[120, 127, 157, 158]

Using data from the preceding iteration, a common and basic level of feedback is to present: a) the respondent's own previous repsonses and b) the group central tendency with a measure of dispersion, such as mean, median, mode standard deviation, or interquartile

range.[122, 125, 130, 175, 197-202] Individuals own opinions are often forgotten in the time between iterations and respondents often have to be reminded of their previous beliefs as well as the summarized group opinion.[120] This degree of feedback is not administratively cumbersome and is relatively free from researcher bias. However, it only provides the respondent with a sense of their position on a topic relative to the group and does not provide rationale for the answers among the group. Therefore, there is a risk that any movement towards consensus may be due to normalative social influence and not the persuasiveness of the shared information (Section 9.2.3).[120, 122, 149, 156-158]

As an addition, some studies also feed back group comments.[130, 175] The content of the comments are often study specific but usually contain the repsondent rationale for their answers.[120, 122, 130] Feeding back the reasons for responses improves accuracy of the group decision.[203] Whatever the content, these comments can be presented as a list of all available comments, as a researcher-selected sample of important or persuasive comments, or as a thematic summary generated using standardized qualitative research techniques. There is a difficult balance that must be struck when trying to ease the burden for the respondents while avoiding researcher bias. Providing a verbatim list of all the group comments is cumbersome for the respondents, thus risking attrition, but would eliminate researcher bias dervied from manipulation of the comments into themes. However, even if done systematically, any steps by the reseachers to analyze, simplify and present the meaning of statements risks bias from the researchers.[127] There is little guidance to help construct a method of standardized feedback.

5.6.11. CONSENSUS

The primary outcome of a Delphi study is the consensus of opinions from the respondent population. As with any research, a well-defined, *a priori* determined definition of the primary outcome is critical but is not always done in published Delphi research.[127, 182] There is no consensus among Delphi researchers regarding the definition of consensus.[120, 149, 186, 204] What is agreed upon, however, is that whatever definition of consensus is used, it must be defined *a priori*.[131] There have been numerous definitions of consensus used.

The most common way to define consensus is to set a proportion threshold on a Likert scale which is taken as a groups consensus to include or exclude the item.[120, 127] For example, some researchers defined consensus when 80% of respondents chose "agreement" selections or "disagreement" selections.[120] Expanding on this, agreement consensus may be considered if respondents "strongly agreed" or "agreed" with a statement or disagreement consensus may be considered if they "strongly disagreed" or "disagreed." The exact proportional threshold is highly variable ranging between 51%-100% of respondents in the literature, with 70-80% being the most common.[120, 127, 143, 144, 182, 205, 206] Similarily, the UCLA RAND method includes a definition of diasagreement when at least 1/3 of respondents rate an item or question at the opposite end of the scale to their peers.[129, 182] In a review of Delphi studies, it was noted that 57% of authors used a proportion threshold to define consensus.[182]

Often measures of central tendency are used by authors in their consensus definitions with roughly 21% of authors using this method. [182] In the UCLA RAND method, for example, inclusion consensus is reached on a 9-point likert scale when the median answer is greater than

or equal to 7 and exclusion consensus is reached when the median is less than or equal to 3.[129, 182] Although the UCLA RAND method is often discussed as a rigourous methodology in the field, only 5% of studies used this consensus definition.[182] Some authors may use a central tendency cutoff as the primary definition for consensus or they may use it in combination with a proportional threshold definition.[120, 175] For example, a definition for agreement consensus may require 70% of subjects to rate 3 or higher on 4 point likert and require a median score greater than 3.25.[129]

Other than definitions based on proprotion threshold and central tendency, other quantitative methods used to define consensus include statistical measures of dispersion, inter-rater or intra-class correlation. Such measures included decreasing interquartile range, Kappa statistic, Crohnbach's alpha, intraclass correlation coefficient, and Kendal's W.[182] These are rarely used and a review of Delphi methodology stated that only 14% of studies used at least one of these methods.

Another method to define consensus is worth noting. Two percent of authors used a strategy of ranking or rating items to establish priorities from a list of questions, items, or choices.[120, 182] The usefullnes of this strategy is limited to certain research questions in which the authors are trying to reach a consensus on priority setting, such as in curriculum development or research prioritization.

What has been discussed so far is the definition for consensus, however the definition for non-consensus is equally important. The definition for non-consensus is often unstated but,

practically, items that are considerered “non-consensus” are those that are left without consensus at the end of the pre-set number of iterations.

5.6.12. ANALYSIS AND REPORTING

Analysis techniques vary depending on the method of data collection and study design, however there is little guidance on how to balance quantitative or qualitative analysis techniques and little guidance on how to present the data generated.[127] The statistical analysis employed in Delphi study reporting is often descriptive and simple.[127, 149] Often for each item under study, a measure of central tendency of the Likert answers is reported to illustrate average opinion and a measure of dispersion to illustrate the divergence of opinion. This is often median and interquartile range due to the non-normal nature of Likert data.[184] This method of reporting does not illustrate the distribution of Likert answers well. For example, a bimodal distribution can be obscured if only the median and interquartile range are reported.[121] Another method which can remedy this is to present the same data using frequency counts for each Likert answer and percentages.[126, 159, 207] Such reporting can be done for each item in each iteration, but at minimum should be provided for items in the final consensus. Additionally, some authors have reported different degrees of consensus such as low, moderate, or high consensus.[175] This was accomplished using the degree of tightness of the dispersion around the measure of central tendency or the absolute proportion of respondents selecting certain Likert answers.

When studies use qualitative methodology the reporting standards are more rigorous and mirror the well-described standards from the qualitative literature. Content analysis is performed using qualitative data analysis techniques and this strategy is often used to create

themes to feed back to the respondents during subsequent iterations, or to inform future survey items if using open-ended questions in iteration 1.[121, 125, 149] A variation of qualitative techniques have also been employed such as analyzing and reporting frequency counts of particular words, phrases, or themes.[125, 149]

5.6.13. BASIC QUALITY STANDARDS OF A DELPHI STUDY

As can be appreciated from the former review, the main guidance researchers have when planning a Delphi study is from the popularity of certain techniques in the literature. However, an increasing number of authors have put forward minimal standards in planning and reporting Delphi studies, listed here:

1. A well-defined purpose to the study and subsequently a well-defined research question.[175]
2. Enrollment criteria of the respondents must be detailed, made a priori, and reported in a way that is reproducible.[128, 175, 182, 189-193] Considerations for enrollment criteria may include membership in an organization, an objective recognized authority, relevant clinical expertise, a field of work, a geographic scope, or membership of a certain profession or stakeholder group.[175] In a review, 65% of Delphi studies achieved this standard.[182]
3. A decision must be made regarding the amount and content of information provided to the respondents prior to the first iteration.[128, 189-193] This information is meant to provide the respondents with the minimal background knowledge required to answer the questions.

4. The conditions determining the number of iterations should be made *a priori* and be clearly reported, including conditions for stopping the study early.[182] It was found that 81% of Delphi studies properly defined the number of iterations but only 15% defined the conditions for early stopping.[182]
5. The definition for consensus on item inclusion, item exclusion, and non-consensus must be made *a priori* and clearly reported.[128, 175, 182, 189-193] It was found that 59% of Delphi studies defined criteria for excluding items from the final consensus.[182]
6. The data collection instrument, which is almost always a survey, must be pilot tested.[175]
7. A description about the amount of formal feedback provided to the respondents before subsequent iterations.[128, 189-193]
8. When conducting the study, the following standards should be followed:
    a. An audit trail of notes should be kept to clearly demonstrate unexpected problems and the rationale for decisions.[208]
    b. More than a single researcher should be used to analyze data and populate future surveys.[208]
9. When reporting the study, the following standards should be followed:
    a. A clear description of methods must be reported including responses to, at minimum, the standards described above.[175]
    b. A description of the levels of anonymity enforced.[128, 189-193]
    c. A flow chart of respondent participation through the iterations including at minimum the response rates.[128, 175, 189-193]

d. The data analysis must be clearly justified and reported.[175]

e. An inclusive and transparent report of the results including the distribution of the respondent answers to the items, a sample of respondent comments, and a demonstration of the evolving consensus over the iterations.[128, 175, 189-193]

f. Conclusions must be reported in the context of the constraints put on the consensus by the composition of the respondent group and other decisions made in the methodology.[175]

g. Limitations must be acknowledged.[175]

6. RESEARCH QUESTIONS, OBJECTIVES, AND AIMS

Considering the theoretical grounding of the Cohen 2011 conceptual definition, and its predominance in the literature, it was used as a base to guide for the further development of a clinical definition. To operationalize this conceptual definition, it is necessary to identify specific, clinical, operational items under each of the four conceptual domains. Therefore, the specific questions for this study were:

1. **What is known from the existing literature about potential variables used to define or describe children with medical complexity, and how will they inform the four domains on the initial Delphi survey?**
2. **What items within each domain of this conceptual definition do experts in the field collectively believe, through iterations of consensus building, should be included for that domain to be fulfilled?**

These questions inform the overall objective of this research which is to develop a consensus among the CMC professional community in Canada and the U.S.A. regarding a standardized clinical definition of CMC using a Delphi method. Specifically, the study sought to identify operational criteria (items) within each domain required to fulfill that domain. We achieved this objective via two Specific Aims:

- **Aim 1: Developed a Draft Delphi Survey to Operationalize the Cohen 2011 Conceptual Definition.**
    - Aim 1.1: Identified members of the Delphi survey development committee.

- Aim 1.2: Identified potential items from the literature potentially pertinent to the four domains: family needs, healthcare usage, functional limitations, and diagnostic conditions variables.
- Aim 1.3 Constructed the first iteration of the Delphi survey
- Aim 1.4: Performed face validation and pilot testing of the draft Delphi survey and adjusted the survey appropriately.

- **Aim 2: Performed a Delphi Survey Process**
  - Aim 2.1: Recruited participants to the Delphi process.
  - Aim 2.2: Engaged the Delphi participants towards consensus on the item list from Aim #1.

7. METHODS

7.1. AIM 1: DEVELOPED A DRAFT DELPHI SURVEY TO OPERATIONALIZE THE COHEN 2011 CONCEPTUAL DEFINITION.

7.1.1. AIM 1.1: IDENTIFIED MEMBERS OF THE DELPHI SURVEY DEVELOPMENT COMMITTEE.

In addition to the MSc thesis student and the MSc supervisor, this committee required at minimum two complex care clinicians with ≥5 years experience in the field, two researchers with ≥5 publications on child health research, and one expert in Delphi methods with at least one prior Delphi study publication. To minimize bias, committee members were not respondents in the Delphi survey.

The committee had four functions. First, it reviewed the literature for variables to inform potential Delphi survey items (Aim 1.2 – Section 7.1.2). Second, it made final decisions regarding the final composition of the first iteration survey (Aims 1.3 – Section 7.1.3; Aim 1.4 – Section 7.1.4). Third, after the first iteration survey, a sub-committee composed of the MSc thesis student and the two complex care clinicians reviewed the respondents' suggested changes to existing items and suggestions for novel items (Aim 2.2 – Section 7.3). Last, a sub-committee composed of the MSc thesis student and the MSc supervisor reviewed each iteration of the Delphi survey before release to the respondents (Aim 2.2 – Section 8.2.2).

7.1.2. AIM 1.2: IDENTIFIED POTENTIAL ITEMS FROM THE LITERATURE POTENTIALLY PERTINENT TO THE FOUR DOMAINS: FAMILY NEEDS, HEALTHCARE USAGE, FUNCTIONAL LIMITATIONS, AND DIAGNOSTIC CONDITIONS VARIABLES.

The initial survey presented items with the potential to define the respective domain in the conceptual definition by Cohen *et al.*, 2011. [36] The items on this initial survey were drawn from variables within pertinent articles studying CMC, identified using the scoping literature review methodology adapted from Arksey & O'Malley 2005.[209] This methodology allows the mapping of key concepts of a research field when the field is broad, complex, imprecisely defined, and has not yet been comprehensively reviewed.[209] The goal of this review was to identify a broad array of biopsychosocial variables in the literature that are associated with CMC.

First, relevant articles were identified using search criteria purposefully designed in conjunction with a librarian to be a broad review of the CMC literature (Appendix 2). To identify pertinent articles, the Medline, Embase, and Scopus databases were searched for English language articles from January 1998 until September 2019. In the U.S.A., the Maternal and Child Health Bureau (https://mchb.hrsa.gov/) first presented their definition of Children with Special Healthcare Needs and prioritized these children for ongoing research in 1998.[78]

Second, articles identified with this search strategy had their abstracts screened by a single reviewer (KM) to determine the relevance for full text review and data extraction. The full text was reviewed if the relevance was unclear following abstract review. A single reviewer strategy was chosen over a double reviewer strategy for practical reasons, specifically the lack of funds to pay a clinical expert for the substantial time commitment and the paucity of local

experts with necessary clinical experience. Articles were chosen for full review and data extraction if they fulfilled both of the following criteria: (1) publication in a peer-reviewed journal, and (2) assessed the definition of CMC or the epidemiology, etiology, natural history, or management of CMC. This included CMC populations identified using a variety of research definitions. Articles were excluded based on the following criteria: a) scope limited to children with specific chronic conditions (ex. Chronic kidney disease, respiratory failure) b) case studies or case series with less than 10 children, c) studied the education of medical learners regarding CMCs, or d) evaluated the impact of CMCs in adulthood, including transition planning. Inclusion and exclusion criteria were developed post-hoc as familiarity was gained with the literature, an accepted method in Scoping literature review methodology.[209]

Third, a single reviewer (KM) reviewed the methods and results sections of the articles that passed the second screening step, and extracted variables having the potential to fulfill any of the four domains of the conceptual framework by Cohen 2011.[36] Within these articles, chosen variables could be: dependent variable/covariates, dependent variables/results, qualitative themes that provided insight into CMC, or social determinants of health using the Artiga and Henton 2018 framework as a guide.(Figure 2)[119] For example, a study examining hospitalizations in CMC demonstrated a mean of 3 hospitalizations per year, which informed an item under the “healthcare use” domain. Some items were reworded to make them more relevant to the Delphi research question. For example, the CMC hospitalizations variable was converted to an item worded as “Over the past 12 months, the child had a substantial number of unplanned, inpatient hospitalizations, independent of the length of the hospitalization.”

Finally, from this draft list of items, the Delphi survey development committee chose a final list of items for presentation to the Delphi respondents in the first iteration survey. Items were chosen by the committee if they were maximally objective and had conceptual and practical consistency with the related Cohen 2011 domains. An iterative process of discussion and modification was used to select items and improve wording until the committee was content with the potential item list to be presented to the Delphi respondents. Modifications could include: a) adjustment of the syntax of an item, b) collapse of multiple items into a single item, c) omission of items, or d) addition of new items deemed pertinent (Appendix 2; Appendix 3). It was important to word the items in a way that satisfied the dynamic nature of a CMC definition and make clear the respondents would have to consider items as important descriptors at any age, level of development or family situation.

7.1.3. AIM 1.3: CONSTRUCTED THE FIRST ITERATION OF THE DELPHI SURVEY

Items were organized into the four survey sections corresponding to the domains of the clinical conceptual definition. To be considered a child with medical complexity under this definition, a child must possess at least one item in each of these four domains. Items related to the social determinants of health were embedded within the family needs domain. For presentation to the Delphi respondents, the order of the sections (domains) was randomized, to minimize bias from partially completed surveys

All items asked respondents to indicate their agreement with its inclusion as being sufficient, if present in a patient, to qualify that domain as being fulfilled. Their degree of agreement was rated on a 5-point bipolar Likert scale with anchors: strongly agree, agree,

neutral, disagree, and strongly disagree. These questions were referred to as Level 1, because some, but not all, items also had a Level 2 to consider. Continuing the example of hospitalizations per year, in the Level 1 item, respondents were asked to indicate their agreement with the importance of including the number of hospitalizations per year as an item in the final definition. Subsequently, in the Level 2 question, they were prompted to choose the threshold number of hospitalizations they deemed necessary to fulfill the item. For this first survey iteration only, respondents had the opportunity to suggest a threshold (level 2 item) with a free-text answer. In subsequent iterations level 2 item scales were centred on the median response from the free text answer and the extremes of the scales were the interquartile range. For example, when determining the threshold number of hospitalizations per year, the median number from the first iteration responses may be 3 hospitalizations per year with an interquartile range of 1-5. Therefore, the subsequent integer scale in future iterations would read as 1 – 2 – 3 – 4 – 5. In iterations 2-4 respondents selected a threshold number (level 2 item) of hospitalizations from this interval scale.

Additionally, voluntary comment sections were available to the respondents for every item (level 1 and 2) which is meant to record the rationale for each respondents' choices and, in iteration 1, to suggest changes to the syntax of the items. A comment section was also presented at the end of each domain section in the first iteration survey allowing respondents to suggest items that should be considered in subsequent iterations.

7.1.4. AIM 1.4: PERFORMED FACE VALIDATION AND PILOTING OF THE DRAFT DELPHI SURVEY AND ADJUST THE SURVEY APPROPRIATELY.

Three survey pilot testers tested the initial survey and critiqued it for face validity, readability, and ease of use. One had experience in survey-based research methodology, and two in Complex Care Pediatrics. These three specialists were not members of the MSc thesis committee, members of the Delphi survey development committee, or Delphi survey respondents. The pilot testers were instructed to critique the understandability of the survey language, ease in progressing through the survey mechanics, and clarity of survey instructions. The survey pilots communicated critiques to the MSc student for correction. Modifications were made to the syntax of the items to improve clarity and to the instructions to remedy confusion regarding the purpose and mechanics of the survey. A single round of survey piloting was performed.

7.2. AIM 2: PERFORMED A DELPHI SURVEY PROCESS

7.2.1. AIM 2.1: RECRUITED PARTICIPANTS TO THE DELPHI PROCESS

7.2.1.1. DELPHI RESPONDENT GROUP COMPOSITION

Eligibility criteria were either of: (a) ≥2 years of clinical experience managing CMCs, and/or (b) experience studying these children for research purposes, as evidenced by having been first or senior author on at least one peer-reviewed article in the past 3 years on the topic of CMCs. There were no exclusion criteria. CMC caregivers were not recruited for this study with the rationale provided in section 11.6.

7.2.1.2. RESPONDENT GROUP SIZE

Our goal was to obtain a minimum of 23 participants for each iteration with rationale described in section 5.6.6. To guarantee this sample size, potential attrition was considered. In the literature of Delphi studies for pediatric disease definition generation, when Delphi surveys were sent to an unsolicited population of experts, the average response rates for the first, second, third and fourth iterations were 58%, 49%, 18%, and 17%, respectively.[140-146] In the literature, when Delphi surveys were sent to previously consented experts, the average response rates were 96%, 69%, 51%, and 100%, respectively.[141, 142, 144, 145] In the current study, there was an initial unsolicited invitation for consent and then multiple ongoing Delphi iterations among these consented respondents. Considering the published response rates and our minimum goal of 23 respondents for each iteration, it was estimated that invitations had to initially be sent to a minimum of 135 individuals (23 respondents / 17% response rate) and a minimum of 45 (23 / 51% response rate) consented respondents in the first iteration were necessary to ensure the study was completed with a minimum of 23 respondents in each iteration.

7.2.1.3. RECRUITMENT STRATEGIES

Criterion sampling was used to construct a sample of potential Delphi respondents, diverse in geography and expertise. The respondents were recruited in the following of ways:

1. Potential respondents meeting the clinical expert criteria were recruited by sending electronic invitations to:
    a. Members of the Complex Care Section of the Canadian Pediatric Society.

b. The Complex Care google-group, a listserv of multi-disciplinary, complex care clinicians from across Canada and U.S.A.
c. Members of the Complex Care Special Interest group of the Pediatric Academic Society.
d. Members of the Complex Care Committee of the American Academy of Cerebral Palsy and Developmental Medicine.

2. Potential respondents meeting the research expert criteria were recruited by sending electronic invitations to first-listed and senior authors of the publications identified in Aim 1.2.

Initial contact with potential survey participants was done through email or listserv posts and included an information package containing a brief introduction of the topic, the study goals, an introduction to the planned survey process, and the Research Ethics Board consent statement (Appendix 4). By replying with interest to the professional email of the MSc student (KM), the participants gave electronic consent to participate. An email contact list was created of interested individuals with the email linked to a sequentially assigned participant identification number. A master key connecting the individual's identity, email and identification number was kept in paper format and housed in a locked file cabinet in a locked office at the Health Sciences Centre in Winnipeg, Manitoba. The email of the respondent, the identification number, and the respondent's subsequent survey answers were housed on a secure REDCap server at the Centre for Healthcare Innovation of the University of Manitoba in

Winnipeg, Manitoba. The identities of the potential respondents remained anonymous to the other respondents.

On each survey iteration, demographic information was collected on the respondents through self-report, including profession, geographic location to the city level, number of peer-reviewed research publications in the field, and the number of years practicing clinically in the field. This information was used to check the respondent's eligibility criteria, to describe the respondent population for each iteration, and to identify any differential attrition based on demographic characteristics. For respondents who did not volunteer their demographic information, eligibility criteria were confirmed using publicly available sources such as institutional websites.

7.3. AIM 2.2: ENGAGE THE DELPHI PARTICIPANTS TOWARDS CONSENSUS ON THE ITEM LIST FROM AIM #1.

7.3.1. LOGISTICS OF THE DELPHI PROCESS

The Delphi surveys was delivered online using the REDCap survey program from the University of Manitoba (Iteration 1 and 2 survey presented in Appendix 5 and Appendix 6, respectively). Each iteration was open for responses for three weeks with additional time available for extension upon request. Analysis and construction of the subsequent iteration occurred between iterations and this inter-iteration period was targeted to be no longer than three weeks. A maximum of four iterations was pre-determined for this study to allow ample opportunity to debate the considerable number of, sometimes complex, items. In addition, we

wished to respect the time and effort of the respondents by limiting it to a reasonable number, with particular emphasis to avoid an open-ended number of iterations.

To address the risk of non-response bias, the following efforts were undertaken:

i. When there was two weeks, one week and three days remaining to finish the survey, completion reminders were sent to respondents if there were survey sections yet to be completed.
ii. A statement of scarcity was included in the introductory email and each survey.
iii. Each survey after the initial iteration included a statement reminding the respondents that each item under consideration is constructed using their previous answers.
iv. A $10 incentive was offered to respondents before initiation of each survey iteration. Although the evidence for the use of incentives is mixed, it was decided to offer these incentives. In addition to the possibility of benefit, it was concluded that the expectation for incentives that has developed over years of survey research among respondents should be satisfied.

7.3.2. DELPHI ITERATIONS AND CONSENSUS

Each iteration of Delphi surveys aimed to further refine the developing consensus on items until the threshold for consensus was reached. In the first iteration the items constructed following the literature review was presented to the respondents for consideration. When considering the level 1 items (section 7.1.3), the consensus threshold was 80% agreement or disagreement (80% of respondents strongly agree or agree that item should be included in the definition or 80% disagree or strongly disagree, respectively). A proportional consensus of 80% was chosen as this is the most common method of consensus definition and the 80% threshold

is among the most common thresholds in the literature (section 5.6.11). When considering level 2 items, the consensus threshold was 70% for a particular integer on the Likert scale (70% of respondents chose the same integer). The lower consensus threshold for level 2 items was necessary due to the system of collapsing Likert answers into groups. For Level 1 items the Likert answers were collapsed into three possible outcomes (agreement, neutral and disagreement) while level 2 answers had five possible outcomes corresponding to the answers on the Likert scale. With fewer outcomes to choose from in the Level 1 items, a more conservative threshold value is justified relative to the level 2 items.

Items that reached consensus were omitted from consideration in subsequent survey iterations. Items reaching consensus for inclusion (agreement) were used in the final definition whereas items reaching consensus for exclusion (disagreement) were removed from future consideration, and not included in the final definition. Items that did not reach any consensus were presented to respondents in the subsequent survey iteration. To aid in re-evaluation of each item by the respondents, the items without consensus were presented alongside information from the preceding iteration including:

i. A histogram illustrating the distribution of answers on the level 1 Likert scale and the level 2 integer scale from the entire respondent population.
ii. The individual respondent's previous answer to the respective item.
iii. Comment themes as described in section 7.3.3.

Level 1 items that failed to reach consensus by the end of the fourth iteration were removed from the draft definition, along with corresponding level 2 item. For items where, after the final Delphi iteration, no level 2 integer had met the 70% threshold, but the

corresponding level 1 item had reached inclusion consensus, a set of rules decided the final level 2 integer selected for the clinical definition. These two rules, applied sequentially, were generated *a priori* by the authors:

i. The mode was selected as the consensus minimum if that value received >50% of the total answers.

ii. If the mode received <50% of total answers and multiple integers were within 5% of the total population of each other, the lower integer was selected as the consensus minimum. For example, if there was a total population of 45 respondents then 5% of the total population is 3 respondents. So, if selection "4" received 19 responses and selection "5" received 21 responses these two integers were within 3 votes of each other. By our rule the consensus selection is "4" as it is the lower of the two integers. This rule also applied to bimodal distributions. This lower integer is selected so that the final clinical definition has higher sensitivity due to the lower value.

The goal of this project was to present an standardized clinical definition for children with medical complexity. To reiterate, these items will contribute to resolving subjectivity of the Cohen *2011* conceptual definition. As such, the final definition followed the rules of this conceptual definition that one affirmative item within each of the four domains fulfilled that domain. The two levels for each item were amalgamated into one item under each variable in the final definition. For example, consider that the consensus determined that annual hospitalizations should be included in the final definition, and that level 2 consensus was reached that three hospitalizations is the minimum to achieve this item. Then the final item

presented in the definition would be “Greater than or equal to three hospitalizations in a year” under the “Health care Use” domain.

7.3.3. RESPONDENT COMMENTS

Respondents had the opportunity to provide comments with each item in each iteration of the survey. For every survey, respondents were encouraged to discuss their rationale for their answers to each level 1 and level 2 item. The first iteration survey had some unique comment features; in addition to rationalizing their answers, the respondents had the opportunity to suggest changes to item wording to make them more relevant for that domain. Additionally, the respondents could suggest new items for each domain that were not presented in the first iteration.

With each iteration respondent comments were reviewed in full three times. Upon first review respondent comments were reviewed by the MSc student and content codes were generated and assigned to the text corresponding to the content of the comments. Each comment could have multiple content codes. After the first comment review, the content code library was reviewed by the MSc student, code language was adjusted to better reflect comment meaning, and the codes were collapsed into overarching thematic categories. During the second comment review, the comments were reviewed again by the MSc student and thematic categories were assigned. During the final comment review, thematic categories were again reviewed and related categories were collapsed further into a second library of thematic categories. Comments were labelled with the finalized thematic categories by the MSc student.

Comment themes contributed to subsequent survey iterations in two ways, with differing methods for utilizing these themes:

i. The themes informed structured feedback presented to the respondents in the subsequent iteration, for the purpose of aiding in re-evaluation of their answers. Following each iteration, we identified the most common two themes arguing for the acceptance of the level 1 item into the definition, and the most common two themes arguing against the acceptance of the level 1 item into the definition. To move forward with a theme, we required at least two respondents to have had the same thematic sentiment. In a similar manner, the most common two themes rationalizing the respondent answers to each level 2 item were identified. These chosen themes (maximum of four for level 1 items and a maximum of two for level 2 items) were discussed by the MSc thesis student and the MSc supervisor for approval before they were presented in the subsequent survey.

ii. After only the first iteration, the themes informed how to change or add items for the second iteration. The wording of items was altered and new items were added if either of two conditions were met. First, a theme associated with changing or adding an item had support from ≥10% of respondent comments. Second, if the sub-committee composed of the MSc thesis student, two complex care physicians, and one child research expert agreed that a theme was pertinent to change or add an item, despite failure to meet the 10% threshold. The MSc student used the themes identified by these two methods to add items or alter the wording of the corresponding item. Added or altered items were presented to the sub-committee, iteratively deliberated, and further revised before presentation in the subsequent iteration. Revisions were meant to ensure clarity of syntax and to ensure the changes reflected the essence of the theme.

7.4. POST-ITERATION MODIFICATION OF ITEMS

After the iterations were commenced, several of the items that reached inclusion consensus were complementary to others and had the potential to be collapsed into single items. This collapse was important to simplify the final definitional tool and limit the effort needed to complete it. The final list of included items were sent to the Delphi Survey Development Committee for their opinions on which could be collapsed. The committee opinions were collated by the MSc student and through an iterative process of debate a final list of items were organized into a final definitional tool. This modification was not planned for *a priori*.

7.5. DATA ANALYSIS

Descriptive statistics were reported, separately for each of the four survey iterations, as count and percentages of: overall survey completion rate, the individual survey items, respondents' demographics, and the commenting rates per item. The proportion of respondents agreeing or disagreeing with level 1 items and the proportion selecting each potential cut-off integer value for the level 2 items is reported as the percentage selecting "strongly agree" and "agree," the percentage selecting "strongly disagree" and "disagree," and the percentage selecting each level 2 integer, respectively.

To examine for evidence of attrition bias several steps were taken:

i. The proportion of "returning respondents", "new respondents" and "withdrawn respondents" was reported as a count and percentage for each item over each iteration. "Returning respondents" referred to respondents who completed the current and previous iteration, "new respondents" referred to those who completed the current

iteration but not the previous iteration, and “withdrawn respondents” referred to those who did not complete the current iteration but completed the previous iteration. Together these three groups represented “all respondents.”

ii. The proportion of “all respondents” and “new respondents” agreeing and disagreeing with the item from the current iteration was reported as a count and percentage. A chi-square test was used to determine whether there was a significant difference between “new respondents” and “all respondents” regarding the proportion of agreement and disagreement with each item; significance was determined to be $p<0.05$. The significance of the chi-square tests were adjusted using Simes’ False Discovery Rate method to account for multiple comparisons. This method controls for the false discovery rate (number of falsely rejected null hypotheses) using a step-wise procedure which produces p-values adjusted for the number of comparisons tested.[210] This procedure was completed using Stata version 17.[211]

iii. The proportion of “withdrawn respondents” agreeing and disagreeing with the respective item in the previous iteration was reported as a count and percentage. A chi-square test was used to determine whether there was a significant difference between the agreement and disagreement of the “withdrawn respondents” in the previous iteration and the agreement and disagreement of “all respondents” in the previous iteration. The significance of the chi-square tests were adjusted using the Simes False Discovery Rate method to account for multiple comparisons.

8. RESULTS

8.1. AIM 1: DEVELOPED A DRAFT DELPHI SURVEY TO OPERATIONALIZE THE COHEN 2011 CONCEPTUAL DEFINITION.

8.1.1. AIM 1.1: IDENTIFIED MEMBERS OF THE DELPHI SURVEY DEVELOPMENT COMMITTEE.

This committee consisted of the MSc thesis student (KM), the MSc supervisor (AG), two complex care clinicians (GR and EC), three researchers with expertise in child health research (EC, GR and CR), and two experts in Delphi methods (EC and KS). The sub-committee tasked with reviewing the respondent comments in the first iteration, requesting changes to first iteration items, and suggesting new items for inclusion in future iterations included the MSc thesis student (KM) and two complex care clinicians (GR and EC). Last, a sub-committee composed of the MSc thesis student (KM) and the MSc supervisor (AG) reviewed each iteration survey before release to the respondents.

8.1.2. AIM 1.2: IDENTIFY POTENTIAL ITEMS FROM THE LITERATURE POTENTIALLY PERTINENT TO THE FOUR DOMAINS: FAMILY NEEDS, HEALTHCARE USAGE, FUNCTIONAL LIMITATIONS, AND DIAGNOSTIC CONDITIONS VARIABLES.

To populate the first iteration survey with potential items, 1411 abstracts were reviewed; 1086 were excluded, resulting in 325 articles selected for full-text review (Figure 3). Of these 325, 132 articles contributed variables that informed potential Delphi items in iteration 1. Figure 4 reports that 199 variables were extracted from the 132 articles, with 88, 37, 56 and 16 variables informing potential items in the family needs, healthcare use, functional

limitations, and diagnostic conditions domains, respectively. The Delphi survey development committee refined these variables by using 13 to directly inform a final item, collapsing 147 related variables into 41 items, omitting 37 variables from use, and creating *de novo* a single item. The single item created was DC1 (Table 1 – "Diagnostic Conditions" item 1).[a] This process resulted in 28 items populating the family needs domain in the first iteration of the survey, 14 items in the healthcare use domain, 9 items in functional limitations, and 4 items in diagnostic conditions. The second column of Table 1 lists the original items and links the items to their identification codes used throughout the remainder of this document.

A detailed description of the variables and items contributed by each article can be viewed in Appendix 1. On average each article contributed 4.88 variables which informed 4.81 potential items for consideration in the first iteration. There were multiple patterns of variable and item contribution leading to these means. Some articles contributed multiple unique variables that were subsequently collapsed into one item; some contributed single variables that informed multiple items; some contributed single variables which informed a single item; and some contributed variables that were omitted from future use as items.

For each item presented in the initial survey, Appendix 3 reports the number of articles that contributed to the creation of that item. A mean of 12.6 articles contributed to each item. This mean was skewed to the right by several items in the *Healthcare Use* and F*unctional Limitations* domains which were common defining items of CMC and/or of common research interest in the literature. Several items across all four domains had less than five articles

[a] DC1 = "The child does not need a medical diagnosis in order to be considered a child with medical complexity as long as the other 3 domains (healthcare use, functional limitations, and family needs) are fulfilled."

contributing to their creation. The Delphi survey development committee decided to include these latter items due to the high likelihood that the Delphi respondents would deem them important to the definition.

8.2. AIM 2: PERFORM A DELPHI SURVEY PROCESS

8.2.1. AIM 2.1: RECRUIT PARTICIPANTS TO THE DELPHI PROCESS.

A total of 82 professionals were recruited to participate as Delphi respondents. It is not possible to know the exact size of the population the sample was drawn from as a membership roster for many of the professional societies is not available. One respondent partially completed the first survey and then withdrew. The remaining 81 respondents continued, and each respondent was eligible to complete the subsequent surveys, independent of whether they completed any previous iteration(s). Table 2 reports the percentage of new, returning, and withdrawn subjects for each iteration. Table 3 reports the proportion of respondents who completed zero, one, two, three or all four iteration surveys. Table 4 reports the proportion of respondents who fully completed, partially completed or did not complete surveys across the four iterations.

The demographic distribution of respondents changed only slightly over the iterations (table 5). Of the respondents who completed any part of a survey, an average of 94% volunteered their location of primary practice at least once over the four iterations, 93% volunteered their years of experience, and 93% volunteered their number of publications as first or last author in the last three years (measure of research expertise in the field). Across iterations, an average of 18% practiced in Canada and 82% in the U.S.A. It is noteworthy that a

disproportionate number of respondents practiced in Ohio (24% of American respondents). There was a bimodal distribution in experience with an average of 51% having >15 years clinical experience and 25% having 5-10 years of clinical experience. Regarding research experience, the distribution of journal publications was skewed to the left, with 50% having 0 publications as first or last author in the past 3 years and 42% having 1-5 publications. Most respondents were physicians (66%) followed by nursing professionals (19%), but there was a wide representation of professionals with allied health, administrators and primary researchers contributing. Of note, a respondent could indicate more than one profession.

8.2.2. AIM 2.2: ENGAGE THE DELPHI PARTICIPANTS TOWARDS CONSENSUS ON THE ITEM LIST FROM AIM#1.

8.2.2.1. LOGISTICS OF THE DELPHI PROCESS

The four iterations of the Delphi process with three inter-iteration periods took 166 days to complete during spring to fall of 2020 (Figure 5). Each iteration took 24-25 days with 21 scheduled days of survey availability and a routine 3-4 day extension. The inter-iteration periods were determined by the time it took to prepare the upcoming survey and the availability of the Delphi survey development committee.

Table 4 reports the completion rates of each iteration determining the proportion of fully-completed, partially-completed or incomplete surveys. Incomplete surveys were those that were left completely blank. A survey was partially completed if any question was answered on the survey. The trend showed declining response rates over iteration 1-3, and a recovery in response rate in iteration four. On average across iterations, 70% of potential respondents fully

completed the survey, 4% partially completed the survey, and 26% did not initiate the survey. Respondents could skip questions or cut the survey short, therefore response rates for each item differ somewhat from the overall response rate of the survey. However, the rates and trends closely mirror that reported for the survey as a whole (Appendix 7).

Considering the 82 respondents did not have to complete a former iteration survey to qualify to complete a latter survey, some respondents completed 0, 1, 2, 3, or 4 surveys. Fifty-two percent of respondents completed all four iterations with the remaing proportion completing 0-3 iterations (Table 3). In iteration 2-4, 69-74% of respondents returned from the previous iteration, as well 9-16% were new respondents relative to the previous iteration and 11-19% of the previous iteration repsondents did not return. The majority of respondents completed all iterations, however a substantial number came and went throughout the iterations leaving the respondents group heterogenous iteration to iteration.

8.2.2.2. DELPHI ITERATIONS AND CONSENSUS

Table 6 reports the percentage of respondents who agreed or disagreed with each level 1 item across the four iterations, while Table 7 reports summary statistics of Table 6. In iterations 1-4, the number of level 1 items reaching consensus for inclusion (agreement with the item) was 25, 10, 10, and 3, for a total of 48 items, respectively. Of this total, 24 level 1 items reached consensus in the *Family Needs* domain (86% inclusion consensus rate within *Family Needs*), 13 reached consensus in *Healthcare Use* (93%), 9 reached consensus in *Functional Limitations* (100%), and 2 in *Diagnostic Conditions* (50%). Additionally, one level 1

item reached consensus for exclusion (Table 1 - DC2)[b]. The average proportion of agreement decreased as the iterations progressed (0.74 → 0.71 → 0.70 → 0.63) coinciding with a higher proportion of contentious level 1 items remaining as time progressed. Similarly, the average percentage of disagreement increased as the iterations progressed (0.13 → 0.16 → 0.19 → 0.23) (Table 7). When examining only level 1 items that reached consensus for inclusion, the average proportion of agreement was relatively high in the first iteration (0.90) and plateaued in iterations 2-4 (0.85 → 0.84 → 0.85).

There were 21 level 2 items for consideration in this survey with 6 in the *Family Needs* domain, 11 in *Healthcare Use*, 1 in *Functional Limitations* and 3 in *Diagnostic Conditions* (Table 8). Five level 2 items reached consensus during the iterations, with 4 after the fourth iteration (3 in *Healthcare Use* and 1 in *Diagnostic Conditions*) and 1 after the third iteration (*Healthcare Use*). After the iterations were complete and obeying the rules set forward to deal with unresolved level 2 items (section 7.3.2), 11 level 2 items reached consensus after the iterations were complete, with 3 from *Family Needs*, 6 from *Healthcare Use*, 1 from *Functional Limitations* and 1 from *Diagnostic Conditions*. Of note, one level 1 item reached consensus for exclusion (Table 1 - DC2) and hence the corresponding level 2 item was removed from further consideration.

Respondents had the opportunity to participate in each survey iteration, independent of their participation in previous iterations. The completion rates of the four iterations (Table 4) suggest that individual respondents came and went throughout the iterations setting up a

[b] DC2 = The child must possess ≥1 medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain. However, the degree of chronicity and complexity of the diagnosis does not matter.

situation with potential for ongoing selection bias, specifically voluntary response bias and attrition bias (Section 9.2.3). Table 9 and 10 reports the number of new, returning, and withdrawn respondents in each iteration, relative to the previous iteration, stratified by each level 1 item. Across iterations 2-4 and across all considered Level 1 items, an average of 7.7 respondents were new to the iteration, 47.9 returned from the previous iteration, and 11.8 did not return from the previous iteration.

Table 9 examines the dynamic nature of agreement from a previous iteration to a current iteration. It reports, within an iteration for each level 1 item, the overall proportion of agreement, the proportion of agreement from new respondents, the proportion of agreement from withdrawn respondents in the previous iteration, and the overall proportion of agreement from the previous iteration. Two situations could theoretically exist as iterations progress that could contribute to biased amount of agreement: 1) the loss of disagreement from withdrawn respondents and 2) the gain of agreement in new respondents. After adjusting for multiple comparisons with the Simes False Discovery Rate method there was no significant examples of a biased increase in agreement.

Table 10 examines the proportion of disagreement from a previous iteration to a current iteration. Again, biased disagreement could theoretically arise through two situations: 1) loss of agreement from withdrawn respondents and 2) gain of disagreement from new respondents. After adjustment for multiple comparisons with the Simes False Discovery Rate method, there were no significant examples of a biased increase in disagreement.

Respondent Comments

Appendix 8 presents the comment themes for each item that were presented to the respondents. The average rate of comment submission (those leaving comments divided by those that completed the item) was 23% with a minimum of 4% and a maximum of 57% across all items and iterations. The table separates the comments into those that argued for the inclusion of the level 1 item in the definition (labelled as affirmative), those that argued against level 1 inclusion (dissenting), and those that explained the rationale behind choosing a level 2 item value. Fourth iteration comments are not included because they did not inform a future survey and comments from a level 1 or level 2 item that reached consensus are not included for the same rationale.

In the first iteration respondents had the opportunity to submit comments suggesting changes to the wording of level 1 or level 2 items. Table 1 illustrates the original wording of the first iteration level 1 items and level 2 items, as well as the altered item wording that was used for consideration in iteration 2-4. In total, 24 of the 55 level 1 items had changes to the wording and 18 of the 21 level 2 items had changes. The number of level 1 items that changed in the Family Needs domain, Healthcare Use, Functional Limitations, and Diagnostic were 15 of 28 total, 3 of 14, 2 of 9, and 4 of 4, respectively. The number of level 2 items that changed in the Family Needs domain, Healthcare Use, Functional Limitations, and Diagnostic were 6 of 6, 11 of 11, 1 of 1, and 0 of 3, respectively.

Post-Iteration Modification of Items

Prior to post-iteration modification of items there were 48 items that reached inclusion consensus. After collapsing items, there were 39 items included on the final definitional tool (25

in the *Family Needs* domain, 10 in *Healthcare Use*, 2 in *Functional Limiations* and 1 in *Diagnostic Conditions*) (Table 11). Within the *Functional Limitations* domain both items referring to the presence of limitations in vital body structure and functions (VBSF) and in activities of daily living (ADL) (Table 1 - FL2 and FL3)[c,d] reached consensus to include. Additionally, the 6 items presenting differing levels of complexity and dependence on medical technology (Table 1 - FL4-9)[e,f,g,h,i,j] reached consensus to include. For simplicity of our definitional tool, FL2 and FL3 were collapsed into one item and considering the presence of medical technology depends on a limitation in either an ADL or VBSF, FL4-9 were redundant and removed from the final tool (Table 11 – Item 3)[k]. Similarily, items referring to the presence of chronic conditions and

[c] FL2 = Currently, the child has impactful limitations in performance of activities of daily living (ADLs).
[d] FL3 = Currently, the child has impactful limitations in vital body structures or functions (VBSFs).
[e] FL4 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in IMMEDIATE, significant impact on physiologic function.
[f] FL5 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in significant impact on physiologic function that is NOT IMMEDIATE but would occur within 1 WEEK.
[g] FL6 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in significant impact on physiologic function that would occur after 1 WEEK.
[h] FL7 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would make it IMPOSSIBLE to perform the activity or participate in the life situations.
[i] FL8 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would SEVERELY LIMIT the performance of the activity or participation in the life situations.
[j] FL9 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would PARTIALLY LIMIT the performance of the activity or participation in the life situations.
[k] Item 3 = Currently, the child has impactful limitations in vital body structures or functions or in the performance of activities of daily living.

complex chronic conditions (Table 1 - DC3 and DC4)[l,m] both reached consensus for inclusion. Considering complex chronic conditions (DC4) are a subset of chronic conditions (DC3) the former was redundant and removed (Table 11 – Item 1).[n] Last, FN5 and FN6 both reached consensus and described the amount of caregiver time spent on activities of daily living (Table 1 - FN5)[o] and on direct medical care (Table 1 - FN6).[p] Considering the level 2 item for both reached consensus on "10 hours per week" the items could be collapsed into a single item (Table 11 – Item 21).[q]

[l] DC3 = The child must possess ≥1 'chronic' medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain.
[m] The child must possess ≥1 medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain. However, the condition must be considered a 'complex' medical diagnosis.
[n] Item 1 = The child must possess a minimum of 1 diagnosed, or unknown but suspected, chronic medical condition(s).
[o] FN5 = Over the past 12 months, there is an impactful amount of caregiver time spent on the direct medical care of the child.
[p] FN6 = Over the past 12 months, there is an impactful amount of caregiver time spent on care related to activities of daily living beyond what is age-appropriate need.
[q] Item 21 = Over the past 12 months, there is a minimum average of 10 hours/week of caregiver time spent on the direct medical care of the child or on care related to activities of daily living beyond what is age-appropriate need.

9. DISCUSSION

Without a standardized definition of Children with Medical Complexity, multinational stakeholders have difficulty determining generalizable prevalence estimates, epidemiological measures, and efficacious interventions. Single centre studies difficult to generalize, and multi-centre studies are difficult to standardize across sites. The definitional framework for CMC created by *Cohen et al.* 2011 requires that a definition fulfill four domains: 1) increased family needs, 2) increased healthcare use, 3) presence of functional limitations, and 4) presence of a chronic condition. These four domains were collated from work done by dozens of researchers using similar definitions in the literature.[36, 108] Our current study, utilizing the Delphi methodology, is the first to provide consensus-based criteria to fulfill these four domains, taking the field closer to an precisely-defined, universal definition. This was completed through the efforts of a multi-disciplinary group of experts from Canada and the U.S.A. Thus the definition proposed by this research is based on the work of dozens of researchers and refined by the opinions of dozens of frontline Complex Care clinicians.

As will be discussed in the following sections, four foundational domains, and the corresponding items, routinely appear in other definitions, giving further weight to their importance. Furthermore, the van der Lee *et al.* 2007 and Cohen *et al.* 2011 frameworks require that when fullfilling the domains one needs to consider the age and developmental stage of the child. Childhood is a dynamic time in which abilities and needs mature rapidly. In the current study, experts were asked when choosing level 1 and level 2 items to answer based on what is beyond the expections for children of the same age or developmental stage. Therefore, the experts appropriately judged items representing all four foundational domains,

and the items (level 1) and fulfillment thresholds for each item (level 2) were considered in an appropriate age and developmental context.

9.1. FINAL DEFINITIONAL TOOL

The Delphi process culminated in 48 items (89% item inclusion rate) reaching consensus for inclusion in the definitional tool. Comparing this rate of consensus to that found in the literature is difficult as each Delphi research question, research context, and expert group is unique. Having said that, recent Delphi studies, if they reported the consensus rate at all, report a range of 29%-100% of items reaching consenus.[194, 212-231]

After modification of the items due to item redundancy, the final definitional tool had 39 items that could fulfill one of the four definitional domains. Modification of consensus items is a relatively common occurrence in the Delphi literature. Some authors will alter or remove items due to redundancy, which was the motivation for alteration in this study.[223] Other studies alter wording and syntax of consensus items to improve usability which is often done in patient and public consultation, focus groups or consensus meetings after the initial Delphi study.[213, 223, 227, 232-234]

9.2. PROGRESSION TOWARDS CONSENSUS

9.2.1. RESPONDENT DEMOGRAPHICS

The expert group in this study represented a diverse group of professionals. They worked in a wide geographic range within Canada and the U.S.A with a slight over-representation of Canadian experts (18%) likely due to active recruitment from the Complex Care Section of the Canadian Pediatric Society. Of the experts from U.S.A., 24% worked in Ohio.

While the reason for this latter disproportion is not entirely clear, it may be a consequence of the snowball sampling technique paired with influential, engaged experts in the Ohio medical community. Additionally, Ohio has a high concentration of well-established complex care, clinical programs. Regarding clinical experience of the experts, there was a noticeable under-representation of experts with 10-15 years of experience relative to the flanking categories (5-10 years and >15 years). Pediatric complex care is a relatively new field skewing the experience to the lesser, and the ">15 years" category as defined encompasses a greater range relative to the other categories. Last, the professional diversity is impressive with representation from 13 categories, ensuring representation from varied experiences. However, 66% of the experts were physicians and therefore this group had the potential to dominate the consensus. Although we have no way of knowing the professional representation of the societies from which the expert group was sampled, the membership of at least two of the sampled professional societies are likely dominated by physicians (American Academy of Pediatrics and Canadian Pediatric Society).

9.2.2. RESPONSE RATE

Relative to other Delphi studies, the response rate in the present study is very good, reducing the likelihood of attrition bias. The response rate (including those that fully and partially completed a survey) was 81.5%, 72.5%, 68.7%, and 70% in iterations 1-4, respectively. These rates are in keeping with reported response rates in the recent Delphi literature of 55%-100%, dependent on the iteration.[169, 194, 212-220, 233-245]

9.2.3. PROGRESSION TOWARDS CONSENSUS

Agreement in a Delphi reflects complex individual decisions with multiple, sometimes covert, influences.[246] In the current study, respondents were far more likely to agree with inclusion of an item than to disagree. The average proportion of respondents agreeing with an item ranged between 0.74 and 0.63 depending on the iteration. This is compared to average disagreement ranging between 0.23 and 0.13. Interestingly, the proportion of level 1 item agreement progressively decreased and disagreement progressively increased as the iterations progressed. The agreement eventually led to the acceptance of 89% of potential items for the definitional tool. In short, a pattern was seen of higher agreement than disagreement and a progression through the iterations of decreasing agreement and increasing disagreement.

First, this pattern may represent the legitimate, cumulative consensus with retention of contentious level 1 items as the iterations progressed. This is an expected feature of consensus building technique as quick acceptance of non-contentious items in the early iterations would show higher agreement and lower disagreement. As non-contentious items are removed and a higher proportion of contentious items remain, agreement would decrease and disagreement would increase. Another interpretation of this pattern could be that CMC domains are inherently inclusive and respondents felt that a wide range of biopsychosocial items may fulfill these domains. This would suggest that there a many ways to be complex due to the extensive number of variables in a CMC's life that can contribute to medical complexity. Alternatively, the seemingly disproportionate amount of agreement may reflect bias in the design, including acquiescence bias, majority opinion bias, and satisficing. However, steps were taken in the design of the survey to avoid these biases.

9.2.3.1. ACQUIESCENCE BIAS

In survey research, acquiescence bias refers to the tendency for respondents to disproportionately select a positive response regardless of their true beliefs.[247, 248] In populations of social media users, 10-20% of respondents succumb to this bias when answering survey questions.[249] Associated demographic and methodological risk factors include cultural differences, lower levels of education, and older age.[247, 250, 251] A form of social desirability bias, termed agreeableness, may explain the cultural differences seen in the prevalence of acquiescence bias. Pressure to conform to social norms leading to agreeable behaviour may unconsciously influence a respondent to express more agreement than their opinion would otherwise allow.[248] Additionally, agreeableness may incline respondents to yield to the perceived authority of the interviewer, leading respondents to demonstrate more agreement to answers presented by these authority figures.[248, 249, 252]

The present study limited the affect of agreeableness by recruiting a geographically and professionally diverse expert group which would dilute the effect of any one agreeable cultural norm. Additionally, the respondent group was composed of experts in the field who would not likely see a broad authority gap between themselves and the research team. Last, the survey was administered anonymously online, lessening the pressure for social desirability bias that is seen in interviewer-administered questionnaires.

Survey question design has also been implicated in encouraging acquiesence. When asking for levels of agreement, questions with positive wording tended to lead to greater levels of agreement.[247] For example, a hypothetical question asking “The sun is warm on my face” will induce a greater proportion of agreement than the proportion of disagreement induced by

the negatively worded reciprocal “The sun is not warm on my face”. In the present study, the questioning did not likely contribute to systematic bias as the items were worded as a neutral descriptor of a hypothetical child, and balanced Likert scales were used. Similarly, a question worded to ask directly whether a respondent agrees or disagrees with a statement has been associated with acquiesence bias. For example, “Do you agree that the sun feels warm on your face” is more prone to bias than the neutrally worded “The sun is warm on my face.” Again, this study did not include this method of item wording.

By their nature, Delphi studies that ask for inclusion consensus are at risk for acquiescence bias for two main reasons: 1) despite careful wording, questions are still asking for agreement whether items should be included and 2) feedback always adds the temptation for respondents to acquiesce to the authority of the majority (majority opinion bias). A blunt instrument to limit bias, including acquiesence bias, in Delphi studies is to utilize a conservatively high consensus threshold. If 10-20% of people answering surveys provide answers biased by acquiesence, a high consensus threshold (80% in the present study) should provide buffer against these indviduals. Compared to other recent Delphi studies, our 80% consensus is higher than most. Most commonly the threshold is 70 or 75% in the recent literature with a range from 70-95%.[31, 169, 212, 213, 215, 216, 219, 221-223, 227, 228, 232-234, 243, 245, 253-255]

9.2.3.2. MAJORITY OPINION BIAS

It is critical in a successful Delphi study to provide the opportunity for an individual respondent to change their opinion in subsequent iterations. Two factors have been found to contribute to the willingness to change opinion: respondent seniority and the provision of

structured feedback.[256] However, respondent seniority has not consistently been found to be a factor in opinion change, leaving structured feedback as the predominant factor.[257, 258] The use of structured feedback tends to shift opinion toward the majority, termed here as majority opinion bias, especially if the majority is viewed by the respondents as representing an authority.[257, 259-263] For example, Barrios *et al.* 2021 discovered that if ≥75% of the respondents favoured an particular opinion then a further shift toward the majority opinion occurred.[246] Further evidence to this is that statistical feedback, such as measures of central tendency and dispersion, provoke opinion change to a greater extent than argumentative feedback, indications of opinion rationale.[258, 263, 264] Thus structured feedback moves respondents towards the majority opinion, especially if the majority opinion is clearly described with descriptive statistics.

Although majority opinion bias may explain the disproportionate proportion of agreement, there are several factors arguing against this in the present study. First, the anonymity of the respondents minimizes the perception of seniority or authority among peer respondents. Second, the structured feedback was not solely presented statistically, as argumentative feedback was jointly presented. Third, the highest amount of agreement appeared in the first iteration, i.e. before respondents had the opporuntity to view peer feedback and be influenced by the majority. Finally, despite a disproportionate presence of agreement through the iterations, the proportion of agreement decreased, and proportion of disagreement increased as iterations progressed. The opposite would have been expected if respondents were swayed by the majority opinion. Although not likely a factor in this study, it is

interesting that anonymous group feedback was designed to suppress the influence of the loud, influential expert but may have replaced this influence with the tyranny of the majority.

9.2.3.3. SATISFICING

Last, a respondent behaviour called satisficing may bias reponses towards higher agreement and may be associated with questionnaire design. Satisficing is a “good-enough” behaviour in which the respondent selects the answer that is close enough to their beliefs without extensively considering the nuance of the question or each individual answer.[248, 249] It is a method to conserve cognitive effort by the respondent.[248] Satisficing may occur when the survey is directed to an audience not maximally engaged with the research question, when the questions are not clear and when the survey is too long without an adequate amount of time to complete.[265] Some researchers believe acquiesence bias may be nested within the overarching satisficing phenomenon.[249]

In the current study the proportion of agreement was maximum in the first iteration and decreased as iterations progressed. This coincided with a progressively decreasing survey time commitment. If satisficing was present secondary to prolonged survey duration, our observation would fit this satisficing scenario. Efforts taken when planning and piloting the first iteration survey minimized survey length but the first survey still took the survey pilots roughly 45 minutes to complete.

However, countering the likelihood of satisficing, respondents were chosen from a population of professionals with demonstrated comittment to the field which hopefully translated into maximal engagement with this study. Further effort was taken to ensure they had met the inclusion criteria necessary to demonstrate expertise in the field. Last, maximal

clarity of each item and minimal confusion with the mechanics of the survey was sought. Each survey iteration presented instructions for the survey, instructions for each domain section, definition of terms in each item, and was pilotted prior to use.

9.3. ***ITEMS POPULATING THE DEFINITIONAL DOMAINS***

9.3.1. FAMILY NEEDS

9.3.1.1. RESULTS FROM THE CURRENT STUDY

The *Family Needs* domain was orginally devised by Cohen *et al.* to represent "family-identified health care service needs such as medical care, specialised therapy and education needs."[36] Subsequent criticisms of the Cohen 2011 framework pointed out that the impacts of social determinants of health (SDH) were omitted.[35] Preexisting detrimental SDH may negatively affect a families ability to cope with the impact of service needs. Conversely, the family may develop detrimental SDH, not previously present, as an consequence of the service need. Potential *Family Needs* items populating the first iteration survey purposefully represented: 1) preexisting features of the family, including SDH, that would impact coping with the increased need of the CMC and 2) features of the family, including SDH, that resulted due to the impacts of the increased need. The expert consensus included multiple items from both of these scenarios, including items representing SDH.

The items in the *Family* Needs domain in this definitional tool seem to naturally cluster, post hoc, into seven sub-domains:

i. **Financial disadvantage (4)** - Items 17-20 (Table 11)[r,s,t,u] represent difficulties with family finances either as a preexisting issue (item 18 and 20) or as a result of caring for a CMC (item 17-19).

ii. **Caregiver time (3)** - Items 21-23 (table 11)[v,w,x] represent the impacts on caregiver time directly (Item 21) or indirectly (Item 23) resulting from the care commitments needed by a CMC. Item 22 refers to inadequate respite relative to the famly needs which is another proxy for impacts on caregiver time.

iii. **Family disruption (4)** - Disruption in the family's social, psychological or physical stability due to the caregiving demands of the CMC is represented in items 24-27 (table 11)[y,z,aa,bb] with items referring to situations such as family relationship disruption (item 24), family isolation (item 26), or caregiver pyschological and physical health (item 27).

[r] Item 17 = In order to care for the medical needs of their child, the caregiver(s) are experiencing an impactful amount of planned or unplanned work disruption.
[s] Item 18 = Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month causing difficulties in providing the necessary care for the child.
[t] Item 19 = Over the past 12 months, the caregiver(s) are incurring an impactful amount (% of the family income) of expenses for the child's medical care beyond what is expected from raising a child without medical issues.
[u] Item 20= Currently, the child lacks medical insurance to cover basic medical needs.
[v] Item 21 = Over the past 12 months, there is a minimum average of 10 hours/week of caregiver time spent on the direct medical care of the child or on care related to activities of daily living beyond what is age-appropriate need
[w] Item 22 = Currently, the caregiver(s) are receiving inadequate amount of respite care at home relative to their self-reported needs.
[x] Item 23 = Due to the time and effort to care for the child's higher needs, the family is currently having difficulty performing their daily family activities
[y] Item 24 = Currently, there are impactful problems in the family relationships, such as communication difficulties or family conflicts, that are resulting from the demands of caring for a child with medical issues.
[z] Item 25 = The child is experiencing or witnessing impactful disruption in their social life.
[aa] Item 26 = Currently, the primary caregiver(s) are isolated from community or family support which is either negatively impacting the care of the child OR is a result of the care needs of the child.
[bb] Item 27 = The primary caregiver(s) have developed, or had a worsening of, a physical, psychological, or cognitive ailment since assuming the care of the child.

iv. **Unmet social and medical needs (7)** – Items 28-34 (table 11)[cc,dd,ee,ff,gg,hh,ii] describe situations of unmet medical and educational needs of the CMC. These may refer to non-direct evidence of unmet needs, specifically unplanned school absences (item 28) and missed medical appointments (item 29), or directly asking about unmet medical care needs (item 30). Additionally, three items refer to risk factors for unmet needs, specifically referring to ever-changing care needs (item 31) and difficulties with care coordination (items 32 and 33). Last, a single item refers to consequences of unmet care needs (item 34).

v. **Caregiver factors (2)** – Suboptimal parental health understanding and difficulties with language proficiency are represented in items 35 and 36[jj,kk], respectively. Both these items represent caregiver factors that put them at a disadvantage when managing the care of CMC.

[cc] Item 28 = Over the past school year, the child is experiencing a minimum average of 6 days/month of unplanned school absences due complications of their medical issues, assuming the child is regularly attending school.
[dd] Item 29 = Over the past 12 months, the child has missed an impactful number of scheduled, medically-necessary appointments including, but not limited to, preventative or well child care visits.
[ee] Item 30 = Currently, the child is failing to receive management that is considered medically necessary and/or is part of the parent's care goals for the child.
[ff] Item 31 = Over the past 12 months, caregivers are having difficulty providing recommended care because the chronic care routines for the child are not stable and frequently changing.
[gg] Item 32 = Currently, the care of the child is impacted by inadequate care coordination.
[hh] Item 33 = Over the past 12 months, there was a minimum average of 5 hours/week of caregiver time spent on care coordination for the child.
[ii] Item 34 = Currently, the child is experiencing complications resulting from unmet care needs.
[jj] Item 35 = Currently, the primary caregiver(s) have suboptimal health literacy and numeracy relative to that needed to care for the child.
[kk] Item 36 = The caregiver(s) are having difficulty accessing and delivering care to the child with medical issues because they are not proficient in the predominant language of their current community.

vi. **Geographic access to care (2)** – These items ask about remote or rural status of the child (item 37)[ll] and access to adequate transportation (item 38).[mm]

vii. **Child fragility (1)** – This single item asks about the risk for medical decompensation (item 39).[nn] This item was presented as a proxy for the inherent stress and fear experienced by caregivers when managing a child with such fragilty.

Four items did not reach consensus for inclusion after the fourth iteration. Two represented potentially disadvantageous caregiver factors that could contribute to difficulty delivering care including recent immigration to their current country (Table 1 – FN 22)[oo] and being a single parent (FN23).[pp] As evident from the predominant comment themes arguing for exclusion of these items, the respondents felt these two items depended on too many other factors to independently fulfill the *Family Needs* domain (Appendix 8 – Item FN22 and FN23). Another item asked whether lack of enrollment in a "medical home" would fulfill the domain (Table 1 – FN14)[qq] and the respondents felt this item was too specific and many children receive adequate care in other primary care models or subspeciality care models (Appendix 8 – FN14). Last, FN18 (table 1)[rr] asked whether caregivers lacked training or experience. The respondents commented that most parents are not trained or experienced early in the child's medical

[ll] Item 37 = Currently, the caregiver(s) are having difficulty accessing care because the child lives in a remote or rural setting.
[mm] Item 38 = Currently, the caregiver(s) lack access to reliable, specialized transportation necessary for the child's care.
[nn] Item 39 = Currently, the child is at risk for rapid medical decompensation or has had frequent life-threatening events at home.
[oo] FN22 = The primary caregiver(s) recently immigrated to their current country of residence.
[pp] FN23 = The current primary caregiver of the child is a single parent.
[qq] FN14 = Currently, the child is not enrolled in a primary care practice that meets the definition of a 'medical home' as defined by the American Academy of Pediatrics.
[rr] FN18 = Currently, the child is cared for by primary caregiver(s) who are not fully trained or experienced in the child's care needs.

course but with training this improves (Appendix 8 - FN 18). Overall, exclusion of these four items demonstrated that broadly defined items were unacceptable and the respondents seemed to prefer items more directly related to the experience of CMC.

The *Family Needs* domain in the Cohen *et al.* 2011 framework was meant to integrate increased time devoted to direct care, frequent medical appointments, increased care coordination, and financial burden into the definition.[36] Our new definitional tool (Table 11) expands on this with pointed items that touch on all of these impacts of caring for a child with CMC. Expanding on the original framework, many items represent pre-existing risk factors for these detrimental impacts (as seen in item 18, 20, 31-33, 35-39).[ss,tt,uu,vv,ww,xx,yy,zz,aaa,bbb] Additionally, SDHs were represented with items providing descriptors of five of the six concepts of the Artiga and Henton 2018 model of SDH including: 1) "economic stability", 2) "Neighborhood and Physical Environment", 3) "Education", 4) "Community and Social Context", and 5) "Health Care System" (Figure 2).[119] The final concept of SDH without representation in

[ss] Item 18 = Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month causing difficulties in providing the necessary care for the child.
[tt] Item 20 = Currently, the child lacks medical insurance to cover basic medical needs.
[uu] Item 31 = Over the past 12 months, caregivers are having difficulty providing recommended care because the chronic care routines for the child are not stable and frequently changing.
[vv] Item 32 = Currently, the care of the child is impacted by inadequate care coordination.
[ww] Item 33 = Over the past 12 months, there was a minimum average of 5 hours/week of caregiver time spent on care coordination for the child.
[xx] Item 35 = Currently, the primary caregiver(s) have suboptimal health literacy and numeracy relative to that needed to care for the child.
[yy] Item 36 = The caregiver(s) are having difficulty accessing and delivering care to the child with medical issues because they are not proficient in the predominant language of their current community.
[zz] Item 37 = Currently, the caregiver(s) are having difficulty accessing care because the child lives in a remote or rural setting.
[aaa] Item 38 = Currently, the caregiver(s) lack access to reliable, specialized transportation necessary for the child's care.
[bbb] Item 39 = Currently, the child is at risk for rapid medical decompensation or has had frequent life-threatening events at home.

this definitional tool is “Food security” which did not present in the original scoping review as a variable associated with CMC and was not requested for addition by the expert panel. The presence of items covering the SDH helps resolve some of the original cirticisms of the Cohen *et al.* 2011 definitional framework (section 5.5).[35]

9.3.1.2. COMPARISON TO OTHER DEFINITIONS USING FAMILY NEEDS

There are limited examples in the literature of clinical definitions utilizing family needs as a component. The National Survey for Children with Special Healthcare Needs (NS-CSHCN), the Complex Care for Kids Ontario Program (CCKO), and the World Health Organization – International Classification of Functioning framework (WHO-ICF) provide the major definitions in the literature that share *Family Needs* items.

National Survey for Children with Special Healthcare Needs

The National Survey for Children with Special Healthcare Needs (NS-CSHCN) contains multiple questions highlighting a variety of family needs and SDH.[266] This survey is delivered to CSHCN and questioning starts with five screening questions (CSHCN screener) designed to identify these children. The five screener questions do not touch on *Family Needs* topics and hence the definition of CSHCN does not include *Family Needs* items. However, beyond the initial screener questions, the greater NS-CSHCN questionnaire touches on *Family Needs* themes similar to the definitional tool in this study including:

i. **Financial disadvantage** including questions about total household income, adequate health insurance, medical expenses, financial problems due to the care of the child, and work disruption.

ii. **Caregiver time disruption** including questions about caregiver time spent on direct care and difficulties securing respite. Notable, is the addition of Item 7 (Table 11)[ccc] regarding disruptions in family time in our definitional tool which is not present in the NS-CSHCN.

iii. **Family Disruption** is hardly covered with just a single question regarding the need for caregiver mental health counselling and a question regarding the impact of a child's condition on the child's ability to do things. This entire subdomain in our definitional tool seems to be a novel addition relative to the questions asked by the NS-CSHCN.

iv. **Unmet social and medical need** including a direct question regarding presence of unmet care needs and several questions on the presence of care coordination, the caregiver time spent on care coordination, and the stability of the child's care routine. Our new definitional tool contains additional items regarding missed medical appointments and school days.

v. **Caregiver factors** as they are presented in our definitional tool are indirectly covered in the NS-CSHCN with questions relating to parental education status, caregiver primary language and the need for an interpreter.

vi. There were no questions in the survey related to the final two subdomains: **Geographic access to care** and **Child fragility.**

The overlap in *Family Needs* themes between our definitional tool and the NS-CSHCN is expected. CMC are a subset of CSHCN and descriptive items of each group from the literature

[ccc] Item 7 = Due to the time and effort to care for the child's higher needs, the family is currently having difficulty performing their daily family activities.

will be similar. Our study however identified items that experts felt were important enough to contribute to a definition of CMC, whereas similar items in the NS-CSHCN were chosen to describe CSHCN, but not necessarily to define them.

Complex Care for Kids Ontario

The Complex Care for Kids Ontario (CCKO) is a Provincial-wide, multi-site clinical program for managing CMC in the Canadian province of Ontario.[267] The entrance criteria for this program represents a standardized, widely-used definition. Although *Family Needs* items do not represent a signficant portion of the CCKO entrance criteria, this program does have some overlapping items compared to our definitional tool (table 11). Under the CCKO "Fragilty" domain there are a number of items referring to the risk of unpredictable, life-threatening health deterioration. This compares to item 39 (table 11).[ddd] Additionally, under the "Technology Dependent" domain an item exists stating that the child is dependent on caregivers for ADLs or requires constant medical supervision, comparable to item 21.[eee] Additionally, the entire "Geography" domain demands the presence of significant challenges to seeking services due to rurality or care access. Compared to the mandatory nature of this domain in the CCKO entrance criteria, the current definitional tool has Item 37 in the *Family Needs* domain, referring to difficulty accessing care due to a rural or remote living setting.[fff] Last, under the "Complexity" domain an item stating that the family has circumstances that

[ddd] Item 39 = Currently, the child is at risk for rapid medical decompensation or has had frequent life-threatening events at home.
[eee] Item 21 = Over the past 12 months, there is a minimum average of 10 hours/week of caregiver time spent on the direct medical care of the child or on care related to activities of daily living beyond what is age-appropriate need.
[fff] Item 37 = Currently, the caregiver(s) are having difficulty accessing care because the child lives in a remote or rural setting.

impede their ability to provide care or decision making. This a more general item encompassing the spirit of the entire *Family Needs* domain. In this current study an attempt at operationalizing the family needs domain was undertaken whereas the CCKO incorporated the *Family Needs* concept in a more broad way.

World Health Organization – International Classification of Functioning

Although the NS-CSHCN and the CCKO represent practical definitions in use epidemiologically and clinically, respectively, the WHO-ICF framework is a conceptual model, albeit with a coding strategy (Figure 6).[95] A similarity between the results of the current study and the WHO-ICF is expected considering the WHO-ICF was an influence on van der Lee *et al.* 2007 when crafting the definitional framework for children with chronic conditions.[108] The WHO-ICF model conceives of disability as an equilibrium of three interacting factors: 1) Body structures and function, 2) Activity, and 3) Participation. The relationship of these three factors influence, and are influenced by, three further factors: a) Health condition, b) Environmental factors, and c) Personal factors. As can be seen, the biopsychosocial considerations of the ICF model mirrors the same considerations informing the four domains in the Cohen *et al.* 2011 concept, and hence our definitional tool. The presence of environment factors in this model acknowledges the impact Family Needs has on health and disability. Environmental factors refers to the social model of disability in which physical factors (ex. climate, terrain), social attitudes, institutions, and laws impact disability and health.

9.3.1.3. DIFFICULTIES IN OPERATIONALIZING THE FAMILY NEEDS CONCEPT

The definition of CMC in the current study and the definitions of children with chronic disease developed by Stein *et al.* 1997 (Section 9.3.2.3), van der Lee *et al.* 2007 and the CSHCN screener all require the presence of a chronic condition, measures of functional limitation, and measures of increased healthcare. [79, 108, 266] The predominant difference is the family needs component required in the CMC definition. Cohen *et al.* 2011 made the argument that the *Family Needs* domain is necessary to differentiate CMC from children with chronic health conditions as defined in van der Lee *et al.* 2007.[36, 108] This domain also seems to be the most difficult to standardize. The other three domains have a narrower focuss. The *Family Needs* domain is broader and standardization has to account for numerous social and environmental elements. Although this broad spectrum was limited to items with a previously demonstrated association with CMC in the literature, and further limited based on the opinions of the experts, in reality a far greater number of *Family Needs* influences likely exist. A child's "family need" may be a highly individualized constellation of interacting factors. Despite this, the rate of consensus for inclusion in the *Family Needs* items was in line with the other domains and like the other domains there was no extra items suggested by the experts in iteration 1. However, compared to the other domains, the wording of *Family Needs* items contain more subjectivity because they were represented this way in the literature and/or at the request of the experts during consensus building.

9.3.2. HEALTHCARE USE

9.3.2.1. RESULTS FROM THE CURRENT STUDY

Increased healthcare utilization has been advocated by some researchers as a proxy to identify CMC.[268] Using children with neuromuscular complex chronic conditions (CCC) as an example this logic is illustrated. Compared to all children with any CCC in Ontario, Canada, children with CCC plus neurologic impairment were more likely to experience hospital readmission, homecare use, medication use, and unplanned visits to the Emergency Room.[269] Furthermore, care coordination and case management interventions have been shown to decrease rates of hospitalization in children with neuromuscular CCC requiring technology assistance.[58] Together this evidence suggests that increased healthcare use is associated with the presence of neuromuscular CCC in children and in these children healthcare use is decreased in response to care coordination and case management, which is a common management strategy for CMC. It is possible that increased healthcare use may be an indicator of a child's degree of complexity, however, the literature making this connection is scarce and using increased healthcare utilization as the sole identifier of complexity should be used with caution. Having said this there are many complex care clinical programs that lean heavily on healthcare utilization metrics as a clinic enrollment criteria out of necessity (e.g. availability of health database) and priority (e.g. lowering costs in a Medicaid system).[39, 45, 267] With our current knowledge, using increased healthcare use as one of multiple domains to identify CMC, as the Cohen *et al.* 2011 framework suggests, and ensuring increased healthcare use is fulfilled using objective items, as this study contributed, seems to be a more appropriate method to define CMC.

The Cohen *et al.* 2011 diagnostic framework included the *Healthcare Use* domain to capture the the high utilization of health resources by this population.[36] Examples of this included "frequent and prolonged hospitalization, multiple surgeries, or the ongoing involvement of multiple subspecialty services and providers."[36] Other authors have also identified the intensity and breadth of healthcare use to be a major component of complexity. "Healthcare" in this context includes medical treatments (e.g. diets, medications, therapies and medical technology) and health services (e.g. subspecialist care, primary care, homecare).[118] Our definitional tool operationalizes this concept through the demonstration of consensus on different types and thresholds of healthcare usage associated with CMC in the literature. It was notable that 10 out of 11 level 2 items reached consensus, aiding in operationalizing the domain.

This domain is primarily populated by items describing thresholds for the need for homecare personal or the excessive need for school nurse time, medical interventions, healthcare providers, outpatient clinic visits, ER services, hospital service, and/or ICU services. All items were operationalized with a level 2 item. Only one item did not reach consensus out of a total of 14 items presented. That item represented the need for healthcare services to be delivered in multiple healthcare and non-healthcare settings (table 1 – HU1).[ggg] The comments indicated that the experts felt this it depended on too many other factors, such as the chronicity of service in these settings and the number of services offered in a single setting (Appendix 8 – HU1).

[ggg] HU1 = Over the past 12 months, healthcare services for the child are delivered in at least 3 healthcare and/or non-healthcare settings.

9.3.2.1. COMPARISON TO OTHER DEFINITIONS USING HEALTHCARE USE

Four definitions, NS-CSHCN, “children with chronic conditions”, “chronic health conditions in childhood”, and CCKO, identified from the literature use measures of healthcare utilization in the definitions and provide comparisons to our findings in this study. As previously indicated however, there are numerous other researchers and clinicians who utilize some measure of healthcare utilization to assist in identifying CMC.[268]

National Survey for Children with Special Healthcare Needs

Of the five NS-CSHCN screener questions, two directly relate to increased health utilization.[266] In other words, the definition of a CSHCN includes healthcare use, in addition to 2 questions on functional limitations, and a single question regarding chronic medical conditions. The two questions are in reference to using any medicines other than vitamins and using more medical care, mental health care, or educational services than is age-appropriate. These latter questions are broad but do encapsulate all the items selected in our study. Outside of the screening questions, the NS-CSCHN collects extensive, and more pointed, information relating to healthcare utilization. There are many questions regarding the frequency of accessing homecare, as well as primary, subspecialty, emergency, and hospital care. Furthermore, data is collected on the number of prescription medications and medical equipment in use. Thus, increased healthcare use is a fundamental part of the definition of CSHCN, albeit with broader and more subjective questions than in our study. Additionally, healthcare use is valued when describing these children in the greater questionnaire with questions mirroring the items in this study precisely.

Chronic Health Conditions in Childhood

As previously introduced, van der Lee *et al.* in 2007 reviewed the literature on definitions of "chronic health conditions in childhood."[108] Those authors stated that differences in definitions could be at least partically attributed to unstandardized terminology, which is also a difficulty in CMC iterature and a justification for the importance of the current study. A framework for a tiered definition of this concept was proposed by those authors, based on their review findings. The first tier allowed for the broad identification of children with chronic conditions using ICD-10 lists of *a priori* defined chronic conditions. The second tier narrowed this overly broad group of children to those with functional limitations. The third tier directly related to healthcare use and further narrowed this group to those that are "in need of medical care or related services in addition to what is usual for this age."[108] Therefore, the final refining descriptor in this definition leans completely on healthcare. However, this is a broad, subjective statement relative to our study because these authors were proposing a conceptual definition whereas our goal was to standardized and existing conceptual defintion. As the van der Lee *et al*. 2007 framework was the basis for the Cohen *et al.* 2011 CMC definitional framework, similarities with our study results are expected.[36]

Children with Chronic Conditions

Stein *et al.* in 1997 proposed a definition for children with chronic conditions.[79] Much like our study, those authors started with a multi-domain, consequence-based, conceptual definition which required: 1) presence of a biological, psychological, or cognitive disorder, 2) duration of at least 12 months, and 3) presence of consequences of the disorder including the

concequence domains of “functional limitation”, “compensatory mechanisms or assistance dependency”, and “excessive service use or need”.[82] The authors then moved to operationalize the dimensions by creating discreet and meaningful question sequences (termed “items”) that asked, first, whether the child experienced a consequence, then second, whether the consequence was due to a chronic disorder, and last, whether the disorder lasted greater than 12 months. Like the present study, items were identified from the literature, existing national surveys, interviews with caregivers, and from expert opinion. The authors created operational topics within each consequence domain that organized the 39 identified items. There were 16 operational topics, eight of which were embedded under the “excessive service use or need” domain.

Like the Cohen *et al.* framework, Stein *et al.* saw the value in defining children with chronic conditions by including measures of increased healthcare use. Like our study, Stein *et al.* included the experience of hospitalization, regular doctor visits, and regular nursing care or treatments. However, Stein *et al.* preferred a more general description for these items as they did not further differentiate treatments by complexity level, differentiate doctors visits by type of provider, and differentiate hospitalizations by admissions, length of stay or readmission. However, Stein *et al.* was more specific than our study by including the need for special “arrangements at school” or “instructions or other educational services” whereas our study included only the need for a school nurse. Interestingly, Stein *et al.* included the need for therapy in this domain whereas our study included it in the *Functional Limitations* domain suggesting some potential interplay or overlap between definitional domains (Section 9.3.5). Last, Stein *et al.* added novel topics such as the “need for pyschological services” and the “need

for service that were unobtainable" neither of which were identified in our scoping review or presented to our expert panel for consideration during the first iteration.

Complex Care for Kids in Ontario

The entrance criteria for the CCKO, a large, multisite, clinical program, is not highly dependent on measures of increased healthcare utilization.[267] In the "Technology Dependent and/or Users of High Intensity Care" domain there is an item referring to the presence of a chronic condition that requires a great level of care. A sub-descriptor of this item refers to the need for constant nursing supervision or monitoring, medication administration and therapy delivery. This sub-descriptor relates to several items in the present study including those describing the need for homecare personnel (Table 11 - Item 5)[hhh], and the delivery of interventions for chronic conditions and delivery of complex interventions (Item 6 and Item 7)[iii,jjj]. In the "Complexity" domain there is an item describing the need for a minimum of 5 healthcare providers, with a direct reference to school nursing, in a minimum of 3 healthcare settings. Like the CCKO above, our study directly references the need for a minimum of 3 providers (Item 8)[kkk] and references the need for a school nurse (Item 4)[lll]. However, our expert panel felt a school nurse alone, for a minimum of 5 hours per week, would satsify the *Healthcare Use* domain. In contrast to this study, the CCKO definition seems to priorize

[hhh] Item 5 = Currently, the child requires homecare personnel.
[iii] Item 6 = At any one time over the past 12 months, the child needed a minimum of 4 interventions for chronic medical issues.
[jjj] Item 7 = Over the past 12 months, the child needed a minimum of 1 medical intervention with COMPLEX administration at any one time.
[kkk] Item 8 = Over the past 12 months, the child had a minimum of 3 healthcare providers involved in their care.
[lll] Item 4 = Currently, the child requires a school nurse to manage care needs for a minimum average of 5 hours/week, assuming the child regularly attends school.

measures of primary care with no reference to inpatient useage. Inpatient measures may have been deliberately omitted considering the goal of the CCKO program was to prevent emergency room and preventable hospitalizations. Therefore, waiting for a child to fulfill certain inpatient healthcare use criteria put an unnecessary delay for intervention.

9.3.3. FUNCTIONAL LIMITATIONS

9.3.3.1. RESULTS FROM THE CURRENT STUDY

Functional limitations are often described using the WHO-ICF philosophy, specifically being based on limitations of body structures and function, performance of activities, and participation in society. [36, 95] However, to better represent the degree of severity of the limitation experienced by CMC, Cohen *et al.* 2011 argued that these children typically require assistance from medical technology to augment vital body structure and/or function or in activites of daily living.[36] Other authors share this recognition and see technology assistance as a defining feature of CMC, a feature that demonstrates their fragility.[270-272] In the current study, the expert panel agreed that medical technology fulfilled the *Functional Limitations* domain, but they also included the concept that just the presence of a functional limitation fulfilled the domain. The spectrum of severity was not deemed necessary to fulfill the *Functional Limitations* domain by this group of experts. Experts were originally presented with 6

items (Table 1 – FL4-9)[mmm,nnn,ooo,ppp,qqq,rrr] that allowed for a choice between different degrees of severity of limitation, but chose to include all severities of limitation. By opening this domain to children with any functional limitation, it will include children who were traditionally viewed as not severe enough to be included as a CMC.

Our *Functional Limitations* consensus was subsequently collapsed into only 2 items, if the child either: 1) Required greater than 3 hours per week of therapy or 2) had a functional limitation in vital body structure and function or in activites of daily living. The former item is referring to efforts to adapt to a functional limitation through therapy, while the latter refers directly to the presence of a functional limitation with some added description.

[mmm] FL4 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in IMMEDIATE, significant impact on physiologic function.
[nnn] FL5 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in significant impact on physiologic function that is NOT IMMEDIATE but would occur within 1 WEEK.
[ooo] FL6 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. AND Misuse or discontinuation would result in significant impact on physiologic function that would occur after 1 WEEK.
[ppp] FL7 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would make it IMPOSSIBLE to perform the activity or participate in the life situations.
[qqq] FL8 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would SEVERELY LIMIT the performance of the activity or participation in the life situations.
[rrr] FL9 = The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. AND Misuse or discontinuation would PARTIALLY LIMIT the performance of the activity or participation in the life situations.

9.3.3.1. COMPARISON TO OTHER DEFINITIONS USING FUNCTIONAL LIMITATION

Presented for comparison are four definitions from the literature incorporating functional limitations measures. Also, comparison is made to the trends of using functional limitations in smaller, regional definitions.

<u>World Health Organization – International Classification of Functioning</u>

The items in this domain relate to the model of the WHO-ICF. [95]The WHO-ICF is described in greater detail in the *Family Needs* section above and is represented in Figure 6. As it relates to the *Functional Limitations* domain, item 3 (table 11)[sss] has direct reference to two of the concepts of disability in the ICF: 1) limitations of a body structure or function and 2) limitations in activities of daily living. The other item (table 11 - Item 2)[ttt] refers to the delivery of therapy to treat limitations in body structure and function, activity, and/or participation. This again relates to the WHO-ICF framework except it focuses on the treatment as a proxy for the limitation.

<u>National Survey for Children with Special Healthcare Needs</u>

The NS-CSHCN screener identified these children with two questions related to functional limitation: 1) is there a limitation in the ability to do the things most children can do and 2) is there a need for special therapy.[266] These questions are similar to the two items chosen by the experts the *Functional Limitations* domain in our definitional tool. The NS-CSHCN

[sss] Item 3 = Currently, the child has impactful limitations in vital body structures or functions or in the performance of activities of daily living.

[ttt] Item 2 = Over the past 12 months, the child required a minimum average 3 hours/week of prescribed, specialized therapy.

itself collects a substantial amount of more detailed information on the need for therapy, as well as limitations from disability including the impact on their activities, senses, vital body functions, mobility, fine motor skills, communication, and the ability to participate in a variety of life activities (ex. sports, play, school, etc.).

Children with Chronic Conditions

In general, the *Functional Limitations* domain in this study has similar items to the 8 operational topics presented by Stein *et al.* 1997 in their two consequence domains: "reliance on compensatory modalities" and "functional limitations."[79] As discussed above, the expert panel reached consensus to include more general items in this domain, so it is not surprising that the operational topics in Stein *et al.* demonstrating more specificity. Under the "functional limitations" consequence domain, operational topics include, "unable to play with other children," "restricted in activities," "difficulty feeding, dressing, washing, toileting, walking," and "difficulty hearing, seeing or communicating." Stein *et al.* were more specific with examples of limitations in body tissues and function, activities, and participation, whereas the current study simply referred to limitations in "vital body structures or functions" and "activities of daily living." Under the "Reliance on compensatory modalities" consequence domain, topics include "medication use," "special diet," "special equipment," and "personal assistance." In the current study, "medication use" and "special diet" were covered in the *Healthcare Use* domain when discussing the number of chronic and complex interventions. Again, this demonstrates the potential for overlap and interplay between domains (Section 9.3.5).

Complex Care for Kids Ontario Program

On a regional scale, the CCKO uses entrance criteria that requires the child be dependent on a medical device that compensates for vital bodily functions. The two items in the definitional tool from the current study provide a broader definition of *Functional Limitations* compared to that in the CCKO. The experts in our study decided on a more inclusive definition as compared to the CCKO, which focused on children on the more severe end of the functional limitation spectrum. Additionally, the experts preferred items that added subjectivity to the items realtive to the more easily operationalized concept in the CCKO: the presence of medical technology.

Local Clinical Definitions

On a local scale there are a number of studies and complex care clinics across North American that utilize measures of functional limitations as inclusion criteria for CMCs. [24, 34, 269, 273] The functional limitations concept, in this sense, is often intertwined with the need for skilled, in-home nursing care and the need for medical technology as a proxy for severe functional limitation.[24, 34, 269, 273] Again, the experts in this study chose to utilize a broader, more inclusive operationalization of Functional Limitations.

9.3.4. DIAGNOSTIC CONDITIONS

9.3.4.1. DECATEGORIZATIONAL DEFINITIONS

Prior to discussing the Diagnostic Conditions domain, a presentation of the concept of decategorizational definitions is pertinent. The operationalized definition presented in the

current study is an example of a decategorizational definitions which is a legacy passed from the Cohen *et al.* 2011 and van der Lee *et al.* 2007 definitional frameworks.[36, 108] The decategorization view of definition generation suggests that the consequences of a child's health problems are not deterministically tied to the underlying disease entity.[79, 82, 274] Although the scientific community may be able to define a pathophysiologic disease processes, this does not adequately correlate with the consequences that a child experiences and the care needs that the child requires.[79, 82] There is great heterogeneity of health consequences within children with a certain condition and unless the actual consequences are considered in a definition these children cannot properly be defined. Decategorizational definitions group children by considering their closely shared biopsychosocial consequences independent on which disease entity is underlying.

Additionally, different disease entities share health consequences and splitting these children up into disparate groups is counterproductive.[79, 82] This argument is particularly persuasive when considering the predominant motivation to categorize children, which is to assign pertinent and specific management strategies and resources to that group.[275] A group of children with shared health consequences would often more closely match the resources to manage them relative to a heterogeneous group of children that happened to share the same disease entity. It must be acknowledged however that there are disease specific treatments that must still be reserved for groups of children sharing the same pathophysiologic process.

The early evolution of the definitions for CMC focussed on compiling lists of disease entities as the working definition of CMC. The earliest evidence of this is the CCC list created by Feudtner *et al.* in 2000.[87] This group defined complexity as the possession of a health

condition on a prescribed list of CCCs. Interestingly, the list was compiled by a group of experts guided by a decategorizational conceptual definition. A disease entity would be included in the list if it was "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."[87] Since this time multiple definitions have been proposed that utilize diagnosis lists to define CMCs such as the Patient Medical Complexity Algorithm.[276] However these definitions do not incorporate other domains of complexity. Generally these definitions were designed to be used on the population level, using adminstrative datasets where ICD codes are more easily accessible. Additionally, these datasets offer limited oportunities to operationalize other aspects of complexity, such as the three other domains described here. With time the definition of CMC, as evident by the definitional frameworks proposed by van der Lee 2007 and Cohen *et al.* 2011, took on a decategorizational view. However, these still required a disease entity to be present as a domain, granted the disease entity did not have to be any particular condition or even be yet diagnosed. Additionally, as previous illustrated, many definitions incorporate other domains of complexity to better align with a decategorizational definition philosophy. However, most of these definitions still reference the need for a diagnostic condition of some sort as one of the minimum requirements including the CCKO (a chronic condition is needed), WHO-ICF (references the presence of a health condition), and the NS-CSHCN (requires the presence of a chronic medical condition).[24, 34, 95, 266, 267, 269, 273]

9.3.4.1. RESULTS FROM THE CURRENT STUDY

Despite the benefits, a decategorizational approach leads to two problems that must be overcome: 1) a clear definition must be implemented that is independent of diagnosis, a task already tackled by Cohen *et al.* 2011, and 2) the definition must be operationalized with maximal precision.[79] The items selected for inclusion by the expert panel in this study put greater focus on a shared health state even though the *Diagnostic Conditions* domain is included as a mandatory component of the definitional tool. The experts were given the choice to omit the need for a diagnostic condition all together (Table 1 - DC1)[uuu] and depend on the other domains alone to define CMC. Alternatively, the experts could have fulfilled the *Diagnostic Condition* domain with a condition without any limitations (Table 1 – DC2)[vvv], a condition that is chronic (Table 1 – DC3)[www], or a condition that is chronic and complex (Table 1 – DC4). The experts reached consensus for inclusion of a chronic condition (Table 1 - DC3) and a complex chronic condition (Table 1 – DC4)[xxx] to fulfill this domain, which was subsequently collapsed into a single item (Table 11 – Item 1)[yyy].

[uuu] DC1 = As long as the other 3 domains (healthcare use, functional limitations, and family needs) are fulfilled, the DIAGNOSTIC CONDITIONS domain is not needed for a child to be considered medically complex.
[vvv] DC2 = To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed medical condition (s) OR unknown but suspected medical condition(s). However, the degree of chronicity and complexity of the condition does not matter.
[www] DC3 = To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed 'chronic' medical condition(s) OR unknown but suspected 'chronic' medical condition(s). Note: Hover your cursor on the highlighted text to view a detailed definition.
[xxx] DC4 = To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed 'complex' medical condition (s) OR unknown but suspected 'complex' medical condition(s). Note: Hover your cursor on the highlighted text to view a detailed definition.
[yyy] Item 1 = The child must possess a minimum of 1 diagnosed, or unknown but suspected, chronic medical condition(s).

The results from this work are notable for several reasons. First, by reaching an exclusion consensus that a diagnostic condition without limitations does not fulfill this domain (Table 1 – DC2), the expert panel made clear that if a diagnostic condition is necessary it cannot be anything less than a chronic condition. Second, and perhaps of most note, consensus was not reached on omitting the Diagnotic Conditions domain all together. The agreement on this item was bimodal with 65% agreeing to omit, 30% disagreeing and only 5% neutral on the subject throughout the iterations (Table 6 – DC1). Presence of this item in the original iteration was meant to allow the experts to debate whether the other three domains were all that were needed to define complexity, following a decategorizational approach. Comment themes in support of acceptance of this item suggested that the other three domains were all that was necessary to be considered complex and diagnosis was superfluous (Appendix 8 – DC1). Interestingly, other comment themes highlighted that undiagnosed conditions, novel conditions or changed diagnoses complicate this domain too much and thus argued in support of removing the need for a Diagnostic Condition. However, comment themes arguing against exclusion of this domain made reference to the impossibility of fulfilling the other 3 domains without having a diagnostic condition of some sort, or referencing the risk of having children with minor conditions meeting the definition. This is in agreement with past precendent as many other non-categorical definitions require the presence of a undefined chronic condition for fulfillment.[79, 108, 266, 267] Repeatedly, and in this study, the perception is that a chronic condition is mandatory to define CMC and that the complexity of the condition depends on the other domains.

9.3.5. INTERPLAY BETWEEN THE DOMAINS

The four domains suggested by Cohen *et al.* 2011 have become a foundation for the definition of CMC. It has been presented that medical complexity is a multifaceted entity that requires more than a diagnosis to define. The four domains parse out these facets requiring *Family Needs*, increased *Healthcare Use*, and *Functional Limitations*, in addition to a *Chronic Condition*. A somewhat challenging consideration is the observed interplay of different domains with each other resulting in item overlap between domains, item redundancy between domains or item impact on other domains.

9.3.5.1. ITEM OVERLAP BETWEEN DOMAINS

Items residing in a specific domain may have features that fit within other domains. An example is item 2 (table 11)[zzz] which refers to the presence of prescribed specialized therapy, either delivered by therapists or by the parents with therapist guidance. The expert panel consensus was that this item fit in *Functional Limitations*, but unfortunately no comments were left by the experts justifying the inclusion in this domain. The research team originally included the item in the *Functional Limitations* domain as it was felt this therapy was indicative of a underlying functional limitation. However, delivery of a health service such as this can also find a home in the *Healthcare Use* domain.

[zzz] Item 2 = Over the past 12 months, the child required a minimum average 3 hours/week of prescribed, specialized therapy

9.3.5.2. ITEM REDUNDANCY BETWEEN DOMAINS

Some items may have partial redundancy with items in other domains. For example, item 6 and item 7 (table 11)[aaaa,bbbb] within the *Healthcare Use* domain refer to the presence of an impactful number of interventions for chronic medical issues, including general interventions and complex interventions, respectively. These items have significant overlap with item 21 (table 11)[cccc] in the *Family Needs* domain, which refers to an impactful amount of caregiver time spent on the direct medical care of the child. An affirmative answer to one item in one domain is associated with an affirmative answer to another item in a different domain.

9.3.5.3. ITEM IMPACT ON OTHER DOMAINS

Some items may directly confound another domain. For example in the *Family Needs* domain, Item 20 (table 11)[dddd] refers to the lack of medical insurance, item 29[eeee] refers to the child missing an impactful number of medically-necessary appointments, and item 30[ffff] refers to the failure to receive management deemed medically necessary. These items are indicative of a family at risk of financial hardships due to lack of insurance and indicative of a family having difficulty with care coordination. However, the presence of these items could also theoretically reduce the amount of *Healthcare Use* due to lack of access, not because the child

[aaaa] Item 6 = At any one time over the past 12 months, the child needed a minimum of 4 interventions for chronic medical issues.
[bbbb] Item 7 = Over the past 12 months, the child needed a minimum of 1 medical intervention with COMPLEX administration at any one time.
[cccc]Item 21 = Over the past 12 months, there is a minimum average of 10 hours/week of caregiver time spent on the direct medical care of the child or on care related to activities of daily living (ADLs) beyond what is age-appropriate need.
[dddd] Item 20 = Currently, the child lacks medical insurance to cover basic medical needs.
[eeee] Item 29 = Over the past 12 months, the child has missed an impactful number of scheduled, medically-necessary appointments including, but not limited to, preventative or well child care visits.
[ffff] Item 30 = Currently, the child is failing to receive management that is considered medically necessary and/or is part of the parent's care goals for the child.

is not in need of this healthcare. The underpinnings of this scenario were described by Berry *et al.* 2015 in which the authors stated, “For many children with medical complexity, high health resource utilization is a direct manifestation of the complex interactions among the children's health problems, functional limitations, and healthcare needs.”[118]

It was asked of the experts to consider whether potential items fulfill the respective domain, thus trying to marry an item to a specific domain. Despite interplay between domains, the consensus bore out that this expert panel felt the final items best fulfilled their respective domain. The foundations of this issue seems to do more with overlap between the conceptual domains than the operationalized items themselves. Unmet family needs, such as poor care coordination or disadvantageous social determinants of health, has been shown to increase healthcare utilization.[39, 45, 61, 277-279] Increased healthcare utilization has detrimental effects on family functioning.[272] Functional limitations often lead to increased healthcare utilization and increased family impact and need.[280, 281] Finally, and most obviously, a child’s diagnostic condition sets the stage for the functional limitations, healthcare utilization and family need. These scenarios highlight the multi-dimensional interaction of characteristics that define complexity. The WHO-ICF philosophy likewise highlights that indviduals’ functioning is a complex interplay between conceptual features of their life (figure 6).[95]

9.4. ***DEFINITIONAL CONCEPTS RELATED TO THE DEFINITION OF CHILDREN WITH MEDICAL COMPLEXITY***

There are multiple health states across ages, health states, and geographic space that seem to mirror the issues seen in the definition of CMC. Like CMC, these are not diagnostic

terms but rather a description of shared phenotypes, functional status, social needs, and care needs. Many of these health states utilize a decategorizational approach and all lack a consensus definition, thus limiting standardization of care and research. These entities are in a variety of states of operationalization at this moment, some solely conceptual and others fully operationalized. Two of the most pertinent entities are presented below.

9.4.1. FRAILTY

The concept of frailty in the adult population has parallels to medical complexity in the pediatric population. Many biologic, clinical and socioeconomic factors contribute to frailty which, in the most general terms, is seen as an individual's decreased reserve, decreased ability to maintain homeostasis, and decreased resistance to external stressors leading to functional decline.[99, 282, 283] Therefore, this definition fits into a decategorizational philosophy. Like our definition of CMC, numerous combinations of these inter-related factors can lead to frailty as a common result, and the factors are dynamic leading to waxing and waning of the presence and degree of frailty over time. [99, 282-284] Many authors have developed frailty definitions linking the concept to disability, comorbidity, and/or old age.[285-287] Common features of these definitions are the incorporation of associated, inter-related factors such as declines in lean body mass, strength, endurance, balance, walking performance, and activity levels.[287-291] Learning from these earlier definitions, Fried *et al.* in 2001 proposed an influential, unified phenotypic definition in which three or more of the following concepts had to be present: 1) unintentional weight loss, 2) self-reported exhaustion, 3) weakness, 4) slow walking speed, and 5) low physical activity. The definition is decategorizational and primarily utilizes physiologic measures with two references to activity limitation (4 and 5).[283] Interestingly, Fried's

definition of frailty encapsulates the concept of CMC and therefore frailty could be seen as a feature of CMC. Additionally, as for CMC, the impact of frailty on health has led several international organizations, such as the European Union, to call for the unification of the multitude of definitions into a single standardized definition as frail individuals are high users for healthcare and social resources.[292, 293]

Like CMC, frailty places individuals at higher risk for adverse health outcomes such as disability and functional limitation, increased healthcare utilization (including ER presentations and preventable hospital admissions), higher healthcare costs and death.[282, 283, 294, 295] Fried *et al.*, using their proposed definition, found frailty was associated with poorer outcomes including incident falls, worse mobility, worse ADL performance, hospitalizations and death.[283] Additionally, frailty shares risk factors with CMC. Fried *et al.* found frailty was associated with lower education, lower income, poor health, higher rates of comorbid conditions and disability.[283] Other studies have also shown an association with lower SES measures.[296] Contrary to CMC, where these associated factors occur in early life, frailty in the adult population most often progresses during a persons final 5 to 10 years after having lived a full life.[297-299] Frailty can be seen however in younger adults after experiencing acute illness or injury or younger adults who have lived with chronic disease since childhood.

There have been several studies attempting to reach a consensus definition for frailty using consensus-building methodologies, a step that had not been taken by Fried *et al.* First, Rodriguez-Manas *et al.* in 2013 published the results from the "Frailty Operative Definition – Consensus Conference Project."[217] In that Delphi study, 18% of statements regarding diagnostic criteria were accepted resulting in 6 conceptual domains required for a definition of

frailty. The domains included: 1) suboptimal nutritional status, 2) mental health difficulties, 3) cognitive impairments, and 4) a detriment on a physical performance assessment which must include 5) gait speed and 6) mobility. Unfortunately, there was no consensus reached on the constituent elements required to operationalize this conceptual definition. Unlike CMC, as defined in the present study, frailty utilizes several physiologic measures (1, 5, and 6 above) and other than this focuses exclusively on limitations of body tissues, particularly cognitive and mental health impairments (3 and 4 above), with no specific reference to activity or participation limitations. This definition takes a definite decategorizational approach with no reference to the need of a diganosis.

Morley *et al.* in 2013 published results from a consensus meeting aiming to create an operational definition of frailty.[300] The expert group agreed on a conceptual definition stating that frailty is "A medical syndrome with multiple causes and contributors that is characterized by diminished strength, endurance, and reduced physiologic function that increases an individual's vulnerability for developing increased dependency and/or death." Unfortunately an operationalized definition was out of reach in this study and the experts rather stated that a definitive diagnosis of frailty should come from a geriatrician considering each case individually using other well-validated models of frailty. The concept agreed on here is decategorizational and again focuses greatly on physiologic concepts. The consensus however did discuss an increased risk of dependency on caregivers which covertly references increased care needs and functional limitation, both concepts embedded in the definition of CMC presented in the present study.

Although these two conceptual, consensus definitions have been created, no operational definition has yet been accepted. This is likely due to several reasons including: 1) its inherent overlap with other concepts like ageing and disability, 2)the often isolated work of researchers in the field leading to numerous clinical definitions, and 3) its complex, multidimensional etiology.[301, 302] Interestingly, these reasons can also be seen as contributors to the difficulty and delay in operationalizing the definition for CMC. The spectrum of frailty and resilience is immense, like the spectrum of medical complexity in children, which makes sense considering frailty can arise from the interaction of a vast number of biopsychosocial variables/items.[284]

These definitions reflect a philosophy that informs many diagnostic instruments designed to identify and measure frailty. Independent of the components of these instruments, their performance seems to be influenced by setting and population suggesting that there are more unmeasured factors influencing a comprehensive definition of these individuals.[284, 303-305] Several instruments like the "Frailty phenotype model" describe frailty by fulfilling a set number of domains but these domains focus solely on the physiologic consequences of frailty like physical performance, weight loss, and mood and cognition.[283, 302, 306, 307] They have minimal focus on socioeconomic, diagnoses or healthcare use factors.

Contrarily, one of the most popular scales is the Frailty Index (FI).[303, 308] The FI utilizes a deficit accumulation philosophy in which the number of biosychosocial deficits reflect the degree of frailty.[309] The FI requires the Comprehensive Geriatric Assessment (CGA) to function which itself is an extensive, standardized assessment that is performed by a geriatrician.[309] The CGA measures functioning in a large number of domains including

cognition, emotion, motivation, communication, strength, mobility, balance, nutrition, ADLs, sleep, social engagement, relationships, housing, supports, caregiver stress, medical conditions and medications. As can be seen, CGA items represent a broad assessment of biopsychosocial health of an individual. The FI score represents the number of deficits recorded on the CGA. Like the philosophy of the Cohen *et al.* 2011 CMC definiton, the FI describes frailty through the presence of items across a extensive range of domains representing family and patient need (ex. caregiver stress, housing), health resource use (ex. medication use), functional limitation (ex. ADLs, mobility), and presence of medical conditions. Also, like the current study the FI has well-defined items that can measure deficits in each of these domains. Unlike the CMC model desribed in this study, there is no threshold to define frailty when using the FI. Therefore it seems the FI is a measure of frailty severity rather than a definition of frailty itself. A major limitation of the FI is that it requires the collection of an extensive amount of data. This problem must be considered in the future when using the definitional tool for CMC as it contains 39 items requiring collection.

9.4.2. NEUROIMPAIRMENT AND NEURODISABILITY

Frailty seems to describe an entity that is similar to the concept of CMC while also describing a feature seen in some CMC. Similarily the concepts of neuroimpairment and neurodisability show a similar relationship with CMC. Neuroimpairment is an extensively, and highly variably, used term in the literature and was summarized well in a 2019 scoping review by Allen *et al.*[310] This group reviewed 42 papers that utilized various definitions of neuroimpairment. They uncovered seven themes that were commonly used by researchers to describe this group of patients.

The most common theme was the concept of intellectual disability, as previously clinically defined, showing up in 80% of definitions in the literature. This was closely followed by mobility impairment (showing up in 70% of definitions) and communication impairment (30%). The remaining themes were described in a small plurality of cases including dependence on others for decision-making, a statement of chronicity, involvement of the central nervous system or the peripheral nervous system, and care needs in excess of those of typically developing children. Despite this initial, critical step by Allen *et al.* 2019, there has not yet been movement on determining which themes are critical in the definition and how to operationalize them to ensure a standardized definition. Interestingly, *Allen et al.* 2019 also predicted several problems in the progression to an international definition for neuroimpairment that have already been seen in the definition of CMC. [310] Problems may include the difficulty in defining each theme across broad health and social cultures and the disagreements when considering what themes to include.

Neurodisability is a very similar entity that was defined in 2013 by Morris *et al.*[311] Although, neurodisability has a separate definition to neuroimpairment, it likely represents a synonymous condition. Using Delphi methodology, Morris *et al.* 2013 presented a definitional statement which was altered through consensus over three rounds by a multi-disciplinary group of experts and caregivers. The consensus definitional statement was, “Neurodisability describes a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations. A specific diagnosis may not be identified. Conditions may vary over time, occur alone or in combination, and include a broad range of severity and complexity. The impact may include

difficulties with movement, cognition, hearing and vision, communication, emotion, and behaviour."[311] Although these authors chose to create a statement as an outcome, the statement shares many of the themes identified by Allen *et al.* 2019 including an insinuation to intellectual disability, a statement of chronicity, and reference to functional limitations including mobility and communication impairment. Further work to operationalize this statement has not yet been undertaken.

When compared with CMC, neuroimpairment and neurodisability are surprisingly similar. The seven themes of neuroimpairment in Allen *et al.* 2019, require measures of functional impairment, dependence on caregivers which will inevitably culminate in increased caregiver needs, and chronicity of the health state. Like the CMC definition, the neurodisability definition describes a group of chronic conditions that may be diagnosed or not, and that must be associated with funtional limitations. Unlike the CMC and neuroimpairment definition, this definitional statement lacks any reference to increased caregiver needs. Although there is no reference to increased healthcare use in either definition, the definitions as they are presented will likely associate with increased healthcare utilization whether used in the definition or not.

The similarity should not be surprising considering, as conceptually defined, children with neuroimpairment and neurodisability seem to represent pathophysiologic subcategories of CMC as defined by *Cohen et al. 2011*. [36] It is known that CMC who have a clinical diagnosis associated with neurologic impairment seem to have more detrimental outcomes and therefore seem to represent a subset of children with the highest need.[269, 273, 312]

10. CONCLUSIONS AND FUTURE WORK

We know that CMC have disproportionately high impacts on family functioning, as well as on healthcare and societal system resources. Therefore proven care models are necessary to optimally manage these children.[26-33] However, before models of care can be designed and studied, a definition of CMC is required that is "clear, reproducible, and comparable across studies."[24, 31, 34-50] Much effort has been taken to create a standardized, clinical definitional framework for these children, culminating in the framework designed by Cohen *et al.* 2011. Unfortunately, practical recognition of these children in the clinical realm remains a subjective exercise, drawing on care providers experiences and held perceptions.[36, 51] The absence of a standardized, consensus definition of CMC hampers progress in the field by limiting the generalizability of clinical research and by limiting the ability to create a validate administrative data definition.

The work presented here represents an important step in creation of an standardized clinical definition. This definition would enable generalizable research into CMC epidemiology, complex care management models, treatment for relevant presentations, and multi-centre trials. Use of this definition will expand our understanding and management of these children, as investigators and clinicians would be able to share relevant strategies for a standardized description of our shared population.

Our next planned steps in the MSc student's program of research are: follow-up survey with the respondent group, incorporation of caregiver opinion, trial in clinic populations, creation of an administrative data definition, and creation of subclassified definitions based on patterns of resource need. First, a follow-up survey will be paired with an overview of the study

findings and sent to the expert respondent group for opinions on the results. Specifically, we will ask about potential uses of the definitional tool, barriers to implementation, and general opinions about the diagnostic tool. This survey will not be a referendum on the final results of this Delphi.

Second, this proposed definitional tool will be presented to a representative group of North American CMC caregivers in a Delphi study. Caregiver and patient-centred research ensures the population under study is represented in research and is critical to identify unmet research needs, design patient-oriented outcomes, and refine the design of clinical studies. Considering a population definition is a foundation to performing research, it is likewise critical to incorporate the caregiver voice into its design.[313] The goal is to reach consensus and eventual acceptance of the tool after incorporating any proposed alterations by the caregiver group.

Third, with possible modifications from the caregiver input, the tool will be introduced to a multi-site clinical setting. This will allow the opportunity to study face validation of the definitional tool. Additionally, using the entrance critieria of multiple clinics as the reference standard and using this tool as a parallel method of defining the children, we can measure indicators of test accuracy and identify missing items. Using factor analysis, items that consistently cluster together can be collapsed into a single item thus simpilying the tool. This tool was designed to be a definitional aid but it should be noted that this tool could be also used as a diagnostic checklist or a needs assessment of CMC. After the multi-site studies, future studies may present themselves to adapt the current tool to these extrapolated uses.

Fourth, an administrative data definition of CMC will be developed to describe the epidemiology of CMC on the national scale. A refined clinical definition is needed to: a) act as a base to build the administrative definition from and b) to define a clinical population required as a comparator when determining the administrative definition's accuracy. Validation of the administrative definition will require a comparative population of CMC and non-CMC children. The 3M Clinical Risk groupings will be used to identify populations at high and low-likelihood of being CMC and these children will be screened through chart review using this tool.[96] This must be a multi-site effort using locations with robust administrative data and the resources to screen our clinical population.

Finally, the validated administrative definition will be used to understand the clustering of children within the CMC definition based on patterns of need and resource utilization. As many different combinations of items can fulfill this definition, CMC are a hetergenous group of children. It is assumed that children within the greater CMC construct will experience a variety of different outcomes and will require a variety of different resources and clinical programming. By identifying these children in population data sets, performing factor analysis, and subclassifying these children into disparate outcome-based and needs-based subcategories, we could better tailor resource utilization within the greater population of CMC.[31, 34, 35]

11. LIMITATIONS

11.1. USES OF THE DEFINITION

The objective of this study was to create a standardized, clinical definition for CMC. This was done by gathering a group of experts from heterogenous backgrounds. Consensus from a varied expert panel should identify a definition that can be used across multiple settings which may explain the inclusive nature of this definitional tool. However, depending on the specific needs or mandates of the clinician or researcher, the performance of the definitional tool will vary. For example, a child with severe cerebral palsy, requiring multiple pieces of medical technology, has had multiple admissions in the past year, and whose caregiver has signficant financial hardship will be classified the same as a child with mild autism spectrum disorder, receiving occuptional therapy, who has contact with multiple outpatient care providers, and who lives rurally. A very inclusive complex care program or research study will have a different experience with this definitional tool than a program that wishes to focus on the child most extreme end to medical complexity. However, all of the children identified by this tool will share the same core domains and represent a population of children in the upper tiers of complexity. Therefore the key unifying feature is the disproportionately higher needs these children require from family, the healthcare system and the social system. This definition will allow us to identfiy this group and understand their epidemiology, impact, and treatment modalities, however our planned future work to subclassify CMC based on a more refined description of resource need and outcomes will be helpful in resource planning.

11.2. SINGLE ARTICLE REVIEWER AND CODER

For pragmatic reasons, specifically limitations in resources and time, there were several instances in the methodology where a single coder or reviewer system was utilized when a double system may have been more methodologically robust. When creating the first iteration survey, the extensive literature review was screened for inclusion and items were extracted by a single reviewer, the MSc student (Section 7.1.2). To limit bias from any single reseracher, the framework for Scoping Reviews developed by Arksey and O'Malley suggests that inclusion and exclusion criteria for articles should be applied by two researchers and data extraction should be completed by two reserachers.[209] In systematic reviews, which have more robust methodology relative to the scoping review, this is considered good practice and has been shown to be superior to single reviewer systems.[314-316] However, a study comparing the performance of a single review to a double reviewer system concluded that substantially more mistakes were missed in the single reviewer system but that a single reviewer system is acceptable in situations of resource limitation as long as the reviewer is experienced in the field.[315] The reviewer in the current study had been a CMC physician for four years. It is acknowledged that a single review system has the risk to introduce bias and a second reviewer would lower this risk. However, in the Delphi studies, all decisions by the single reviewer are considered and "double checked" by the expert panel which would theoretically futher lower the risk of this bias.

Additionally, when coding narrative comments in Qualitative methodology it is advised to avoid single, unsupervised coders.[317] There are multiple strategies to code the narratives collected from sources such as comment boxes or interviews but they all involve other team

members to involve themselves with the process to avoid unconcious bias from a lone coder. For example, a double coder system with opportunity to discuss and resolve disagreements is often employed. Other systems include single coder systems with team review of the coding manuals or member checking of the single coders interpretation.[317] In this study a single coder of respondents' narrative comments was employed with supervisor review of the final comment themes. The coding was employed in the first iteration when respondents were asked to add or change presented items, and in every iteration when respondents rationalized their level 1 and 2 item choices. This single coder system used may increase the risk of unconcious bias during interpretation of the comments.

11.3. ATTRITION AND ACCRETION

In the current study expert repondents were allowed to come and go throughout the iterations. Respondents were not excluded due to a previous missed iteration. The majority of respondents completed all iterations and/or returned from the previous iteration, however a substantial number came and went throughout the iterations leaving the respondents group heterogenous iteration to iteration. As a result, bias from attrition or accretion may have influenced the results of this study. Arguing against the presence of bias is that there was no difference detected between new respondents, withdrawn respondents, and all respondents when examining the proportion of agreement and disagreement (table 9 and table 10). Of course comparing agreement in new repsondents to returning respondents and comparing agreement of withdrawn respondents to the respondent group in the previous iteration may be a flawed comparison. The experience of each group through the iterations is different but these comparisons may still give us an idea of the tendencies of these groups.

11.4. VOLUNTEER BIAS

As with any research, voluntary participation opens the possibility for volunteer bias. Clinical experts and research experts were engaged through unsolicited emails through four academic societies and through their contact information on publications, respectively. First, it is unclear whether members of a professional organization are a representative sample of all CMC experts in the field. Second, without access to the roster and demographics of all members of the societies it is impossible to gauge whether our sample was representative of the society membership. Without the ability to compare, one must consider theoretical reasons or evidence that the sample may differ from the general population of CMC experts.

First, financial incentives were offered for participation in this research. There is evidence that financial incentives are more appealing to individuals with lower socioeconomic status, thus introducing volunteer bias.[318] Considering the sample of experts in the current study were all employed health professionals, the influence of a $10 per iteration incentive is likely negligible. Second, the state of Ohio had disproportionately increased representation in this study (24% of American respondents) and it is unclear why there was increased interest in this region. It is unclear whether this is an accurate representation of the professional society membership, and it is unclear whether these individuals have significantly different opinions compared to the greater population. These facts will have to balanced when considering the generalizability of these results.

Interestingly, a Delphi study by its nature strives to attract the most engaged experts who can provide the time, passion, and knowledge to the research question.[120, 149, 160] Although the goal is to attract engaged experts that cover the entire spectrum of opinions on

the subject, the fact that they are the most passionate bunch out of a greater population likely introduces volunteer bias.

11.5. PHYSICIAN OVER-REPRESENTATION

Healthcare professionals that care for CMC represent a wide range of disciplines. This study had a multi-disciplinary panel of experts with 13 different professions represented. However, physicians were over-represented as 66% of the total panel were physicians. This is likely because three of the four professional societies were sub-sections within greater physician societies and the Complex Care Google group was originally created and promoted by CMC physicians. When considering the results, one must be aware that this expert panel represents a great deal of physician opinion relative to other professions who may have insight into distinct aspects of CMC lives.

11.6. OMISSION OF CAREGIVERS

It is acknowledged that caregivers should be involved in the generation of any patient-centred definition of CMC.[319] Caregivers have immense experience oriented to the needs of their child and the lived experience of their family in their specific psychosocial situation. However, it was decided to focus on healthcare provider opinions in this current study with plans to expand on this definitional tool using caregiver opinions as a next step. The greater concept of CMC is inclusive, including children with many different needs and many different psychosocial situations. Medical care providers may not have the same understanding of an individual child compared to their caregivers, but they have a better understanding of the full spectrum of medical complexity experienced by the heterogenous group of CMC. Considering

the Delphi methodology requires the respondent group be constructed with individuals of similar expertise (section 5.6.5) it was decided not to blend these differing expert groups together in a single Delphi procedure. Therefore, medical care providers were recruited as it was felt they may be in a better position to provide an overview of the needs of a broad, inclusive definition of CMC. Despite this decision, it is acknowledged that the caregiver opinion is critical and will be incorporated in future work.

12. TABLES AND FIGURES

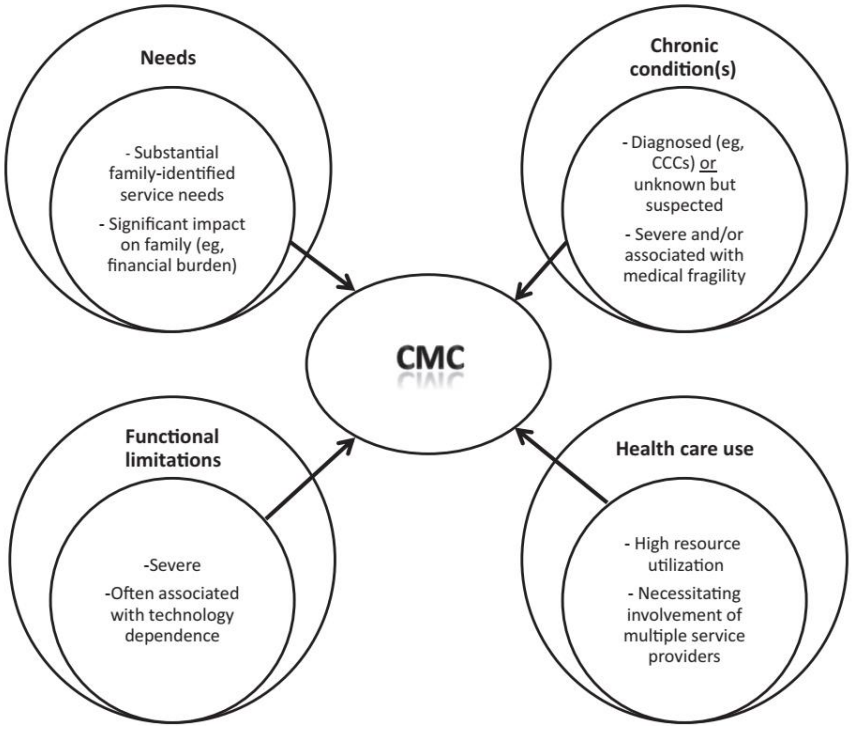

Figure 1: Conceptual definition created by the Cohen et al. 2011.[36] There are four variables that must be considered when defining a child as a CMC: Needs, Chronic condition(s), Functional limitation, and Health care use. CCCs = complex chronic conditions, CMC = children with medical complexity.

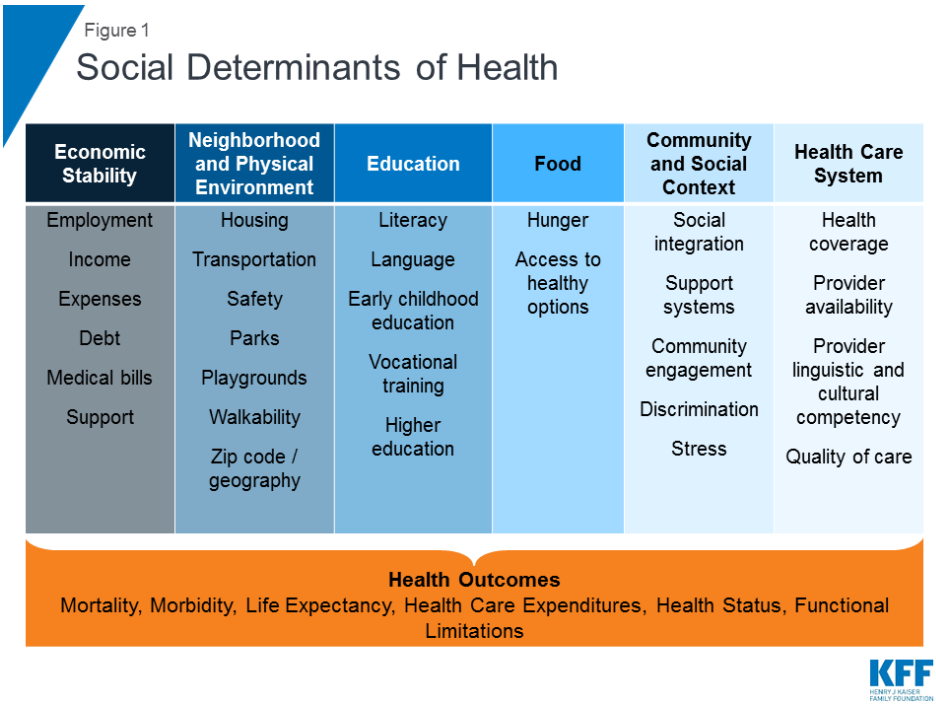

Figure 2: The model proposed by Artiga and Henton 2018 demonstrating the effect of social determinants of health on health outcomes pertinent to CMC and their caregivers.[128]

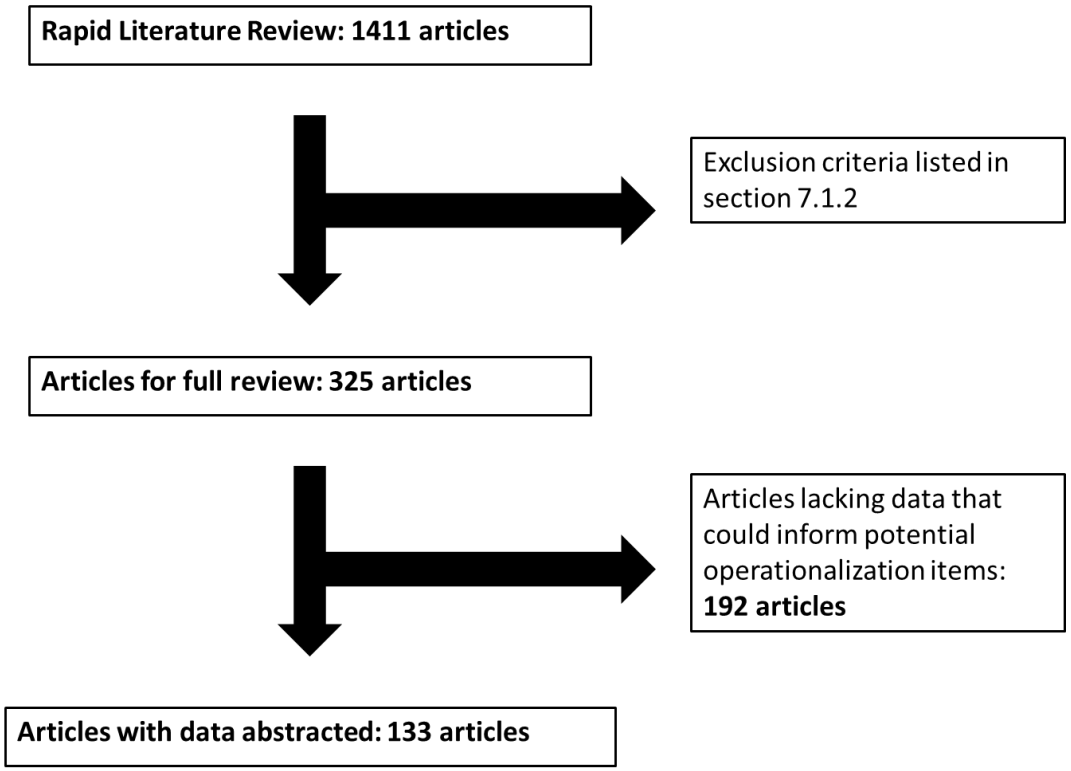

Figure 3: Steps in the article selection process starting with the number of articles from the scoping literature review and culminating in the number of articles containing variables that informed potential items for the first iteration survey.

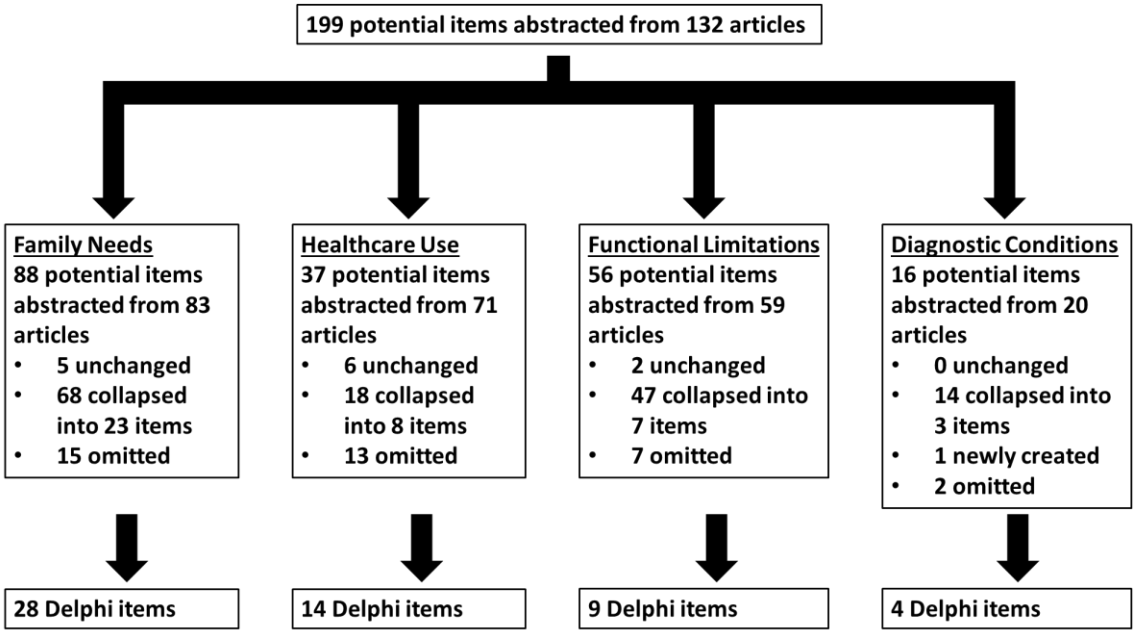

Figure 4: The steps in item creation, stratified by domain, including variable extraction, variable collation, and presentation of the potential Delphi items on the first iteration survey.

Table 1: Presentation of the level 1 items developed by the Delphi survey development committee for presentation in iteration 1, the level 1 item adjusted from respondent suggestions after iteration 1 (used in iterations 2-4), the corresponding level 2 item (iteration 2), and the adjusted level 2 item after iteration 2 (used in iterations 3-4). The level 1 and level 2 items are assigned an item code for practical ease. (FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

Item Code	First iteration Level 1 Item	Adjusted level 1 Item	Second iteration level 2 item	Adjusted level 2 item
Family Needs				
FN1	In order to care for the medical needs of their child, the caregiver(s) are experiencing an impactful amount of planned or unplanned work disruption.			
FN2	Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month.	Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month causing difficulties in providing the necessary care for the child.		
FN3	Over the past 12 months, the caregiver(s) are experiencing an impactful amount of care-related, out-of-pocket expenses.	Over the past 12 months, the caregiver(s) are incurring an impactful amount of expenses for the child's medical care beyond what is expected from raising a child without medical issues.	Over the past 12 months, an IMPACTFUL amount expenditures is defined as ≥___% of the family income. (5%-10%-15%-20%-25%)	Over the past 12 months, an IMPACTFUL amount expenditures for the child's medical care is a MINIMUM of ___% of the family income. (5%-10%-15%-20%-25%)
FN4	Currently, the child lacks medical insurance to cover basic medical needs.			
FN5	Over the past 12 months, there is an impactful amount of caregiver time spent on the direct medical care of the child.		Over the past 12 months, an IMPACTFUL amount of caregiver time spent on this level of care is defined as an average of	Over the past 12 months, an IMPACTFUL amount of caregiver time spent on this level of care is a MINIMUM average of ___ hours/week. (5-10-15-20-25)

			≥___ hours/week. (5-10-15-20-25)	
FN6	Over the past 12 months, there is an impactful amount of caregiver time spent on care related to activities of daily living (ADLs) beyond what is age-appropriate need. Note: ADLs include eating, bathing, dressing, toileting, transferring, and maintaining continence.		Over the past 12 months, an IMPACTFUL amount of caregiver time spent on this level of care is defined as an average of ≥___ hours/week.	Over the past 12 months, an IMPACTFUL amount of caregiver time spent on this level of care is a MINIMUM average of ___ hours/week.
FN7	Currently, the caregiver(s) are receiving inadequate amount of respite care at home relative to their needs. Note: This respite care may be home nursing care or non-nursing care.	Currently, the caregiver(s) are receiving inadequate amount of respite care at home relative to their self-reported needs.		
FN8	Due to the time and effort to care for the child's higher needs, the family is currently having difficulty performing their daily family activities. Note: Examples of these activities include family outings, home family time, and routine care of the other children.			
FN9	Currently, there are problems in the family relationships	Currently, there are impactful problems in the family relationships,		

	such as communication difficulties or family conflicts.	such as communication difficulties or family conflicts, that are resulting from the demands of caring for a child with medical issues.		
FN10	The child is experiencing or witnessing social disruption. Note: Social disruption may include, but not limited to: caregiver substance use, domestic violence, housing insecurity, neglect of the child, or an active child welfare case.	The child is experiencing or witnessing impactful disruption in their social life. Note: This social disruption may include, but not limited to: caregiver substance use, domestic violence, housing insecurity, food insecurity, neglect of the child, or an active child welfare case.		
FN11	Currently, the primary caregiver(s) are isolated from community or family support.	Currently, the primary caregiver(s) are isolated from community or family support which is either negatively impacting the care of the child OR is a result of the care needs of the child.		
FN12	The primary caregiver(s) have developed, or had a worsening of, a physical, psychological, or cognitive ailment since assuming the care of the child.			
FN13	Over the past school year the child is absent from an impactful amount of school, assuming the child is regularly attending school.	Over the past school year, the child is experiencing an impactful amount of unplanned school absences due complications of their medical issues, assuming the child is	Over the past school year, an IMPACTFUL amount of time absent from school is defined as an average of ≥___ days/month, assuming the child is regularly	Over the past school year, an IMPACTFUL amount of these unplanned absences from school is a MINIMUM average of ___ days/month, assuming the child is regularly attending school. (2-3-4-5-6)

		regularly attending school.	attending school. (2-3-4-5-6)	
FN14	Currently, the child is not enrolled in a primary care practice that meets the definition of a 'medical home' as defined by the American Academy of Pediatrics. Note: Place your cursor on the highlighted text to view a detailed definition.	Currently, the child is not followed by a primary care team or specialist care team that provides the care philosophy described in the 'medical home' care model. Note: Hover your cursor on the highlighted text to view a detailed definition.		
FN15	Over the past 12 months, the child has missed an impactful number of scheduled medical appointments including preventative or well child care visits.	Over the past 12 months, the child has missed an impactful number of scheduled, medically-necessary appointments including, but not limited to, preventative or well child care visits.		
FN16	Currently, the child is failing to receive recommended management for their care needs in an amount considered impactful. Note: Management may be considered medication, therapies or services.	Currently, the child is failing to receive management that is considered medically necessary and/or is part of the parent's care goals for the child. Note: Management may be considered medication, therapies or services.		
FN17	Currently, the child is experiencing complications			

	resulting from unmet care needs.			
FN18	Currently, the child is cared for by primary caregiver(s) who are not fully trained or experienced in the child's care needs.			
FN19	Over the past 12 months, the care routines for the child are frequently changing prohibiting the caregiver(s) from becoming accustomed to these routines.	Over the past 12 months, caregivers are having difficulty providing recommended care because the chronic care routines for the child are not stable and frequently changing.		
FN20	Currently, the primary caregiver(s) have suboptimal health literacy and numeracy relative to that needed to care for the child.			
FN21	The caregiver(s) are not proficient in the predominant language of their current community.	The caregiver(s) are having difficulty accessing and delivering care to the child with medical issues because they are not proficient in the predominant language of their current community.		
FN22	The primary caregiver(s) recently immigrated to their current country of residence.		The amount of time since the caregiver immigrated that is considered RECENT is ≥___ years. (1-2-3-4-5)	The amount of time since the caregiver immigrated that is considered RECENT is a less than ___ years. (1-2-3-4-5)
FN23	The current primary caregiver of the child is a single parent.			

FN24	Currently, the child is receiving inadequate care coordination.	Currently, the care of the child is impacted by inadequate care coordination.		
FN25	Over the past 12 months, there was an impactful amount of caregiver time spent on care coordination for the child.		Over the past 12 months, an IMPACTFUL amount of time spent on care coordination is defined as an average of ≥___ hours/week. (3-4-5-6-7)	Over the past 12 months, an IMPACTFUL amount of time spent on care coordination is a MINIMUM average of ___ hours/week. (3-4-5-6-7)
FN26	Currently, the child is at risk for rapid medical decompensation or has had frequent life-threatening events at home.			
FN27	Currently, the child lives in a remote or rural setting.	Currently, the caregiver(s) are having difficulty accessing care because the child lives in a remote or rural setting.		
FN28	Currently, the caregiver(s) lack reliable and accessible transportation for the child.	Currently, the caregiver(s) lack access to reliable, specialized transportation necessary for the child's care.		
Healthcare Use				

HU1	Over the past 12 months, healthcare services for the child are delivered in at least 3 healthcare and/or non-healthcare settings. Note: Healthcare settings may include hospitals, rehabilitation facilities or outpatient settings. Non-healthcare settings may include family homes, schools, or community centers.			
HU2	Currently, the child requires a school nurse to manage care needs for an impactful amount of time assuming the child regularly attends school.		Currently, an IMPACTFUL amount of allotted school nurse time is defined as ≥___ hours per week, assuming the child regularly attends school.	Currently, an IMPACTFUL amount of allotted school nurse time is defined a MINIMUM of ___ hours per week, assuming the child regularly attends school.
HU3	Currently, the child requires homecare personnel. Note: Examples of homecare services include a homecare nurse, respite personnel, or healthcare aide.			
HU4	Over the past 12 months, the child needed an impactful number of medical interventions at any one time. Note: These interventions can be pharmacologic (ex. prescription medications) and/or	At any one time over the past 12 months, the child needed an impactful number of interventions for chronic medical issues. Note: These interventions can be pharmacologic (ex. prescription medications) and/or	Over the past 12 months, an IMPACTFUL number of medical interventions at any one time is defined as ≥___. (2-3-4-5-6)	Over the past 12 months, an IMPACTFUL number of medical interventions at any one time is defined as a MINIMUM of ___. (2-3-4-5-6)

	non-pharmacologic (ex. medical technology, physical therapies).	non-pharmacologic (ex. medical technology).		
HU5	Over the past 12 months, the child needed an impactful number of medical interventions with COMPLEX administration at any one time. Note: These interventions can be pharmacologic and/or non-pharmacologic AND must have increased complexity of administration. For example, a complex intervention may include home total parenteral nutrition administered in a central venous line.		Over the past 12 months, an IMPACTFUL number of medical interventions with complex administration at any one time is defined as ≥___. (1-2-3-4-5)	At any one time over the past 12 months, an IMPACTFUL number of medical interventions with complex administration is a MINIMUM of ___. (1-2-3-4-5)
HU6	Over the past 12 months, the child had a substantial number of healthcare providers involved in their care. Note: Multiple providers with the same specialty from the same practice count as 1 provider.		Over the past 12 months, a SUBSTANTIAL number of involved healthcare providers is defined as ≥___. (1-2-3-4-5)	Over the past 12 months, a SUBSTANTIAL number of involved healthcare providers is a MINIMUM of ___. (1-2-3-4-5)
HU7	Over the past 12 months, the child had a substantial number of outpatient clinic visits.	Over the past 12 months, the child had a substantial number of medically-necessary outpatient clinic visits beyond what is expected for the child's age.	Over the past 12 months, a SUBSTANTIAL number of clinic visits is defined as ≥___. (3-6-9-12-15)	Over the past 12 months, a SUBSTANTIAL number of medically-necessary clinic visits beyond what is expected for the child's age is a MINIMUM of ___. (3-6-9-12-15)

HU8	Over the past 12 months, the child had a substantial number of presentations to the emergency room or urgent care center.	Over the past 12 months, the child had a substantial number of presentations to the emergency room or urgent care center for emergent or urgent medical issues.	Over the past 12 months, a SUBSTANTIAL number of presentations to the emergency room or urgent care is defined as ≥___. (2-3-4-5-6)	Over the past 12 months, a SUBSTANTIAL number of presentations to the emergency room or urgent care is a MINIMUM of ___. (2-3-4-5-6)
HU9	Over the past 12 months, the child had a substantial number of unplanned, inpatient hospitalizations, independent of the length of the hospitalization.		Over the past 12 months, a SUBSTANTIAL number of hospitalizations is defined as ≥___. (1-2-3-4-5)	Over the past 12 months, a SUBSTANTIAL number of hospitalizations is a MINIMUM of ___. (1-2-3-4-5)
HU10	Over the past 12 months, the child had a substantial number of cumulative days in hospital, independent of the number of hospitalizations.		Over the past 12 months, a SUBSTANTIAL number of days in hospital is defined as ≥___. (7-14-21-28-35)	Over the past 12 months, a SUBSTANTIAL number of days in hospital is a MINIMUM of ___. (7-14-21-28-35)
HU11	Over the past 12 months, the child had a substantial number of re-admissions to hospital. Note: Re-admissions are defined as an unplanned hospitalization within 30 days of a preceding hospitalization.		Over the past 12 months, a SUBSTANTIAL number of re-admissions to hospital is defined as ≥___. (1-2-3-4-5)	Over the past 12 months, a SUBSTANTIAL number of re-admissions to hospital is a MINIMUM of ___. (1-2-3-4-5)
HU12	Over the past 12 months, the child had a substantial number of unplanned hospitalizations to the intensive care unit (ICU),		Over the past 12 month, a SUBSTANTIAL number of hospitalizations to the ICU is defined as ≥___. (1-2-3-4-5)	Over the past 12 month, a SUBSTANTIAL number of hospitalizations to the ICU is a MINIMUM of ___. (1-2-3-4-5)

	independent of the length of stay on this unit.			
HU13	Over the past 12 months, the child had a substantial number of cumulative days in the ICU, independent of the number of ICU admissions.		Over the past 12 months, a SUBSTANTIAL number of days in the ICU is defined as ≥___. (3-6-9-12-15)	Over the past 12 months, a SUBSTANTIAL number of days in the ICU is a MINIMUM of ___. (3-6-9-12-15)
HU14	The child experienced an ICU re-admission within 30 days of a previous ICU admission.			
Functional Limitation				
FL1	Over the past 12 months, the child required an impactful amount of prescribed specialized therapy. Note: This therapy may be delivered by occupational therapy, physiotherapy, or speech language pathology.	Over the past 12 months, the child required an impactful amount of prescribed, specialized therapy. Note: This therapy may be delivered by, but not limited to, occupational therapy, physiotherapy, psychology, speech language pathology or by caregivers with consultation from therapists.	Over the past 12 months, an IMPACTFUL amount of prescribed specialized therapy is defined as an average of ≥___ hours per week. (1-2-3-4-5)	Over the past 12 months, an IMPACTFUL amount of prescribed specialized therapy is a MINIMUM average of ___ hours per week. (1-2-3-4-5)
FL2	Currently, the child has impactful limitations in performance of activities of daily living (ADLs). Note: ADLs include eating, bathing, dressing, toileting, transferring, and			

	maintaining continence.			
FL3	Currently, the child has impactful limitations in vital body structures or functions (VBSFs). Note: Examples of VBSFs include the maintenance of breathing, airway patency, blood circulation, digestion, waste excretion, etc.			
FL4	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. (Ex: devices supporting respiration, feeding, or elimination.)AND Misuse or discontinuation would result in IMMEDIATE, significant impact on physiologic function. (Ex: failure of a ventilator providing full mechanical ventilatory support would result in immediate respiratory failure.)			

FL5	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. (Ex: devices supporting respiration, feeding, or elimination.)AND Misuse or discontinuation would result in significant impact on physiologic function that is NOT IMMEDIATE but would occur within 1 WEEK. (Ex: failure of nocturnal, non-invasive ventilation device for nocturnal hypoventilation may cause the child to develop progressive CO2 retention and lethargy after several days.)			
FL6	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function essential to survival. (Ex: devices supporting respiration, feeding, or elimination.)AND Misuse or discontinuation would result in significant impact on physiologic function that would occur after 1 WEEK. (Ex: failure of a			

	gastrostomy tube used for nutritional support may result in nutritional deprivation after 1 week or more.)			
FL7	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. (Ex: mobility device, transfer device, or specialized seating.) AND Misuse or discontinuation would make it IMPOSSIBLE to perform the activity or participate in the life situations. (Ex: Mobility may be impossible for a child fully dependent on a motorized wheelchair should it fail.)	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. (Ex: mobility device, transfer device, specialized seating, sensory augmentation devices, communication augmentation devices, etc.) AND Misuse or discontinuation would make it IMPOSSIBLE to perform the activity or participate in the life situations. (Ex: Mobility may be impossible for a child fully dependent on a motorized wheelchair should it fail.)		

FL8	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. (Ex: mobility device, transfer device, or specialized seating.) AND Misuse or discontinuation would SEVERELY LIMIT the performance of the activity or participation in the life situations. (Ex: Mobility may be severely limited for a child aided by a walker should it fail.)	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age. (Ex: mobility device, transfer device, specialized seating, sensory augmentation devices, communication augmentation devices, etc.) AND Misuse or discontinuation would SEVERELY LIMIT the performance of the activity or participation in the life situations. (Ex: Mobility may be severely limited for a child aided by a walker should it fail.)		

FL9	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age.(Ex: mobility device, transfer device, or specialized seating.)AND Misuse or discontinuation would PARTIALLY LIMIT the performance of the activity or participation in the life situations. (Ex: Mobility may be partially limited by a child aided by ankle-foot orthotics should it fail.)	The child requires a piece of technology that supports or compensates for a limitation in a physiologic function NOT essential for survival but necessary for other 'life activities and participation' appropriate for chronologic age.(Ex: mobility device, transfer device, specialized seating, sensory augmentation devices, communication augmentation devices, etc.)AND Misuse or discontinuation would PARTIALLY LIMIT the performance of the activity or participation in the life situations. (Ex: Mobility may be partially limited for a child aided by ankle-foot orthotics should they fail.)		
Diagnostic Conditions				
DC1	The child does not need a medical diagnosis in order to be considered a child with medical complexity as long as the other 3 domains (healthcare use, functional limitations, and family needs) are fulfilled.	As long as the other 3 domains (healthcare use, functional limitations, and family needs) are fulfilled, the DIAGNOSTIC CONDITIONS domain is not needed for a child to be considered medically complex.		

DC2	The child must possess ≥1 medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain. However, the degree of chronicity and complexity of the diagnosis does not matter.	To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed medical condition (s) OR unknown but suspected medical condition(s). However, the degree of chronicity and complexity of the condition does not matter.		The child must possess a MINIMUM of _____ diagnosed or suspected medical conditions to fulfill the DIAGNOSTIC CONDITIONS domain. However, the degree of chronicity and complexity of the condition does not matter. (1-2-3-4-5)
DC3	The child must possess ≥1 'chronic' medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain. Note: Place your cursor on the highlighted text to view a detailed definition.	To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed 'chronic' medical condition(s) OR unknown but suspected 'chronic' medical condition(s). Note: Hover your cursor on the highlighted text to view a detailed definition.		The child must possess a MINIMUM of ____ diagnosed or suspected 'chronic' medical conditions to fulfill the DIAGNOSTIC CONDITIONS domain. (1-2-3-4-5)
DC4	The child must possess ≥1 medical diagnosis to fulfill the DIAGNOSTIC CONDITIONS domain. However, the condition must be considered a 'complex' medical diagnosis. Note: Place your cursor on the highlighted text to view a detailed definition.	To fulfill the DIAGNOSTIC CONDITIONS domain, the child must possess an substantial number of diagnosed 'complex' medical condition (s) OR unknown but suspected 'complex' medical condition(s). Note: Hover your cursor on the highlighted text to view a detailed definition.		The child must possess a MINIMUM of ____ diagnosed or suspected 'complex' medical conditions to fulfill the DIAGNOSTIC CONDITIONS domain. (1-2-3-4-5)

Table 2: Percentage of respondents in each iteration who were new, returning or withdrawn. New respondents refer to respondents who did not participate in the previous iteration, returning are those that participated in the previous iteration and withdrawn are those that did not participate in the respective iteration but did participate in the previous iteration.

	Iteration 2	Iteration 3	Iteration 4
New respondents	9.4%	12.8%	16.0%
Returning respondents	71.7%	68.9%	73.5%
Withdrawn respondents	18.9%	18.3%	10.5%

Table 3: Proportion of participants who consented to participate (original participants) that completed zero, one, two, three and four surveys.

	N (%) of 82 original participants
Never completed a survey	7 (8.5%)
Completed 1 survey	12 (15%)
Completed 2 surveys	5 (6.1%)
Completed 3 surveys	15 (18%)
Completed 4 surveys	43 (52%)

Table 4: Completion rates of the Delphi surveys across the four iterations. A survey was partially completed if any question was answered on the survey. Incomplete surveys were those that were left completely blank.

	Iteration 1 (%)	Iteration 2 (%)	Iteration 3 (%)	Iteration 4 (%)
Fully complete	60 (73%)	57 (70%)	53 (65%)	57 (70%)
Partially complete	7 (8.5%)	2 (2.5%)	3 (3.7%)	0 (0%)
Incomplete	15 (18%)	22 (27%)	25 (31%)	24 (30%)
Total Participants	82	81	81	81

Table 5: Demographic description of the Delphi respondents across the four iterations.

		Iteration 1	Iteration 2	Iteration 3	Iteration 4
State/Province					
	British Columbia	3	3	2	3
	Alberta	3	3	2	3
	Ontario	3	1	3	2
	Quebec	1	2	1	2
	PEI	0	1	1	1
	California	2	1	2	2
	Colorado	1	1	1	1
	Delaware	1	1	1	1
	Florida	1	1	0	1
	Illinois	3	3	2	3
	Iowa	1	1	1	1
	Maryland	2	4	3	4
	Michigan	1	1	0	1
	Minnesota	3	3	3	3

	Missouri	1	1	1	2
	Nebraska	1	1	1	1
	New York	4	4	4	4
	North Carolina	3	3	3	3
	Ohio	12	12	10	10
	Oregon	1	1	1	1
	Pennsylvania	1	1	1	1
	South Carolina	1	1	1	1
	Tennessee	2	1	1	1
	Texas	3	1	3	2
	Virginia	1	0	0	0
	Washington	2	1	2	1
	Washington DC	1	2	1	1
	Wisconsin	1	1	1	1
	Total Canadian Response	10	10	9	11
	Total U.S.A. Response	49	46	43	46
	Total Response	59 (88%)	56 (95%)	52 (93%)	57 (100%)
Clinical experience working with Children with Medical Complexity					
	2-5 years	9	9	9	5
	5-10 years	16	14	11	15
	10-15 years	7	6	5	3
	>15 years	27	27	27	32
	Total Responses	59 (88%)	56 (95%)	52 (93%)	55 (96%)
Publications as first or last author in the past 3 years					
	0	24	31	26	30
	1-5	28	19	23	22
	6-10	3	4	2	4
	>10	2	1	1	1
	Total Responses	57 (85%)	55 (93%)	52 (93%)	57 (100%)
Profession					
	Occupational therapist	2	0	1	0
	Physician	40	37	36	39
	Physical therapist	2	2	2	2
	Registered Nurse	4	3	4	5
	Nurse Practitioner	7	8	4	8
	Social worker	1	1	1	1
	PhD	2	1	3	3
	Pharmacist	1	1	0	1
	Psychologist	0	0	1	1
	Manager	0	1	1	0
	Nursing Home Administrator	0	1	1	0
	Retired Pediatrician	0	1	0	0
	Physician and PhD researcher	1	0	0	0

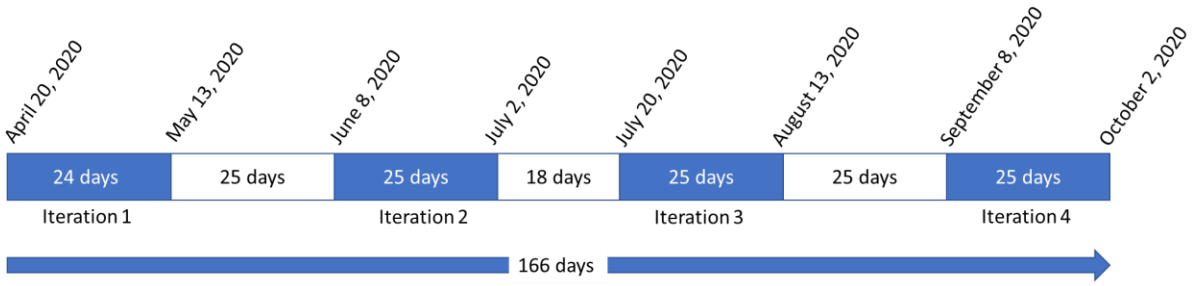

Figure 5: Figure 5 (3): Timeline to complete the four iterations of the Delphi process.

Table 6: The percentage of respondents whom agreed or disagreed with each level 1 item across the four iterations. (= Reached consensus; ~ = item wording changed after iteration 1, FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).*

	Iteration 1		**Iteration 2**		**Iteration 3**		**Iteration 4**	
	Agree	**Disagree**	**Agree**	**Disagree**	**Agree**	**Disagree**	**Agree**	**Disagree**
Family Needs								
FN1	88%*	6%						
FN2~	55%	20%	74%	16%	81%*	11%		
FN3~	78%	6%	86%*	7%				
FN4	67%	18%	72%	19%	79%	17%	88%*	7%
FN5	92%*	2%						
FN6	97%*	0%						
FN7~	71%	11%	79%	11%	85%*	4%		
FN8	92%*	5%						
FN9~	48%	22%	74%	11%	85%*	8%		
FN10~	59%	14%	73%	9%	81%*	10%		
FN11~	64%	8%	80%*	7%				
FN12	86%*	6%						
FN13~	72%	13%	89%*	7%				
FN14~	36%	34%	34%	46%	25%	60%	11%	73%
FN15~	64%	16%	77%	12%	81%*	9%		
FN16~	72%	9%	81%*	9%				
FN17	89%*	2%						
FN18	66%	20%	75%	14%	75%	10%	79%	16%
FN19~	73%	11%	84%*	5%				
FN20	67%	17%	79%	11%	89%*	4%		
FN21~	45%	25%	73%	13%	83%*	9%		
FN22	42%	22%	35%	19%	40%	15%	33%	14%
FN23	46%	30%	39%	39%	49%	34%	51%	30%
FN24~	69%	9%	88%*	5%				
FN25	88%*	3%						
FN26	91%*	5%						
FN27~	45%	23%	61%	14%	75%	11%	82%*	11%
FN28~	70%	13%	84%*	4%				

Healthcare Use								
HU1	71%	22%	71%	22%	67%	22%	77%	18%
HU2	81%*	13%						
HU3	87%*	5%						
HU4~	75%	8%	86%*	7%				
HU5	95%*	3%						
HU6	94%*	5%						
HU7~	78%	8%	91%*	2%				
HU8~	73%	15%	81%*	7%				
HU9	85%*	3%						
HU10	89%*	7%						
HU11	86%*	5%						
HU12	92%*	3%						
HU13	83%*	8%						
HU14	83%*	8%						
Functional Limitation								
FL1~	74%	13%	73%	13%	87%*	7%		
FL2	95%*	2%						
FL3	97%*	3%						
FL4	98%*	2%						
FL5	95%*	2%						
FL6	98%*	0%						
FL7	87%*	3%						
FL8	81%*	6%						
FL9~	56%	18%	79%	9%	87%*	7%		
Diagnostic Conditions								
DC1~	46%	38%	62%	33%	65%	26%	65%	30%
DC2~	25%	67%	16%	76%	7%	93%*		
DC3~	58%	37%	67%	28%	73%	20%	84%*	11%
DC4~	68%	21%	78%	16%	85%*	7%		

Table 7: Summary statistics of the proportion of respondents whom agreed or disagreed with each level 1 item across the four iterations, stratified by domain. Illustrated is: 1) the number of level 1 items reaching consensus for inclusion (Cons. Agree.), 2) the average proportion of agreement across level 1 items (Aver. Agree.), 3) the average proportion of agreement among level 1 items that reached consensus for inclusion (Aver. Cons. Agree.), 4) average change in the proportion of agreement across subsequent iterations (Aver. Agree. Chang), 5) the number of level 1 items reaching consensus for exclusion (Cons. Disagree.), 6) the average proportion of disagreement across level 1 items (Aver. Disagree.), and 7) the average proportion of disagreement among level 1 items that reached consensus for exclusion (Aver. Cons. Agree.), and 8) average change in the proportion of disagreement across subsequent iterations. (FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

Iteration	Domain	1. Cons. Agree.	2. Aver. Agree.	3. Aver. Cons. Agree.	4. Aver. Agree. Change	5. Cons. Disagree.	6. Aver. Disagree	7. Aver. Cons. Disagree.	8. Average Disagree. Change
1	Total	25	0.74	0.90	N/A	0	0.13	N/A	N/A
	FN	8	0.69	0.90	N/A	0	0.13	N/A	N/A
	HU	10	0.84	0.88	N/A	0	0.08	N/A	N/A
	FL	7	0.87	0.93	N/A	0	0.05	N/A	N/A
	DC	0	0.49	N/A	N/A	0	0.41	N/A	N/A
2	Total	10	0.71	0.85	0.10	0	0.16	N/A	-0.03
	FN	7	0.72	0.85	0.11	0	0.14	N/A	-0.03
	HU	3	0.82	0.86	0.08	0	0.10	N/A	-0.04
	FL	0	0.76	N/A	0.11	0	0.11	N/A	-0.05
	DC	0	0.56	N/A	0.07	0	0.38	N/A	-0.03
3	Total	10	0.70	0.84	0.05	1	0.19	0.93	-0.02
	FN	7	0.71	0.84	0.06	0	0.16	N/A	-0.02
	HU	0	0.67	N/A	-0.04	0	0.22	N/A	0
	FL	2	0.87	0.87	0.11	0	0.07	N/A	-0.04
	DC	1	0.58	0.85	0.02	1	0.37	0.93	-0.02
4	Total	3	0.63	0.85	0.02	0	0.23	N/A	-0.01
	FN	2	0.57	0.85	0	0	0.25	N/A	-0.01
	HU	0	0.77	N/A	0.1	0	0.18	N/A	-0.04
	FL	0	N/A	N/A	N/A	0	N/A	N/A	N/A
	DC	1	0.75	0.84	0.06	0	0.21	N/A	-0.03

Table 8: The median of level 2 item answers in the first iteration (Iter.) and the percentage of respondents who selected an integer in the level 1item Likert scale, across the final 3 iterations. The bold text in quotation marks represents the labels of the Likert value for the corresponding level 2 item. (= Reached consensus; ^ = reached consensus as determined by the final level 2 item rules, ~ = item wording changed after iteration 1; !=a item was excluded, FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition, IQR=interquartile range).*

	Iter.	**Iter. 1 median (IQR)**	**Likert 1 label**	**Likert 2 label**	**Likert 3 label**	**Likert 4 label**	**Likert 5 label**	**"Unsure" label**
Family Needs								
FN3~		10 (5-20)	**"5%"**	**"10%"**	**"15%"**	**"20%"**	**"25%"**	**"Unsure"**
	Iter 2		15%	17%	7%	15%	6%	40%
	Iter 3		13%	28%	19%	13%	2%	42%
	Iter 4		12%	28%	5%	5%	0%	49%
FN5~		15.5 (10-25)	**"5"**	**"10"**	**"15"**	**"20"**	**"25"**	**"Unsure"**
	Iter 2		16%	29%	11%	18%	11%	15%
	Iter 3		6%	60%	10%	10%	4%	12%
	Iter 4		9%	68%^	11%	4%	0%	9%
FN6~		18 (9.5-25.75)	**"5"**	**"10"**	**"15"**	**"20"**	**"25"**	**"Unsure"**
	Iter 2		13%	27%	15%	24%	9%	13%
	Iter 3		8%	46%	23%	13%	2%	8%
	Iter 4		11%	65%^	14%	2%	0%	9%
FN13~		4 (3-6)	**"2"**	**"3"**	**"4"**	**"5"**	**"6"**	**"Unsure"**
	Iter 2		23%	8%	17%	23%	4%	25%
	Iter 3		0%	20%	6%	22%	29%	24%
	Iter 4		0%	23%	5%	25%	33%^	14%
FN22~		2 (1-3)	**"1"**	**"2"**	**"3"**	**"4"**	**"5"**	**"Unsure"**
	Iter 2		13%	27%	10%	0%	6%	44%
	Iter 3		12%	39%	39%	20%	20%	41%
	Iter 4		9%	49%^	18%	18%	18%	37%
FN25~		5 (3-8)	**"3"**	**"4"**	**"5"**	**"6"**	**"7"**	**"Unsure"**
	Iter 2		34%	31%	13%	4%	7%	13%
	Iter 3		0%	52%	27%	12%	4%	6%
	Iter 4		0%	0%	68%^	23%	5%	4%
Healthcare Use								
HU2~		5 (3-10)	**"1"**	**"3"**	**"5"**	**"7"**	**"9"**	**"Unsure"**
	Iter 2		7%	33%	21%	9%	5%	26%
	Iter 3		4%	40%	25%	7%	2%	22%
	Iter 4		2%	0%	54%^	0%	25%	19%
HU4~		3 (3-5)	**"2"**	**"3"**	**"4"**	**"5"**	**"6"**	**"Unsure"**
	Iter 2		11%	18%	16%	7%	7%	40%
	Iter 3		9%	25%	15%	5%	2%	44%
	Iter 4		0%	7%	47%^	4%	2%	40%
HU5~		2 (1-3)	**"1"**	**"2"**	**"3"**	**"4"**	**"5"**	**"Unsure"**
	Iter 2		32%	18%	16%	4%	11%	20%
	Iter 3		44%	25%	4%	4%	5%	18%

	Iter 4		65%^	23%	2%	0%	2%	9%
HU6~		3 (3-4)	**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		2%	2%	59%	14%	10%	14%
	Iter 3		0%	4%	71% *	15%	4%	7%
	Iter 4							
HU7~		6 (5.75-12)	**“3”**	**“6”**	**“9”**	**“12”**	**“15”**	**“Unsure”**
	Iter 2		20%	29%	22%	13%	2%	15%
	Iter 3		11%	53%	16%	5%	2%	13%
	Iter 4		13%	74% *	7%	2%	2%	4%
HU8~		4 (2-5)	**“2”**	**“3”**	**“4”**	**“5”**	**“6”**	**“Unsure”**
	Iter 2		14%	42%	14%	7%	9%	14%
	Iter 3		11%	64%	6%	2%	6%	11%
	Iter 4		16%	75% *	5%	0%	0%	4%
HU9~		2 (2-3)	**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		10%	40%	33%	7%	2%	9%
	Iter 3		7%	60%	20%	5%	0%	7%
	Iter 4		7%	72% *	12%	4%	0%	5%
HU10~		14 (6.75-21)	**“7”**	**“14”**	**“21”**	**“28”**	**“35”**	**“Unsure”**
	Iter 2		34%	34%	10%	10%	2%	9%
	Iter 3		44%	39%	6%	4%	0%	7%
	Iter 4		61%^	30%	2%	2%	0%	5%
HU11~		2 (1-2)	**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		33%	44%	14%	4%	0%	5%
	Iter 3		32%	48%	13%	0%	0%	7%
	Iter 4		33%	58%^	4%	0%	0%	5%
HU12~		1 (1-2)	**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		47%	33%	10%	0%	0%	10%
	Iter 3		62%	25%	4%	0%	0%	9%
	Iter 4		68%^	23%	0%	2%	0%	7%
HU13~		6 (3-10)	**“3”**	**“6”**	**“9”**	**“12”**	**“15”**	**“Unsure”**
	Iter 2		28%	38%	9%	5%	3%	17%
	Iter 3		26%	54%	7%	2%	2%	9%
	Iter 4		25%	65%^	4%	0%	2%	5%
Functional Limitation								
FL1~		3 (2-5)	**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		10%	27%	37%	6%	6%	15%
	Iter 3		2%	31%	46%	9%	2%	9%
	Iter 4		0%	28%	61%^	7%	2%	2%
Diagnostic Conditions								
DC2~!			**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		29%	13%	21%	2%	2%	33%
	Iter 3		35%	7%	24%	2%	4%	28%
	Iter 4							
DC3~			**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		42%	8%	31%	0%	0%	19%

	Iter 3		44%	15%	28%	2%	0%	11%
	Iter 4		52%^	20%	21%	0%	0%	7%
DC4~			**“1”**	**“2”**	**“3”**	**“4”**	**“5”**	**“Unsure”**
	Iter 2		59%	6%	22%	0%	0%	14%
	Iter 3		74% *	9%	11%	0%	0%	6%
	Iter 4							

Table 9: Examination of the dynamic nature of agreement from a previous (prev.) iteration (It.) to a current iteration. Table reports, within an iteration and for each level 1 item: 1) the iteration, 2) number of new respondents since the previous iteration ("New"), 3) number of respondents that returned from the previous iteration (Ret.), 4) number of respondents that withdrew since the previous iteration (Withdr.), 5) the overall proportion of agreement (Ov. Agree.), 6) the proportion of agreement from new respondents (New agree.), 7) the chi square statistic (chi sq.), associated p-value (p-val), and associate p-value adjusted by the Sime's False Discover Rate method (Adj. p-val.) comparing new respondent agreement proportion to overall agreement proportion, 8) the overall proportion of agreement from the previous iteration (Ov. Agree. Prev.), 9) the proportion of agreement from withdrawn respondents in the previous iteration (with. Agree.), and 10) the chi square statistic (chi sq.), associated p-value (p-val), and associate p-value adjusted by the Sime's False Discover Rate method (Adj. p-val.) comparing withdrawn respondent agreement proportion to overall agreement proportion in the previous iteration. (~ = level 1 item wording changed after iteration 1, FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

		Respondents			Agreement									
	1. Iter.	2. New.	3. Ret.	4. Withdr.	5. Ov. Agree.	6. New agree.	7. Chi sq. (df=1)	7. p-val.	7. Adj. p-val	8. Ov. Agree. Prev.	9. With. agree.	10. Chi sq. (df=1)	10. P-val.	10. Adj p-val.
FN2~	2	5	52	14	74%	60%	0.509	0.40	0.82	55%	50%	0.141	0.71	0.93
	3	8	45	12	81%	75%	0.187	0.67	0.87	74%	67%	0.306	0.58	0.93
FN3~	2	7	49	15	85%	71%	1.076	0.30	0.82	78%	80%	0.035	0.85	0.93
FN4	2	5	52	14	72%	40%	2.540	0.11	0.82	67%	50%	1.830	0.18	0.82
	3	8	45	12	77%	63%	0.885	0.35	0.82	72%	50%	2.881	0.09	0.82
	4	10	47	6	88%	80%	0.606	0.44	0.82	77%	67%	0.339	0.56	0.93
FN7	2	7	50	12	79%	43%	5.468	0.02	0.56	71%	67%	0.093	0.76	0.93
	3	8	45	12	85%	88%	0.056	0.81	0.88	79%	67%	1.042	0.31	0.93
FN9~	2	6	51	13	74%	67%	0.153	0.70	0.87	48%	23%	3.255	0.07	0.82
	3	8	44	13	83%	88%	0.142	0.71	0.87	74%	77%	0.061	0.81	0.93
FN10~	2	6	50	14	73%	83%	0.304	0.58	0.86	59%	57%	0.023	0.88	0.93
	3	8	44	12	81%	63%	1.684	0.19	0.82	73%	67%	0.219	0.64	0.93
FN11~	2	5	51	13	80%	60%	1.250	0.26	0.82	64%	54%	0.564	0.45	0.93
FN13~	2	6	51	13	89%	83%	0.221	0.64	0.86	72%	69%	0.058	0.81	0.93
FN14~	2	5	51	13	34%	20%	0.437	0.51	0.83	36%	54%	1.828	0.18	0.82
	3	9	43	13	25%	11%	0.941	0.33	0.82	34%	15%	2.091	0.15	0.82
	4	11	45	7	11%	0%	1.360	0.24	0.82	25%	29%	0.060	0.81	0.93
FN15~	2	6	51	13	77%	67%	0.339	0.56	0.86	64%	62%	0.023	0.88	0.93
	3	8	45	12	81%	63%	1.684	0.19	0.82	77%	58%	2.446	0.12	0.82
FN16~	2	6	51	13	81%	50%	3.747	0.05	0.68	72%	69%	0.058	0.81	0.93
FN18	2	6	51	13	75%	50%	2.000	0.16	0.82	66%	62%	0.093	0.76	0.93

	3	8	44	13	73%	75%	0.016	0.90	0.92	75%	69%	0.250	0.62	0.93
	4	11	46	6	79%	73%	0.239	0.63	0.86	73%	83%	0.304	0.58	0.93
FN19~	2	6	51	13	84%	67%	1.290	0.26	0.82	73%	77%	0.106	0.75	0.93
FN20	2	6	50	14	79%	50%	3.042	0.08	0.68	67%	71%	0.101	0.75	0.93
	3	9	44	12	87%	78%	0.645	0.42	0.82	79%	67%	1.042	0.31	0.93
FN21~	2	6	49	15	73%	50%	1.610	0.20	0.82	45%	47%	0.024	0.88	0.93
	3	10	43	12	81%	80%	0.006	0.94	0.94	73%	33%	9.741	<0.01	0.12
FN22	2	6	51	13	35%	17%	0.855	0.36	0.82	42%	46%	0.085	0.77	0.93
	3	8	44	13	38%	50%	0.489	0.48	0.82	35%	31%	0.091	0.76	0.93
	4	11	46	6	33%	27%	0.179	0.67	0.86	38%	67%	2.142	0.14	0.82
FN23	2	7	47	16	39%	29%	0.294	0.59	0.86	46%	50%	0.103	0.75	0.93
	3	10	43	11	47%	60%	0.678	0.41	0.82	39%	27%	0.666	0.42	0.93
	4	10	47	6	51%	40%	0.484	0.49	0.82	47%	67%	0.963	0.33	0.93
FN24~	2	6	51	13	88%	100%	0.818	0.37	0.82	69%	54%	1.367	0.24	0.93
FN27~	2	6	51	13	61%	67%	0.091	0.76	0.88	45%	54%	0.425	0.51	0.93
	3	8	45	12	74%	63%	0.503	0.48	0.82	61%	42%	1.821	0.18	0.82
	4	10	47	6	82%	80%	0.027	0.87	0.92	74%	83%	0.253	0.62	0.93
FN28~	2	6	51	13	84%	67%	1.290	0.26	0.82	70%	69%	0.006	0.94	0.94
HU1	2	9	50	13	71%	67%	0.070	0.79	0.88	71%	77%	0.227	0.63	0.93
	3	7	48	11	67%	71%	0.051	0.82	0.88	71%	82%	0.646	0.42	0.93
	4	9	47	8	77%	67%	0.508	0.48	0.82	67%	75%	0.232	0.63	0.93
HU4~	2	8	48	15	86%	75%	0.804	0.37	0.82	75%	80%	0.200	0.66	0.93
HU7~	2	9	49	14	91%	100%	0.890	0.35	0.82	78%	86%	0.522	0.47	0.93
HU8~	2	10	48	14	81%	50%	6.244	0.01	0.56	73%	86%	1.200	0.27	0.93
FL1~	2	7	49	12	73%	43%	3.196	0.07	0.68	74%	75%	0.006	0.94	0.94
	3	9	45	11	85%	78%	0.346	0.56	0.86	73%	45%	4.375	0.04	0.82
FL9~	2	6	51	11	79%	83%	0.058	0.81	0.88	56%	36%	1.786	0.18	0.82
	3	8	46	11	85%	88%	0.056	0.81	0.88	79%	82%	0.060	0.81	0.93
DC1~	2	7	51	12	62%	71%	0.241	0.62	0.86	46%	33%	0.816	0.37	0.93
	3	8	46	12	65%	63%	0.014	0.91	0.92	62%	67%	0.127	0.72	0.93
	4	10	47	7	65%	50%	0.989	0.32	0.82	65%	86%	1.357	0.24	0.93
DC2	2	7	51	12	16%	0%	1.333	0.25	0.82	25%	50%	4.000	0.05	0.82
	3	8	47	11	7%	0%	0.602	0.44	0.82	16%	18%	0.033	0.86	0.93

DC3	2	8	49	13	67%	63%	0.058	0.81	0.88	58%	54%	0.085	0.77	0.93
	3	9	46	11	73%	100%	3.329	0.07	0.68	67%	73%	0.179	0.67	0.93
	4	9	47	8	84%	78%	0.241	0.62	0.86	73%	88%	0.913	0.34	0.93
DC4	2	8	50	12	78%	50%	3.655	0.06	0.68	68%	67%	0.006	0.94	0.94
	3	8	46	12	85%	75%	0.627	0.43	0.82	78%	58%	2.797	0.09	0.82

Table 10: Examination of the dynamic nature of disagreement from a previous (prev.) iteration (Iter.) to a current iteration. Table reports, within an iteration and for each level 1 item: 1) the iteration, 2) number of new respondents since the previous iteration ("New"), 3) number of respondents that returned from the previous iteration (Ret.), 4) number of respondents that withdrew since the previous iteration (Withdr.), 5) the overall proportion of disagreement (Ov. DisAgr.), 6) the proportion of disagreement from new respondents (New Disagr.), 7) the chi square statistic (chi sq.), associated p-value (p-val), and associate p-value adjusted by the Sime's False Discover Rate method (Adj. p-val.) comparing new respondent disagreement proportion to overall disagreement proportion, 8) the overall proportion of disagreement from the previous iteration (Ov. DisAgr. Prev.), 9) the proportion of disagreement from withdrawn respondents in the previous iteration (With. DisAgr.), and 10) the chi square statistic (chi sq.), associated p-value (p-val), and associate p-value adjusted by the Sime's False Discover Rate method (Adj. p-val.) comparing withdrawn respondent disagreement proportion to overall disagreement proportion in the previous iteration. (~ = level 1 item wording changed after iteration 1, FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

		Respondents			Disagreement									
	1. Iter.	2. New.	3. Ret.	4. Withdr.	5. Ov. Disagr.	6. New Disagr.	7. Chi sq. (df=1)	7. p-val.	7. Adj. p-val.	8. Ov. Disagr. prev.	9. With. Disagr.	10. Chi sq. (df=1)	10. p-val.	10. Adj. p-val.
FN2~	2	5	52	14	16%	40%	2.143	0.143	0.734	20%	21%	0.009	0.925	0.974
	3	8	45	12	11%	0%	0.989	0.320	0.790	16%	17%	0.009	0.925	0.974
FN3~	2	7	49	15	7%	29%	5.204	0.023	0.271	6.3%	7%	0.012	0.911	0.974
FN4	2	5	52	14	19%	60%	5.461	0.019	0.271	18%	36%	3.073	0.080	0.793
	3	8	45	12	17%	38%	2.500	0.114	0.734	19%	42%	4.125	0.042	0.793
	4	10	47	6	7%	10%	0.138	0.710	0.790	17%	33%	1.089	0.297	0.974
FN7	2	7	50	12	11%	43%	7.322	0.007	0.177	11%	17%	0.441	0.507	0.974
	3	8	45	12	2%	0%	0.163	0.686	0.790	11%	25%	2.402	0.121	0.793
FN9~	2	6	51	13	11%	0%	0.742	0.389	0.790	22%	31%	0.614	0.433	0.974
	3	8	44	13	8%	13%	0.272	0.602	0.790	11%	15%	0.212	0.645	0.974
FN10~	2	6	50	14	9%	0%	0.593	0.441	0.790	14%	7%	0.570	0.450	0.974
	3	8	44	12	10%	25%	2.000	0.157	0.734	8.9%	17%	0.971	0.324	0.974
FN11~	2	5	51	13	7%	0%	0.376	0.540	0.790	7.8%	15%	0.937	0.333	0.974
FN13~	2	6	51	13	7%	0%	0.452	0.502	0.790	13%	15%	0.046	0.830	0.974

FN14~	2	5	51	13	46%	40%	0.072	0.788	0.830	34%	31%	0.052	0.819	0.974
	3	9	43	13	58%	67%	0.299	0.584	0.790	46%	62%	1.340	0.247	0.974
	4	11	45	7	73%	82%	0.452	0.501	0.790	60%	43%	0.843	0.359	0.974
FN15~	2	6	51	13	12%	17%	0.142	0.706	0.790	16%	15%	0.010	0.922	0.974
	3	8	45	12	9%	25%	2.501	0.114	0.734	12%	25%	1.920	0.166	0.974
FN16~	2	6	51	13	9%	17%	0.469	0.494	0.790	9.4%	15%	0.479	0.489	0.974
FN18	2	6	51	13	14%	33%	1.799	0.180	0.734	20%	15%	0.203	0.652	0.974
	3	8	44	13	10%	13%	0.080	0.777	0.830	14%	15%	0.011	0.917	0.974
	4	11	46	6	16%	27%	0.990	0.320	0.790	9.6%	0%	0.637	0.425	0.974
FN19~	2	6	51	13	5%	17%	1.819	0.177	0.734	11%	15%	0.212	0.645	0.974
FN20	2	6	50	14	11%	17%	0.221	0.639	0.790	17%	14%	0.089	0.765	0.974
	3	9	44	12	4%	11%	1.148	0.284	0.790	11%	25%	2.402	0.121	0.793
FN21~	2	6	49	15	13%	33%	2.122	0.145	0.734	25%	20%	0.200	0.655	0.974
	3	10	43	12	9%	20%	1.477	0.224	0.734	13%	25%	1.528	0.216	0.974
FN22		6	51	13	19%	67%	8.982	0.003	0.177	22%	15%	0.371	0.542	0.974
		8	44	13	15%	25%	0.627	0.428	0.790	19%	23%	0.135	0.713	0.974
		11	46	6	14%	27%	1.544	0.214	0.734	15%	17%	0.019	0.891	0.974
FN23	2	7	47	16	39%	29%	0.294	0.588	0.790	30%	25%	0.190	0.663	0.974
	3	10	43	11	34%	20%	0.873	0.350	0.790	39%	55%	1.184	0.277	0.974
	4	10	47	6	30%	10%	1.905	0.168	0.734	34%	0%	3.091	0.079	0.793
FN24~	2	6	51	13	5%	0%	0.316	0.574	0.790	9.4%	23%	2.823	0.093	0.793
FN27~	2	6	51	13	14%	17%	0.045	0.832	0.846	23%	23%	0.000	1.000	1.000
	3	8	45	12	11%	25%	1.602	0.206	0.734	14%	17%	0.090	0.765	0.974
	4	10	47	6	11%	10%	0.010	0.919	0.919	11%	0%	0.742	0.389	0.974
FN28~	2	6	51	13	4%	0%	0.250	0.617	0.790	13%	15%	0.046	0.830	0.974
HU1	2	9	50	13	22%	11%	0.635	0.426	0.790	22%	15%	0.371	0.542	0.974
	3	7	48	11	22%	29%	0.200	0.655	0.790	22%	0%	3.103	0.078	0.793
	4	9	47	8	18%	22%	0.098	0.755	0.825	22%	13%	0.378	0.539	0.974
HU4~	2	8	48	15	7%	0%	0.602	0.438	0.790	7.9%	0%	1.287	0.257	0.974
HU7~	2	9	49	14	2%	0%	0.184	0.668	0.790	7.9%	7%	0.016	0.901	0.974
HU8~	2	10	48	14	7%	10%	0.138	0.710	0.790	15%	14%	0.011	0.917	0.974
FL1~	2	7	49	12	13%	29%	1.584	0.208	0.734	13%	8%	0.265	0.607	0.974
	3	9	45	11	7%	11%	0.221	0.638	0.790	13%	45%[b]	9.959	0.002	0.118

FL9~	2	6	51	11	9%	0%	0.593	0.441	0.790	18%	27%	0.604	0.437	0.974
	3	8	46	11	7%	13%	0.442	0.506	0.790	8.8%	9%	0.001	0.981	0.998
DC1~	2	7	51	12	33%	29%	0.051	0.822	0.846	38%	42%	0.081	0.775	0.974
	3	8	46	12	26%	38%	0.599	0.439	0.790	33%	25%	0.347	0.556	0.974
	4	10	47	7	30%	40%	0.476	0.490	0.790	26%	14%	0.524	0.469	0.974
DC2	2	7	51	12	76%	86%	0.384	0.536	0.790	67%	50%	1.569	0.210	0.974
	3	8	47	11	91%	100%	0.791	0.374	0.790	76%	73%	0.054	0.816	0.974
DC3	2	8	49	13	28%	38%	0.397	0.529	0.790	37%	38%	0.006	0.940	0.974
	3	9	46	11	18%	0%	1.976	0.160	0.734	28%	27%	0.005	0.941	0.974
	4	9	47	8	11%	22%	1.112	0.292	0.790	20%	13%	0.245	0.621	0.974
DC4	2	8	50	12	16%	50%	6.881	0.009	0.177	21%	25%	0.116	0.734	0.974
	3	8	46	12	6%	0%	0.511	0.475	0.790	16%	33%	2.580	0.108	0.793

Table 11: The final definitional tool composed with the results of the Delphi procedure and collapsing of related items.

Diagnostic Conditions	
1	The child must possess a minimum of 1 diagnosed, or unknown but suspected, chronic medical condition(s).
Functional Limitations	
2	Over the past 12 months, the child required a minimum average 3 hours/week of prescribed, specialized therapy. Note: This therapy may be delivered by, but not limited to, occupational therapy, physiotherapy, psychology, speech language pathology or by caregivers with consultation from therapists.
3	Currently, the child has impactful limitations in vital body structures or functions (VBSFs) or in the performance of activities of daily living (ADLs). • Note: Examples of VBSFs include the maintenance of breathing, airway patency, blood circulation, digestion, waste excretion, etc. • Note: ADLs include eating, bathing, dressing, toileting, transferring, and maintaining continence.
Healthcare Use	
4	Currently, the child requires a school nurse to manage care needs for a minimum average of 5 hours/week, assuming the child regularly attends school.
5	Currently, the child requires homecare personnel. Note: Examples of homecare services include a homecare nurse, respite personnel, or healthcare aide.
6	At any one time over the past 12 months, the child needed a minimum of 4 interventions for chronic medical issues. Note: These interventions can be pharmacologic (ex. prescription medications) and/or non-pharmacologic (ex. medical technology).
7	Over the past 12 months, the child needed a minimum of 1 medical intervention with COMPLEX administration at any one time. Note: These interventions can be pharmacologic and/or non-pharmacologic AND must have increased complexity of administration. For example, a complex intervention may include home total parenteral nutrition administered in a central venous line.
8	Over the past 12 months, the child had a minimum of 3 healthcare providers involved in their care. Note: Multiple providers with the same specialty from the same practice count as 1 provider.
9	Over the past 12 months, the child had a minimum of 6 medically necessary outpatient clinic visits beyond what is expected for the child's age.
10	Over the past 12 months, the child had a minimum of 3 presentations to the emergency room or urgent care center for emergent or urgent medical issues.
11	Over the past 12 months, the child had a minimum of 2 unplanned, inpatient hospitalizations, independent of the length of the hospitalization.
12	Over the past 12 months, the child had a minimum of 7 cumulative days in hospital, independent of the number of hospitalizations.
13	Over the past 12 months, the child had a minimum of 2 re-admissions to hospital. Note: Re-admissions are defined as an unplanned hospitalization within 30 days of a preceding hospitalization.
14	Over the past 12 months, the child had a minimum of 1 unplanned hospitalization to the intensive care unit (ICU), independent of the length of stay on this unit.
15	Over the past 12 months, the child had a minimum of 6 cumulative days in the ICU, independent of the number of ICU admissions.
16	The child experienced an ICU re-admission within 30 days of a previous ICU admission.
Family Needs	
17	In order to care for the medical needs of their child, the caregiver(s) are experiencing an impactful amount of planned or unplanned work disruption.
18	Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month causing difficulties in providing the necessary care for the child.
19	Over the past 12 months, the caregiver(s) are incurring an impactful amount (% of the family income) of expenses for the child's medical care beyond what is expected from raising a child without medical issues.
20	Currently, the child lacks medical insurance to cover basic medical needs.

21	Over the past 12 months, there is a minimum average of 10 hours/week of caregiver time spent on the direct medical care of the child or on care related to activities of daily living (ADLs) beyond what is age-appropriate need. Note: ADLs include eating, bathing, dressing, toileting, transferring, and maintaining continence.
22	Currently, the caregiver(s) are receiving inadequate amount of respite care at home relative to their self-reported needs.
23	Due to the time and effort to care for the child's higher needs, the family is currently having difficulty performing their daily family activities. Note: Examples of these activities include family outings, home family time, and routine care of the other children.
24	Currently, there are impactful problems in the family relationships, such as communication difficulties or family conflicts, that are resulting from the demands of caring for a child with medical issues.
25	The child is experiencing or witnessing impactful disruption in their social life. Note: This social disruption may include, but not limited to: caregiver substance use, domestic violence, housing insecurity, food insecurity, neglect of the child, or an active child welfare case.
26	Currently, the primary caregiver(s) are isolated from community or family support which is either negatively impacting the care of the child OR is a result of the care needs of the child.
27	The primary caregiver(s) have developed, or had a worsening of, a physical, psychological, or cognitive ailment since assuming the care of the child.
28	Over the past school year, the child is experiencing a minimum average of 6 days/month of unplanned school absences due complications of their medical issues, assuming the child is regularly attending school.
29	Over the past 12 months, the child has missed an impactful number of scheduled, medically-necessary appointments including, but not limited to, preventative or well child care visits.
30	Currently, the child is failing to receive management that is considered medically necessary and/or is part of the parent's care goals for the child. Note: Management may be considered medication, therapies or services.
31	Over the past 12 months, caregivers are having difficulty providing recommended care because the chronic care routines for the child are not stable and frequently changing.
32	Currently, the care of the child is impacted by inadequate care coordination.
33	Over the past 12 months, there was a minimum average of 5 hours/week of caregiver time spent on care coordination for the child.
34	Currently, the child is experiencing complications resulting from unmet care needs.
35	Currently, the primary caregiver(s) have suboptimal health literacy and numeracy relative to that needed to care for the child.
36	The caregiver(s) are having difficulty accessing and delivering care to the child with medical issues because they are not proficient in the predominant language of their current community.
37	Currently, the caregiver(s) are having difficulty accessing care because the child lives in a remote or rural setting.
38	Currently, the caregiver(s) lack access to reliable, specialized transportation necessary for the child's care.
39	Currently, the child is at risk for rapid medical decompensation or has had frequent life-threatening events at home.

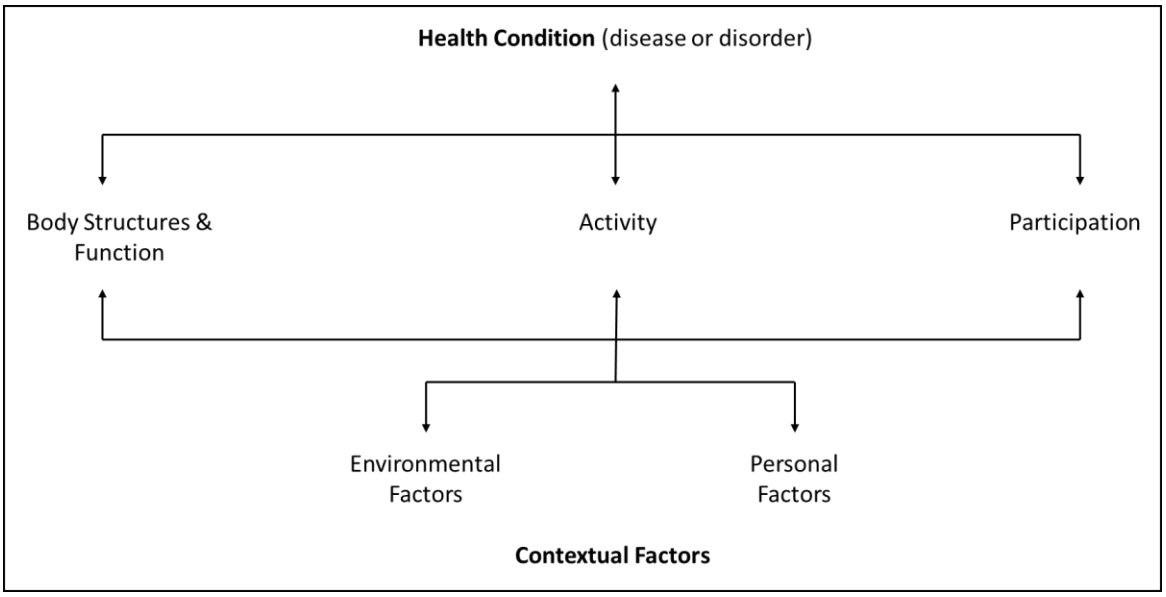

Figure 6: Characterization of the disability model that forms the basis for the World Health Organization - International Classification of Functioning

13. APPENDIX

Appendix 1: The extracted variables and subsequently constructed items from each article. Variables were extracted from the articles and were used by the Delphi survey development committee to construct potential items that were presented on the first iteration Delphi survey. (FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

Article	Variable	Item
International classification of functioning, disability, and health: children and youth version [95]	Child has impairment with limitations in function and participation as described by the World Health Organization - International Classification of Functioning.	Omitted
Technology-Dependent Children: Hospital v. Home Care – A technical memorandum (1987) [320]	Child requires medical technology that if failed would cause adverse health or hospitalization.	FL4, FL5, FL6
	Child requires home intravenous administration of nutrition and drugs.	FL4, FL5, FL6
	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires oxygen therapy at home.	FL4, FL5, FL6
	Child requires cardiorespiratory monitoring at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires renal dialysis at home.	FL4, FL5, FL6
	Child requires urinary catheterization at home.	FL4, FL5, FL6
	Child requires colostomy care at home.	FL4, FL5, FL6
The Child and Adolescent Health Measurement Initiative [321]	Child does not have a medical home.	FN14
	Child requires an educational assistant or school nurse.	HU2
	Child requires multiple, prescribed medications.	HU4 & HU5

	Number of healthcare providers involved in child's care.	HU6
	Child has individualized education plan.	Omitted
	National Survey on Children with Special Healthcare Needs asked about functional limitation - difficulty breathing, swallowing, digesting food, seeing, hearing, moving around, speaking/communicating, and taking care of themselves.	FL2
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Aboneh, E.A., *et al.* (2017) [322]	The child is not receiving prescribed medications.	FN16
	Caregiver has limited English proficiency or English is not their first language.	FN21
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Child is followed by a therapist.	FL1
Abbott, L., *et al.* (2011) [323]	Child requires an educational assistant or school nurse.	HU2
Adams, S., *et al.* (2013) [324]	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Adams, S., *et al.* (2017) [325]	Caregiver has limited English proficiency or English is not their first language.	FN21
	Caregiver is an immigrant.	FN22
	Child requires multiple, prescribed medications.	HU4 & HU5
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Afolabi, T.M., *et al.* (2019) [326]	Child requires home parenteral nutrition.	FL4, FL5, FL6
Agrawal, R., *et al.* (2014) [327]	Total hospital length of stay in a year.	HU10
Agarwal, A., *et al.* (2015) [328]	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6

Agrawal, R., *et al.* (2016) [312]	Amount of healthcare spending on the child.	Omitted
Altman, L., *et al.* (2018) [329]	Child is experiencing housing insecurity.	FN10
	Child is experiencing food insecurity.	FN10
	Caregiver has limited English proficiency or English is not their first language.	FN21
	Caregiver is an immigrant.	FN22
	Caregiver is a single parent.	FN23
	Child lives in a rural or remote location.	FN27
	Caregivers have lower educational attainment.	Omitted
	Number of healthcare providers involved in child's care.	HU6
Amin, R., *et al.* (2014) [330]	Child lives in a rural or remote location.	FN27
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Total hospital length of stay in a year.	HU10
	Total intensive care unit length of stay in a year.	HU13
An, R. (2016) [331]	Caregivers have lower income.	FN2
Ananth, P., *et al.* (2015) [332]	Hospitalization frequency.	HU9
	Total hospital length of stay in a year.	HU10
	Total intensive care unit length of stay in a year.	HU13
Arthur, K.C., *et al.* (2018) [333]	Caregivers are receiving inadequate respite.	FN7
	Child not enrolled in a primary care clinic.	FN14
	Child has not received flu shots.	FN16
	Not enrolled in a dental clinic.	FN16
	Caregivers have lower educational attainment.	Omitted
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
Aruda, M.M., *et al.* (2011) [334]	Frequency of school absences.	FN13
Avritscher, E.B.C., *et al.* (2019) [335]	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
	Frequency of intensive care unit admissions.	HU12
Balistreri, K.S. (2019) [336]	Caregivers are unemployed or underemployed.	FN1
	Caregivers have lower income.	FN2
	Child is experiencing food insecurity.	FN10
Barnert, E.S., *et al.* (2017) [337]	Caregivers are missing work due to the child's care demands or since having a child with medical complexity.	FN1

	Frequency of school absences.	FN13
	Caregiver is an immigrant.	FN22
	Child lives in a rural or remote location.	FN27
Benneyworth, B.D., *et al.* (2011) [338]	Total hospital length of stay in a year.	HU10
	Child requires a ventilator at home.	FL4, FL5, FL6
Berry, J.G., *et al.* (2011) [24]	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
	Total hospital length of stay in a year.	HU10
	Child resides in a long-term care facility.	Omitted
	Child has neurological impairment.	FL2
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Requires permanent indwelling catheters at home	FL4, FL5, FL6
	Child requires a ventriculoperitoneal shunt.	FL4, FL5, FL6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
	Child diagnosed with a medical condition affecting 3+ body systems.	DC4
	Child diagnosed with a severe single system disorder excluding asthma and psychiatric diagnoses.	DC4
Berry, J.G., *et al.* (2013) [339]	Total hospital length of stay in a single stay.	HU10
Berry, J.G., *et al.* (2014) [340]	Child requires multiple, prescribed medications.	HU4 & HU5
	Clinic visit frequency.	HU7
	Primary care clinic visit frequency.	HU7
	Subspecialty care clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Total hospital length of stay in a single stay.	HU10
	Child requires oxygen therapy at home.	FL4, FL5, FL6
	Child requires a nebulizing device at home.	FL4, FL5, FL6
	Child requires a wheelchair.	FL7, FL8, FL9

Berry, J.G., *et al.* (2015) [34]	Child does not have a medical home.	FN14
	Child requires an educational assistant or school nurse.	HU2
	Child requires home nursing care.	HU3
	Number of active medical treatments.	HU4 & HU5
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Hospitalization frequency.	HU9
	Child requires specialized diets.	Omitted
	Child requires frequent home testing or laboratory testing.	Omitted
	Frequency of access to outpatient health services.	Omitted
	Child requires frequent lab or radiological testing.	Omitted
	Child is followed by a therapist.	FL1
	Child has limitations in performing activities of daily living.	FL2
	Child has limitations with breathing, swallowing, digesting food, seeing, hearing, moving around, speaking/communicating, and taking care of themselves as described on the National Survey on Children with Special Healthcare Needs.	FL2
	Pediatric Evaluation of Disability Inventory - Computer Adaptive Test – impairments in daily activities, mobility, and social/cognitive	FL2
	Child has a cognitive limitation causing functional limitations.	FL2
	Child requires home parenteral nutrition.	FL4, FL5, FL6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child has individualized education plan.	Omitted
	Child diagnosed with a complex chronic condition as described by Feudtner, C., *et al.*, 2000.	DC4
Berry, J.G., *et al.* (2016) [341]	Child requires home nursing care.	HU3
	Child lives in a post-acute care facility.	Omitted
Berry, J.G., *et al.* (2019a) [342]	Child requires multiple, prescribed medications.	HU4 & HU5
Berry, J.G., *et al.* (2019b) [343]	Child requires home nursing care.	HU3
	Child is followed by a therapist.	FL1
	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires oxygen therapy at home.	FL4, FL5, FL6

	Child requires cardiorespiratory monitoring at home.	FL4, FL5, FL6
	Requires a mechanical in-exsufflator at home.	FL4, FL5, FL6
	Child requires a high-frequency chest oscillator at home.	FL4, FL5, FL6
Bowles, J.D, *et al.* (2016) [344]	Caregivers experiencing financial burden due to child's medical care.	FN3
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Caregivers are experiencing increased social isolation since having a child with medical complexity.	FN11
	Caregivers experienced emotional or psychological burden after having child with medical complexity.	FN12
	Child's care has not normalized for the caregivers.	FN19
Brady, P.W., *et al.* (2013) [345]	Total hospital length of stay in a single stay.	HU10
Breen, C., *et al.* (2018) [346]	No one is coordinating the child's care.	FN24
	Clinic visit frequency.	HU7
	Total hospital length of stay in a single stay.	HU10
Cady, R.G., *et al.* (2015) [347]	Child's care has not normalized for the caregivers.	FN19
Caicedo, C. (2014) [348]	Caregivers have lost employment since having a child with medical complexity.	FN1
	Caregivers are missing work due to the child's care demands or since having a child with medical complexity.	FN1
	Caregivers are unemployed or underemployed.	FN1
	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Caregivers are spending excessive time of direct care of their child with medical complexity.	FN5
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Child's family experienced negative impacts to normal daily activities since having a child with medical complexity.	FN8
	Child's family experienced negative impacts in their normal social activities.	FN8
	Child's family experienced negative impacts to family relationships since having a child with medical complexity.	FN9
	Child's family experienced increased communication difficulties since having a child with medical complexity.	FN9
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their cognitive wellbeing since having a child with medical complexity.	FN12

	Caregivers have experienced worsened physical health since having a child with medical complexity.	FN12
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Caregivers are spending excessive time on care coordination.	FN25
	Excessive time on transportation between planned or unplanned medical appointments.	FN27
	Child lives in a rural or remote location.	FN27
	Child requires home nursing care.	HU3
	Child is followed by a therapist.	FL1
Caicedo, C. (2015) [349]	Caregivers have lower income.	FN2
	Caregivers are collecting a form of financial assistance.	FN2
	Child has limitations in performing activities of daily living.	FL2
	Child has limitations in mobility.	FL2
	Child has impairment vital life functioning.	FL3
	Child requires home intravenous administration of nutrition and drugs.	FL4, FL5, FL6
	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires oxygen therapy at home.	FL4, FL5, FL6
	Child requires a nebulizing device at home.	FL4, FL5, FL6
	Child requires cardiorespiratory monitoring at home.	FL4, FL5, FL6
	Child requires a medical humidifier at home.	FL4, FL5, FL6
	Child requires a high-frequency chest oscillator at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires technology for mobility augmentation.	FL7, FL8, FL9
	Child requires a wheelchair.	FL7, FL8, FL9
	Child requires orthotic devices.	FL7, FL8, FL9
	Child requires a helmet for falls protection.	FL7, FL8, FL9
	Child requires a hospital bed at home.	FL7, FL8, FL9

	Child requires a bath chair.	FL7, FL8, FL9
	Child requires a stander.	FL7, FL8, FL9
	Child requires a mechanical lift.	FL7, FL8, FL9
	Child requires medical technology. for limitation in senses	FL7, FL8, FL9
	Child requires adapted feeding utensils.	FL7, FL8, FL9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Caicedo, C. (2016) [350]	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Primary care clinic visit frequency.	HU7
	Subspecialty care clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Child is followed by a therapist.	FL1
	Child requires technology for mobility augmentation.	FL7, FL8, FL9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Carnevale, F.A., *et al.* (2006) [351]	Child's family is distressed and not providing regular care.	FN8
Casey, P.H., *et al.* (2011) [39]	Child does not have a medical home.	FN14
	Number of healthcare providers involved in child's care.	HU6
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires a ventriculoperitoneal shunt.	FL4, FL5, FL6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3

Castro-Codesal, M.L., *et al.* (2018) [352]	Child requires a ventilator at home.	FL4, FL5, FL6
Chan, T., *et al.* (2016) [353]	Total hospital length of stay in a single stay.	HU10
	Total intensive care unit length of stay per stay.	HU13
Chien, E.T., *et al.* (2019) [354]	Child lacks preventative or well-child care.	FN14 & FN15
Cohen, E., *et al.* (2010) [355]	Child requires multiple, prescribed medications.	HU4 & HU5
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Total hospital length of stay in a year.	HU10
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Cohen, E., *et al.* (2011) [36]	Caregivers experiencing financial burden due to child's medical care.	FN3
	Caregivers are spending excessive time of direct care of their child with medical complexity.	FN5
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Caregivers need help care coordinating care.	FN24
	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Clinic visit frequency.	HU7
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child has impairment with limitations in function and participation as described by the World Health Organization - International Classification of Functioning.	Omitted
	Child diagnosed with a complex chronic condition as described by Feudtner, C., *et al.*, 2000.	DC4
Cohen, E., *et al.* (2012a) [47]	Caregivers have lower income.	FN2
	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Child requires a level of care equal to a technology-assisted child.	FN5
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Total hospital length of stay in a year.	HU10

	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
	Child has a known or suspected diagnosis of a complex chronic condition that is associated with medical fragility.	DC4
Cohen, E., *et al.* (2012b) [269]	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Primary care clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Frequency of hospital readmissions.	HU11
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Cohen, E., *et al.* (2013) [356]	Caregivers have lower income.	FN2
	Child is experiencing housing insecurity.	FN10
Coller, R.J., *et al.* (2016) [357]	Caregivers have lower income.	FN2
	Caregivers experiencing financial burden due to child's medical care.	FN3
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Caregivers are receiving inadequate respite.	FN7
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Child lacks preventative or well-child care.	FN14 & FN15
	Caregiver is a single parent.	FN23
	Caregivers need help care coordinating care.	FN24
	Caregivers have lower educational attainment.	Omitted
	Medical professionals need to communicate with multiple medical and non-medical institutions.	HU1
	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Child is followed by a therapist.	FL1
	Child has difficulty with physical, cognitive, and mental health function and these affect daily activities as described on the National Survey on Children with Special Healthcare Needs.	FL2
Coller, R.J., *et al.* (2018a) [358]	Child's immunizations are not up to date.	FN16
	Child not receiving recommended well-child care.	FN16
	Clinic visit frequency.	HU7
	Hospitalization frequency.	HU9

	Total hospital length of stay in a single stay.	HU10
Coller, R.J., *et al.* (2018b) [359]	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Condren, M., *et al.* (2019) [40]	Medication reconciliation document is not up to date.	FN16
	Caregiver has limited English proficiency or English is not their first language.	FN21
	Child requires multiple, prescribed medications.	HU4 & HU5
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
	Child diagnosed with a progressive condition associated with deteriorating health and a decreased life expectancy.	DC4
Coquillette, M., *et al.* (2015) [44]	Social worker involved in child's care.	Omitted
	Number of healthcare providers involved in child's care.	HU6
	Child diagnosed with a severe single system disorder excluding asthma and psychiatric diagnoses.	DC4
	Child diagnosed with a severe problem involving more than one body system lasting greater than 12 months.	DC4
Donohue, P.K., *et al.* (2018) [360]	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Child's care has not normalized for the caregivers.	FN19
Desai, A.D., *et al.* (2016) [361]	Child's family experienced negative impacts to normal daily activities since having a child with medical complexity.	FN8
	Caregivers are experiencing increased social isolation since having a child with medical complexity.	FN11
	Child's care has not normalized for the caregivers.	FN19
Dodds, S.D., *et al.* (2015) [362]	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Child requires a baclofen pump.	FL4, FL5, FL6
	Child requires a vagal nerve stimulator.	FL4, FL5, FL6
Dodds, S.D., *et al.* (2016) [363]	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Child requires continuous support for reaching, grasping, and activities of daily living.	FL2
	Child requires a wheelchair.	FL7, FL8, FL9
	Child cannot communicate wants or needs.	Omitted

	Child diagnosed with condition resulting in multisystem impairments.	DC4
Edwards, J.D., *et al.* (2017) [364]	Frequency of intensive care unit readmissions in 30 days.	HU14
Elias, E.R., *et al.* (2012) [365]	Caregivers lack adequate financial resources.	FN2
	Caregivers lack community or family supports and resources.	FN11
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Child does not have a medical home.	FN14
	Child experienced complications from unmet care needs.	FN17
	Caregivers are not fully trained in their child's medical care.	FN18
	Child has unstable and frequently changing medical management.	FN19
	Child lives in a rural or remote location.	FN27
	No identified primary caregiver.	Omitted
	Child requires an educational assistant or school nurse.	HU2
	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Child lives in a post-acute care facility.	Omitted
	Child requires a hospital bed at home.	FL7, FL8, FL9
	Child requires a bath chair.	FL7, FL8, FL9
	Child requires a mechanical lift.	FL7, FL8, FL9
	Child requires an adapted car seat.	FL7, FL8, FL9
Ellzey, A., *et al.* (2015) [366]	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Frequency of school absences.	FN13
	Child experienced complications from unmet care needs.	FN17
	Child has limitations in peer interaction.	Omitted
Fayed, N., *et al.* (2018) [367]	Caregivers lack adequate financial resources.	FN2
	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Child's family experienced negative impacts to normal daily activities since having a child with medical complexity.	FN8
	Child's family experienced negative impacts in their normal social activities.	FN8
	Child's family is distressed and not providing regular care.	FN8
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregivers have experienced worsened physical health since having a child with medical complexity.	FN12
	Child does not have a medical home.	FN14

	Child experienced complications from unmet care needs.	FN17
	Child has limitations in feeding.	FL2
	Levels of child and family satisfaction with medical technology.	Omitted
	Child has limitations in socioemotional functioning.	Omitted
Fortin, K., *et al.* (2016) [368]	Child is involved with a child protective service.	FN10
Foster, C.C., *et al.* (2017) [369]	Caregivers lack adequate financial resources.	FN2
	Child does not have a medical home.	FN14
	Adequacy of caregiver medical literacy.	FN20
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Child lives in a rural or remote location.	FN27
	The goals for the child's medical management are unclear.	Omitted
	Child's care frequently transitions between locations/providers.	HU1
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Friedman, S.L., *et al.* (2016) [370]	Caregivers are missing work due to the child's care demands or since having a child with medical complexity.	FN1
	Child's family experienced negative impacts to normal daily activities since having a child with medical complexity.	FN8
	Child's family experienced negative impacts in their normal social activities.	FN8
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregivers have experienced worsened physical health since having a child with medical complexity.	FN12
	Child has unmet care needs.	FN17
	Caregivers are not fully trained in their child's medical care.	FN18
Feudtner, C, *et al.* (2003) [371]	Hospitalizations frequency is increasing.	Omitted
Feudtner, C., *et al.* (2005) [372]	Child requires home nursing care.	HU3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Total hospital length of stay in a single stay.	HU10
	Child requires medical technology that if failed would cause adverse health or hospitalization.	FL4, FL5, FL6
	Child requires home intravenous administration of nutrition and drugs.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires a nebulizing device at home.	FL4, FL5, FL6

	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires a ventriculoperitoneal shunt.	FL4, FL5, FL6
Feudtner, C., *et al.* (2014) [373]	Child diagnosed with a complex chronic condition as described by Feudtner, C., *et al.*, 2000.	DC4
Gay, J.C., *et al.* (2016) [374]	Child requires home nursing care.	HU3
Geremia, C., *et al.* (2017) [375]	Child requires home intravenous administration of nutrition and drugs.	FL4, FL5, FL6
Giambra, B.K., *et al.* (2018) [376]	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
Gidengil, C., *et al.* (2017) [377]	Caregivers are not fully trained in their child's medical care.	FN18
	Caregivers need help care coordinating care.	FN24
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Caregivers have access to a needed interpreter.	Omitted
	Medical professionals need to communicate with multiple medical and non-medical institutions.	HU1
Golden, S.L., *et al.* (2012) [378]	Caregivers need help care coordinating care.	FN24
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Acting as care coordinators is overwhelming for the caregivers.	FN25
Gold, J.M., *et al.* (2016) [379]	Child lives in a rural or remote location.	FN27
Goossens, E., *et al.* (2016) [380]	Caregivers lack adequate financial resources.	FN2
	Presence of family or child illicit substance use.	FN10
	Frequency of missed medical appointments.	FN15
	Inadequate compliance with child's medications.	FN16
Gordon, J.B., *et al.* (2007) [45]	Caregiver has limited English proficiency or English is not their first language.	FN21
	Caregiver is an immigrant.	FN22
	Child's parents are divorced/separated or got divorced/separated since having a child with medical complexity.	FN23
	Child lives in a rural or remote location.	FN27
	Child's care frequently transitions between locations/providers.	HU1
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Total hospital length of stay in a year.	HU10
	Child diagnosed with a medical condition affecting 3+ body systems.	DC4

Graham, R.J., *et al.* (2018) [381]	Child lives in a rural or remote location.	FN27
Haarbauer-Krupa, J., *et al.* (2019) [382]	Caregivers have lower income.	FN2
Hessels, A.J., *et al.* (2017) [383]	Frequency of school absences.	FN13
	Child requires multiple, prescribed medications.	HU4 & HU5
	Clinic visit frequency.	HU7
	Child has limitations in performing activities of daily living.	FL2
	Child has limitations in mobility.	FL2
	Child has impairment vital life functioning.	FL3
	Child requires technology for mobility augmentation.	FL7, FL8, FL9
	Child requires orthotic devices.	FL7, FL8, FL9
	Child requires a stander.	FL7, FL8, FL9
	Child has impairment in their special senses.	Omitted
	Child has behavioural disruption.	Omitted
Hoffman, A., *et al.* (2018) [384]	Medical professionals need to communicate with multiple medical and non-medical institutions.	HU1
Hudson, S.M., *et al.* (2014) [385]	Caregivers are missing work due to the child's care demands or since having a child with medical complexity.	FN1
	Caregivers lack adequate financial resources.	FN2
	Child's family experienced negative impacts in their normal social activities.	FN8
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregiver experienced excessive worry since having a child with medical complexity.	FN12
	Caregivers experienced emotional or psychological burden after having child with medical complexity.	FN12
	Caregivers are not fully trained in their child's medical care.	FN18
	Caregiver is a single parent.	FN23
	Caregivers need help care coordinating care.	FN24
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Child at risk of rapid decompensation.	FN26
	There are deficiencies with transportation to medical appointments.	FN28
	Child has siblings.	Omitted
Johaningsmeir, S.A., *et al.* (2015) [46]	Caregivers lack adequate financial resources.	FN2
	Child lives in a rural or remote location.	FN27
	Multiple children with medical complexity in the family.	Omitted
	Amount of nursing time spent on care coordination.	Omitted

	Child has poor access to care.	Omitted
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a medical condition affecting 3+ body systems.	DC4
Joly, E. (2017) [386]	Caregivers lack community or family supports and resources.	FN11
	There are deficiencies with transportation to medical appointments.	FN28
	Child lacks access to medical resources.	Omitted
	Child requires a wheelchair.	FL7, FL8, FL9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Keilty, K., *et al.* (2018) [387]	Caregivers are unemployed or underemployed.	FN1
	Caregivers lack adequate financial resources.	FN2
	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Caregiver is an immigrant.	FN22
	Caregiver is a single parent.	FN23
	Caregivers have lower educational attainment.	Omitted
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires oxygen therapy at home.	FL4, FL5, FL6
	Child requires cardiorespiratory monitoring at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Kelly, A., *et al.* (2002) [41]	Caregivers are unemployed or underemployed.	FN1
	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Lack of financial coverage for medications.	FN4

	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Child is experiencing housing insecurity.	FN10
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Caregiver is a single parent.	FN23
	No one is coordinating the child's care.	FN24
	Caregivers need help care coordinating care.	FN24
	Caregivers are spending excessive time on care coordination.	FN25
	Acting as care coordinators is overwhelming for the caregivers.	FN25
	Child at risk of rapid decompensation.	FN26
	There are deficiencies with transportation to medical appointments.	FN28
	Child lacks access to medical resources.	Omitted
	Child's treatments are controversial or ill-defined.	Omitted
	Medical professionals need to communicate with multiple medical and non-medical institutions.	HU1
	Child requires multiple, prescribed medications.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Frequency of access to outpatient health services.	Omitted
	Child is followed by a therapist.	FL1
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child cannot communicate wants or needs.	Omitted
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
	Child diagnosed with an unstable medical condition.	Omitted
Kingsnorth, S., *et al.* (2015) [48]	Caregivers need help care coordinating care.	FN24
	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Child at risk of rapid decompensation.	FN26
	Child is at risk for unpredictable, life-threatening deterioration requiring home monitors.	FN26
	Child's medical services are delivered in 3+ settings.	HU1
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
	Child requires technology that if it failed would cause immediate risk.	FL4, FL5, FL6
	Child requires technology to support a vital life function.	FL4, FL5, FL6

	Child requires home intravenous administration of nutrition and drugs.	FL4, FL5, FL6
	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a ventilator at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
	Child requires oxygen therapy at home.	FL4, FL5, FL6
	Child requires cardiorespiratory monitoring at home.	FL4, FL5, FL6
	Child requires an enteral feeding tube at home.	FL4, FL5, FL6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a chronic condition that is severe or life-threatening, expected to last longer than 6 months, or requires the same level of care as a child with technology assistance.	DC4
Klitzner, T.S., *et al.* (2010) [277]	No one is coordinating the child's care.	FN24
	Number of healthcare providers involved in child's care.	HU6
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Urgent care presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
	Total hospital length of stay in a year.	HU10
Kun, S.S., *et al.* (2012) [388]	Frequency of hospital readmissions.	HU11
Kuo, D.Z., *et al.* (2009) [389]	Caregivers need help care coordinating care.	FN24
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child has impairment with limitations in function and participation as described by the World Health Organization - International Classification of Functioning.	Omitted
Kuo, D.Z., *et al.* (2011) [390]	Caregivers have lost employment since having a child with medical complexity.	FN1
	Caregivers require additional income for medical expenses.	FN3
	Caregivers experiencing financial burden due to child's medical care.	FN3
	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Frequency of school absences.	FN13
	Child has unstable and frequently changing medical management.	FN19

	Caregivers are spending excessive time on care coordination.	FN25
	Child enrolled in early intervention services.	Omitted
	Child enrolled in special education services.	HU2
	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
Kuo, D.Z., *et al.* (2014a) [391]	Child was not receiving necessary services to meet care goals.	FN16
Kuo, D.Z., *et al.* (2014b) [392]	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
Liberman, D.B., *et al.* (2016) [393]	The child is at risk of diminished quality of life.	Omitted
	Child diagnosed with a chronic, life-limiting condition that is, not expected to have a normal lifespan.	DC4
Lichstein, J.C., *et al.* (2018) [394]	Caregivers have lower income.	FN2
	Child is not covered or under-covered by medical insurance.	FN4
	Child does not have a medical home.	FN14
	Caregiver has limited English proficiency or English is not their first language.	FN21
	Caregivers have lower educational attainment.	Omitted
Lin, J.L., *et al.* (2018) [395]	Child requires multiple, prescribed medications.	HU4 & HU5
	Primary care clinic visit frequency.	HU7
	Subspecialty care clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Child requires more medical care than the typical child.	Omitted
	Child is followed by a therapist.	FL1
	Child has limitations in performing activities of daily living.	FL2
Looman, W.S., *et al.* (2018) [396]	Caregiver experienced negative impacts to their physical health since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their social wellbeing since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their cognitive wellbeing since having a child with medical complexity.	FN12
	Caregiver experienced excessive worry since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to healthy communication with supports since having a child with medical complexity.	Omitted
	The child is at risk of diminished quality of life.	Omitted
	Child requires medical technology to maintain their health status.	FL4, FL5, FL6

Mandic, C.G., *et al.* (2017) [397]	Caregivers have lost employment since having a child with medical complexity.	FN1
Marshall, J., *et al.* (2018) [398]	Hospitalization frequency.	HU9
	Child has impairment vital life functioning.	FL3
Matiz, L.A., *et al.* (2019) [399]	Child is experiencing housing insecurity.	FN10
	Child experiencing or witnessing domestic violence.	FN10
	Child is involved with a child protective service.	FN10
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Number of healthcare providers involved in child's care.	HU6
	Primary care clinic visit frequency.	HU7
	Subspecialty care clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Maynard, R., *et al.* (2019) [42]	Child requires an educational assistant or school nurse.	HU2
	Number of active medical treatments.	HU4 & HU5
	Number of therapeutic interventions.	HU4 & HU5
	Number of healthcare providers involved in child's care.	HU6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
McKissick, H.D., *et al.* (2017) [400]	Frequency of unplanned clinical visits per year.	Omitted
	Frequency of planned clinical visits per year.	Omitted
Meier, J.D., *et al.* (2015) [401]	Emergency room presentation frequency.	HU8
	Child requires a tracheostomy at home.	FL4, FL5, FL6
Mesman, G.R., *et al.* (2013) [402]	Caregivers have lost employment since having a child with medical complexity.	FN1
	Child is not covered or under-covered by medical insurance.	FN4
	Caregivers are receiving inadequate respite.	FN7
	Child's family experienced negative impacts to normal daily activities since having a child with medical complexity.	FN8
	Child's family experienced negative impacts to family relationships since having a child with medical complexity.	FN9

	Child's family experienced increased communication difficulties since having a child with medical complexity.	FN9
	Caregivers are experiencing increased social isolation since having a child with medical complexity.	FN11
	Caregiver experienced negative impacts to their physical health since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their social wellbeing since having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to their cognitive wellbeing since having a child with medical complexity.	FN12
	Caregiver experienced excessive worry since having a child with medical complexity.	FN12
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Caregiver experienced negative impacts to healthy communication with supports since having a child with medical complexity.	Omitted
	The child is at risk of diminished quality of life.	Omitted
Miller, A.R., *et al.* (2009) [403]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
Ming, D.Y., *et al.* (2019) [404]	Number of healthcare providers involved in child's care.	HU6
	Child requires technology that maintains daily health functioning.	FL4, FL5, FL6
Murphy, K.L., *et al.* (2012) [405]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Child lives in a rural or remote location.	FN27
Nageswaran, S., *et al.* (2016) [406]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Number of healthcare providers involved in child's care.	HU6
	Hospitalization frequency.	HU9
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Nageswaran, S., *et al.* (2017a) [407]	Caregivers experiencing increased care burden from caring for a child with medical complexity.	FN5 & FN6
	Caregivers are not fully trained in their child's medical care.	FN18
	Number of healthcare providers involved in child's care.	HU6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Nageswaran, S., *et al.* (2017b) [49]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24

	Number of healthcare providers involved in child's care.	HU6
	Undiagnosed clinical condition or unresolved clinical problem after a workup was done	Omitted
Nageswaran, S., *et al.* (2018a) [408]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Number of healthcare providers involved in child's care.	HU6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Nageswaran, S., *et al.* (2018b) [409]	Child has unstable and frequently changing medical management.	FN19
	Child experiencing frequent life-threatening events at home.	FN26
	Child requires a suction machine at home.	FL4, FL5, FL6
	Child requires a tracheostomy at home.	FL4, FL5, FL6
Nageswaran, S., *et al.* (2018c) [410]	There are deficiencies with transportation to medical appointments.	FN28
	Number of healthcare providers involved in child's care.	HU6
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
Nasir, A., *et al.* (2018) [411]	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
Nelson, B.B., *et al.* (2016) [43]	Child has unstable and frequently changing medical management.	FN19
	Child's care has not normalized for the caregivers.	FN19
	Child diagnosed with a chronic condition requiring 2+ subspecialty centres.	HU1
	Child diagnosed with a chronic condition requiring 2+ subspecialty centres.	DC4
Nkoy, F.L., *et al.* (2019) [412]	Caregiver experienced negative impacts to their emotional health since having a child with medical complexity.	FN12
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Caregivers are not fully trained in their child's medical care.	FN18
	Child has unstable and frequently changing medical management.	FN19
	Child's care has not normalized for the caregivers.	FN19
	Child experiencing frequent life-threatening events at home.	FN26
Noritz, G. *et al.* (2017) [413]	Hospitalization frequency.	HU9
	Total hospital length of stay in a single stay.	HU10
Parmeter, J., *et al.* (2018) [414]	Child is involved with a child protective service.	FN10

Peltz, A., *et al.* (2016) [415]	Child lives in a rural or remote location.	FN27
Perros, I., *et al.* (2019) [416]	Number of healthcare providers involved in child's care.	HU6
	Frequency of total hospital encounters.	Omitted
Petitgout, J.M., *et al.* (2018) [417]	Child requires a tracheostomy at home.	FL4, FL5, FL6
Petruik, C., *et al.* (2017) [418]	Child requires a ventricular assist device.	FL4, FL5, FL6
Phillips, C.D., *et al.* (2018) [419]	Frequency of access to outpatient health services.	Omitted
Pizur-Barnekow, K. (2010) [420]	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
Puls, H.T., *et al.* (2016) [421]	Child had admission for failure to thrive.	FN17
Quigley, L., *et al.* (2014) [422]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
Quinn, B.L., *et al.* (2018) [423]	Child experienced complications from unmet care needs.	FN17
Rafferty, K.S., *et al.* (2017) [424]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
Ranade-Kharker, P., *et al.* (2017) [425]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
Ralston, S.L., *et al.* (2015) [426]	Clinic visit frequency.	HU7
	Emergency room presentation frequency.	HU8
	Total hospital length of stay in a year.	HU10
Raymond, J.A. (2009) [427]	There is poor communication or collaboration between professionals and medical and non-medical institutions involved in the child's care.	FN24
	Child requires an educational assistant or school nurse.	HU2
Rehm, R.S., *et al.* (2005) [428]	Child's family experienced negative impacts in their normal social activities.	FN8
	Child has limitations in participation.	FL7, FL8, FL9
Ronis, S.D., *et al.* (2019) [429]	Child relies on caregivers for activities of daily living.	FN6
	Not enrolled in a disease specific care coordination program.	Omitted
	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a condition resulting in neurocognitive impairment and involvement of 3+ body systems.	DC4
Schrager, S.M., *et al.* (2016) [430]	Caregivers have lower income.	FN2

	Child is not covered or under-covered by medical insurance.	FN4
	Child is experiencing housing insecurity.	FN10
	Caregiver is experiencing illicit drug dependency.	FN10
	Child is involved with a child protective service.	FN10
	Caregiver physical disability	FN12
	Caregivers have pre-existing mental health concerns or developed mental health concerns after having a child with medical complexity.	FN12
	Caregiver has limited English proficiency or English is not their first language.	FN21
Sellers, D., *et al.* (2014) [431]	Child has limitations in feeding.	FL2
Simon, T.D., *et al.* (2014) [276]	Child requires medical technology.	FL4, FL5, FL6, FL7, FL8, & FL9
	Child diagnosed with a nonchronic chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC2
	Child diagnosed with a noncomplex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC3
	Child diagnosed with a complex chronic disease as defined by the Pediatric Medical Complexity Algorithm.	DC4
Walter, A.W., *et al.* (2018) [432]	Child's care results in excessive out-of-pocket expenditures for caregivers.	FN3
	Child requires multiple, prescribed medications.	HU4 & HU5
	Emergency room presentation frequency.	HU8
	Hospitalization frequency.	HU9

Appendix 2: Search strategies used to identify potential items to be used in the initial Delphi survey.

Database	Search Terms	Limits
Ovid Medline	• (Child* OR pediatric* OR paediatric* OR youth* OR infant*).ti,ab,kw AND (medical* adj5 complex*).ti,abs,kw	• Years between 1998-Septmeber 2019, inclusive • English language • Humans
Ovid Embase	• (Child* OR pediatric* OR paediatric* OR youth* OR infant*).ti,ab,kw AND (medical* adj5 complex*).ti,abs,kw	• Years between 1998-September 2018, inclusive • English language • Humans
Scopus	• TITLE-ABS-KEY (child* OR pediatric* OR paediatric* OR youth* OR infant*) AND TITLE-ABS-KEY (medical* W/5 complex*)	• Article or in-press article or review • Years between 1998-Septmeber 2018, inclusive • English language • Humans

Appendix 3: For each potential item presented in the first Delphi survey, the number of articles that contributed to the creation of that item. (FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

Item	Number of articles contributing	Item	Number of articles contributing	Item	Number of articles contributing
FN1	10	FN20	1	HU11	2
FN2	14	FN21	7	HU12	1
FN3	8	FN22	5	HU13	2
FN4	5	FN23	6	HU14	1
FN5	13	FN24	20	FL1	8
FN6	12	FN25	4	FL2	10
FN7	4	FN26	5	FL3	3
FN8	9	FN27	11	FL4	46
FN9	2	FN28	4	FL5	46
FN10	10	HU1	8	FL6	46
FN11	5	HU2	7	FL7	33
FN12	16	HU3	11	FL8	33
FN13	6	HU4	21	FL9	33
FN14	10	HU5	21	DC1	Newly created
FN15	3	HU6	27	DC2	1
FN16	6	HU7	23	DC3	7
FN17	6	HU8	19	DC4	14
FN18	6	HU9	22		
FN19	9	HU10	20		

Appendix 4: Recruitment message and consent statement that will be sent to the potential Delphi respondents

Dear member of the Pediatric Complex Care community,

Greetings. I am a clinician-scientist in Pediatrics at the University of Manitoba (Winnipeg, Manitoba, Canada) and, like you, I care for Children with Medical Complexity (CMC). The clinical definition of these children remains subjective resulting in a myriad of regional definitions for CMC. Such subjectivity and variability does not allow a consistent, generalizable definition, a characteristic crucial for broader quality and research initiatives. **I hope you will agree to participate in this research study regarding the clinical definition of CMC.**

The primary goal of this project is to engage experts in this field to reach consensus on the details of a clinical definition of CMC. Movement on a consensus definition will advance the goals of creating international standards of care, including them in generalizable clinical trials, and informing future administrative data definitions. **Your expertise is needed to guide this definition in a direction that best fits your experience and the needs of the CMC community**. You are eligible to participate in this study if you have either: (a) at least 2 years doing clinical work in the pediatric complex care field OR (b) been first or last author on at least 1 journal article in the past 3 years relating to CMC. Further details of the study are presented in the consent statement.

Consent Statement

This study is entitled, "Towards a Clinical Definition of Children with Medical Complexity: A Delphi Process of Clinicians' Opinions." A Delphi process is a consensus building technique where participants complete serial iterations of a survey. Here there will be a **maximum of 4 surveys emailed to participants, separated by several weeks, with an estimated 1-hour commitment per iteration.** Your contact information will not be linked to your survey responses and the responses will only be accessible to the immediate research team. As a token of appreciation for your participation, you will receive a **$10 Starbucks gift card** after completing each iteration.

Participation in this study is voluntary and presents no more than minimal risk. You can withdraw from participation at any time. You are not required to provide any identifiable information on the survey, however in order to describe our respondent population, it will be requested that you provide information on your postal/Zip code, years caring for CMC, authorship of research articles studying CMC, and profession. An email list will be kept in order to send out the survey links, but your name will not be linked to your email. Instead an identification number will be assigned to your email, and the identifier will be linked to your name in a separate document with proper security measures that have been approved by the Research Ethics Board of the University of Manitoba. Digital information will be stored on a password-protected computer and paper information will be stored in a locked file cabinet in a locked office. The survey system itself will not record your email address or Internet Protocol (IP) address.

By responding to this message, you are consenting to participate in this study. Thank you for your consideration.

IF YOU ARE WILLING TO PARTICIPATE IN THIS STUDY OR HAVE ADDITIONAL QUESTIONS, PLEASE CONTACT ME AT ummillak@umanitoba.mb.ca. If you have questions about your rights as a research participant, you may contact the University of Manitoba Bannatyne Research Ethics Board by phone at 204-789-3255 or by email at bannatynereb@umanitoba.ca. **Please forward this invitation to other individuals who you think might qualify and be interested.**

Sincerely,

Kyle Millar, MD, FRCPC
Complex Care Pediatrician
MSc (candidate)
Departments of Pediatrics and Community Health Sciences
University of Manitoba
Winnipeg, Manitoba, Canada

Appendix 5: A text version of the iteration 1 Delphi survey (double click for full survey).

Confidential

Family Needs (Iteration 1)

Record ID

_____

FAMILY NEEDS

INSTRUCTIONS:
You will be asked to determine whether each of the following items, considered ALONE, would satisfactorily fulfill the "FAMILY NEEDS" domain. When considering each item under "FAMILY NEEDS", please assume the other 3 domains (Healthcare Use, Functional Limitations and Diagnostic Conditions) have already been fulfilled.

This section contains items pertaining to increased service needs, impacts on the family due to these needs, and risk factors for greater impacts from the needs.

To reduce the length of the first iteration survey, the FAMILY NEEDS section was split in two and then randomized to two groups of respondents. In the second iteration survey you will likely see items not yet viewed by you. At the end of this section, please continue to suggests items you deem pertinent knowing they may be present in the other half.

A version of the general instructions is available at the end of this section.

Proportion of Responses

_____

You have completed [proportion]% of this survey

FAMILY NEEDS

In order to care for the medical needs of their child, the caregiver(s) are experiencing an impactful amount of planned or unplanned work disruption.

◯ Strongly Agree that this item fulfills FAMILY NEEDS
◯ Agree
◯ Neutral
◯ Disagree
◯ Strongly Disagree that this item fulfills FAMILY NEEDS

(Optional) Comment on the rationale for your answer, on any proposed alterations to the item, or on any other issue with this item.

_____

Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month.

◯ Strongly Agree that this item fulfills FAMILY NEEDS
◯ Agree
◯ Neutral
◯ Disagree
◯ Strongly Disagree that this item fulfills FAMILY NEEDS

(Optional) Comment on the rationale for your answer, on any proposed alterations to the item, or on any other issue with this item.

_____

Appendix 6: A text version of the iteration 2 Delphi survey (double click for full survey).

Family Needs (Iteration-2)

Record ID

_____

FAMILY NEEDS

INSTRUCTIONS:
You will be asked to determine whether each of the following items, considered ALONE, would satisfactorily fulfill the "FAMILY NEEDS" domain. When considering each item under "FAMILY NEEDS", please assume the other 3 domains (Healthcare Use, Functional Limitations and Diagnostic Conditions) have already been fulfilled.

This section contains items pertaining to increased service needs, impacts on the family due to these needs, and risk factors for greater impacts from the needs.

A version of the general instructions is available at the end of this section.

Proportion of Responses

_____

You have completed [proportion_v2]% of this survey

FAMILY NEEDS

Currently, the caregiver(s) lack the financial resources to pay necessary expenses at the end of the month causing difficulties in providing the necessary care for the child.

For your re-evaluation of this item: 1) WORDING WAS ALTERED since iteration 1. 2) Your previous response was [fam_lack_res]. 3) Combined group responses from iteration 1 are displayed below the question.

- ◯ Strongly Agree that this item fulfills FAMILY NEEDS
- ◯ Agree
- ◯ Neutral
- ◯ Disagree
- ◯ Strongly Disagree that this item fulfills FAMILY NEEDS

Appendix 7: Completion rates of the Delphi survey across level 1 items and across iterations. (FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition, iter.=iteration).

	A Items				**B Items**			
	Iter. 1 response rate (n=82)	**Iter. 2 response rate (n=81)**	**Iter. 3 response rate (n=81)**	**Iter. 4 response rate (n=81)**	**Iter. 1 response rate (n=82)**	**Iter. 2 response rate (n=81)**	**Iter. 3 response rate (n=81)**	**Iter. 4 response rate (n=81)**
Family Needs								
FN1	66 (80%)							
FN2	66 (80%)	57 (70%)	53 (65%)					
FN3	64 (78%)	56 (69%)			52 (63%)	53 (65%)	53 (65%)	57 (70%)
FN4	66 (80%)	57 (70%)	53 (65%)	57 (70%)				
FN5	65 (79%)				62 (76%)	55 (68%)	52 (64%)	57 (70%)
FN6	63 (77%)				60 (73%)	55 (68%)	52 (64%)	57 (70%)
FN7	62 (76%)	57 (70%)	53 (65%)					
FN8	63 (77%)							
FN9	64 (78%)	57 (70%)	52 (64%)					
FN10	64 (78%)	56 (69%)	52 (64%)					
FN11	64 (78%)	56 (69%)						
FN12	64 (78%)							
FN13	64 (78%)	57 (70%)			49 (60%)	52 (64%)	51 (63%)	57 (70%)
FN14	64 (78%)	56 (69%)	52 (64%)	56 (69%)				
FN15	64 (78%)	57 (70%)	53 (65%)					
FN16	64 (78%)	57 (70%)						
FN17	64 (78%)							
FN18	64 (78%)	57 (70%)	52 (64%)	57 (70%)				
FN19	64 (78%)	57 (70%)						
FN20	64 (78%)	56 (69%)	53 (65%)					
FN21	64 (78%)	55 (68%)	53 (65%)					
FN22	64 (78%)	57 (70%)	52 (64%)	57 (70%)	43 (52%)	48 (59%)	51 (63%)	57 (70%)
FN23	63 (77%)	54 (67%)	53 (65%)	57 (70%)				
FN24	64 (78%)	57 (70%)						
FN25	64 (78%)				57 (69%)	56 (69%)	52 (64%)	57 (70%)
FN26	64 (78%)							
FN27	64 (78%)	57 (70%)	53 (65%)	57 (70%)				
FN28	64 (78%)	57 (70%)						
Healthcare Use								
HU1	63 (77%)	59 (73%)	55 (68%)	56 (69%)				
HU2	63 (77%)				51 (62%)	58 (72%)	55 (68%)	57 (70%)
HU3	63 (77%)							
HU4	63 (77%)	56 (69%)			49 (60%)	55 (68%)	55 (68%)	55 (68%)
HU5	63 (77%)				56 (68%)	56 (69%)	55 (68%)	57 (70%)
HU6	63 (77%)				56 (68%)	58 (72%)	55 (68%)	
HU7	63 (77%)	58 (72%)			52 (63%)	55 (68%)	55 (68%)	57 (70%)
HU8	62 (76%)	58 (72%)			47 (57%)	57 (70%)	53 (65%)	57 (70%)
HU9	62 (76%)				55 (67%)	58 (72%)	55 (68%)	57 (70%)

HU10	61 (74%)				52 (63%)	58 (72%)	54 (67%)	57 (70%)
HU11	63 (77%)				57 (70%)	57 (70%)	54 (67%)	57 (70%)
HU12	63 (77%)				56 (68%)	58 (72%)	55 (68%)	57 (70%)
HU13	63 (77%)				53 (65%)	58 (72%)	54 (67%)	57 (70%)
HU14	63 (77%)							
Functional Limitation								
FL1	61 (74%)	56 (69%)	54 (67%)		50 (61%)	52 (64%)	54 (67%)	57 (70%)
FL2	62 (76%)							
FL3	62 (76%)							
FL4	62 (76%)							
FL5	62 (76%)							
FL6	62 (76%)							
FL7	62 (76%)							
FL8	62 (76%)							
FL9	62 (76%)	57 (70%)	54 (67%)					
Diagnostic Conditions								
DC1	63 (77%)	58 (72%)	54 (67%)	57 (70%)				
DC2	63 (77%)	58 (72%)	55 (68%)			52 (64%)	54 (67%)	
DC3	62 (76%)	57 (70%)	55 (68%)	56 (69%)		52 (64%)	54 (67%)	56 (69%)
DC4	62 (76%)	58 (72%)	54 (67%)			51 (63%)	53 (65%)	

Appendix 8: The comment response rate and comments for each item that were presented to the respondents. The comments are separated into those that argued for the inclusion of the level 1 item in the definition (labelled as affirmative), those that argued against inclusion (dissenting), and those that explained the rationale behind choosing a level 2 item value. The number of comments left (N) for each iteration and item is presented with the comment rate as a percentage (those leaving comments denominated by those that completed any of the survey). (Iter. = Iteration, FN=Family Needs, HU=Healthcare Use, FL=Functional Limitations, DC=Diagnostic Condition).

Item	Iter.	Sub-Item	N (RR)	Top Comments Presented in Subsequent Iteration
FN2	1	1-affirmative	21 (32%)	Poverty worsens child's medical outcomes.
				Item increases familial stress.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item depends on too many other factors involving the family, healthcare system, and/or social services context.
	2	1-affirmative	25 (44%)	As long as the other domains are met, this item fulfills family needs.
				Poverty could result in, or exacerbate, lack of access to care and/or other negative health outcomes.
		1-dissenting		Social determinants of health and social system difficulties should not define children with medical complexity.
				Item is not related to the care needs of children with medical complexity.
FN3	1	1-affirmative	28 (44%)	Item demonstrates the impact on caregivers of having a child with medical complexity.
				Item increases familial stress.
				Item may decrease money available for necessities.

		1-dissenting		Item depends on too many other factors involving the cost of living, healthcare system context, and/or social service context.
				Item not specifically related to the care or needs of children with medical complexity.
	2	Level 2	21 (38%)	Answer to the follow-up item depends too much on other factors including: a) the families total income or b) the amount the families spend on raising a typically developing child.
	3	Level 2	14 (26%)	Answer to the follow-up item depends too much on other factors including: a) the family's total income or b) what the family is at risk of losing due to the expenditures.
				The answer to this question should be derived from the families not from this respondent group.
FN4	1	1-affirmative	18 (27%)	Care delivery would be very difficult without medical insurance.
				Lacking medical insurance overwhelms finances.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item depends on too many other factors such as family prioritization and/or the healthcare system context.
	2	1-affirmative	17 (30%)	Lack of insurance leads to financial expense that could result in, or exacerbate, lack of access to care and/or negative health outcomes.
				As long as the other domains are met, this item fulfills family needs.
		1-dissenting		Item depends on the region and system the child is in.
				Item does not reach the threshold to fulfill family needs.
				Item not specific enough to children with medical complexity, but rather affects all children.
	3	1-affirmative	10 (19%)	Lack of insurance leads to financial expense that could result in poor access to care, negative health outcomes, or further exacerbate financial disadvantage.
		1-dissenting		Item depends on the region and system the child is in.
FN5	2	Level 2	17 (31%)	Participant rationale for follow-up answers included: a) the amount of time that would be disruptive to caregiver life and work activities, b) minimum time insurance covers for homecare services, c) average time complex children require for care, d) time beyond what is required to care for a child with special health needs, and e) time estimate that includes nighttime care.
	3	Level 2	9 (17%)	Participant rationale for follow-up answers included: a) the minimum amount of time that would be disruptive to work schedules, b) the minimum amount of time before families need help with delivering direct medical care, and c) average time per day complex children require for direct medical care.

				Answer to the follow-up item depends too much on other factors including: a) the amount of time the family spends on similar care for a neurotypical child, b) how much time the caregivers need to care for their own mental and physical health, and c) the intensity of care that must be delivered.
FN6	2	Level 2	13 (24%)	Participant rationale for follow-up answers included: a) the amount of time that would be disruptive to caregiver work activities, b) minimum time insurance covers for homecare services, and c) time beyond what is required to care for a child with special health needs.
	3	Level 2	8 (15%)	Participant rationale for follow-up answers included: a) the minimum amount of time that would be disruptive to the caregiver's self-care, employment, regular chores, and family time, b) minimum amount of time required for a complex feeding regime, and c) the minimum amount of time before families need help with delivering care for activities of daily living.
				Answer to the follow-up item depends too much on other factors including: a) the amount of time the family spends on similar care for a neurotypical child and b) how much time the caregivers need to care for their own mental and physical health.
FN7	1	1-affirmative	20 (32%)	Lack of respite care would lead to a high caregiver burden.
				Item represents a measure of medical complexity.
		1-dissenting		Item not specifically related to the care or needs of a children with medical complexity.
				Item too specific as it does not account for regional differences and/or family preferences.
				It is too hard to define "adequate respite."
	2	1-affirmative	13 (23%)	Item relates to the care needs of the child and contributes to familial burden.
		1-dissenting		Item is not specific enough as "adequate respite" is not adequately defined.
				Item does not reach the threshold to fulfill family needs.
FN9	1	1-affirmative	21 (33%)	Social troubles fulfill the FAMILY NEEDS domain.
				Family stress will hinder the child's care and development.
				The problems raise questions about child's safety.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item depends on too many other factors such as the family social context, family coping, and/or the severity of the problems.
	2	1-affirmative	10 (18%)	Item interferes with the care needs of the child and contributes to familial burden.
		1-dissenting		Item is not specific to children with medical complexity.

				Item depends on too many other familial factors to be a robust measure of family needs.
FN10	1	1-affirmative	16 (25%)	Disruptions increase complexity of condition and/or care.
				Disruptions raise questions about child's safety.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item does not impact the care of children with medical complexity.
	2	1-affirmative	14 (25%)	Item interferes with the care needs of the child and raises safety concerns.
				Item is a marker for other disruption and is worrisome for adverse outcomes.
		1-dissenting		Item is not specific to children with medical complexity.
				Item does not reach the threshold to fulfill family needs or to define children with medical complexity.
FN11	1	1-affirmative	12 (19%)	Isolation would lead to a high caregiver burden.
				Item captures family needs.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item does not impact the care of the child and/or is not due to the difficulty of caring for a children with medical complexity.
				Item depends on too many other factors such as the reason for caregiver isolation.
FN13	1	1-affirmative	22 (34%)	Item captures family needs assuming the school is missed for health reasons.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Item depends on too many other factors such as the school context, caregiver context and/or the child's condition.
	2	Level 2	16 (28%)	Participant rationale for follow-up answers included: a) the amount of time the caregiver is absent from work that an employer would forgive and b) the amount of missed time that seemed significant per week or per month.
				Answer to the follow-up item depends too much on the grade level or functional level of the child.
	3	Level 2	11 (22%)	Participant rationale for follow-up answers included: a) the amount of missed time that impacts the caregiver's ability to work, b) the percentage of missed school days that would prevent the child from keeping up to their peers, and c) the number of weeks off per month that seems substantial.
				Answer to the follow-up item depends too much on other factors including: a) the functional level of the child, b) the amount of school that is done virtually, and c) the functional level of the family.

FN14	1	1-affirmative	24 (38%)	Medical homes are important to coordinate care across providers.
		1-dissenting		Item is too specific as many primary care physicians are not in medical home models and/or care coordination is provided by other clinic models outside of primary care.
				Item not specifically related to the care or needs of children with medical complexity.
	2	1-dissenting	18 (32%)	Item is too specific as many primary care and subspecialty care practices provide good care without abiding by this care philosophy.
				Item depends on the region and system the child is in.
	3	1-dissenting	14 (27%)	Item cannot be generalized to all children with medical complexity because many care models provide good care without abiding by this care philosophy.
				Item depends on the system the child is in and the capabilities of the caregivers.
FN15	1	1-affirmative	18 (28%)	Item indicates sub-optimal management and social risk.
				Item indicates barriers to care.
				Item indicates a common problem in complex care.
		1-dissenting		Item depends on too many other factors such as the caregiver context and/or child's age.
				Item not specifically related to the care or needs of children with medical complexity.
	2	1-affirmative	9 (16%)	Item interferes with the care needs of the child and puts the child at risk for poor outcomes.
		1-dissenting		Item depends on too many other factors related to the family situation, healthcare system and child's health.
				Healthcare system difficulties should not define children with medical complexity.
FN16	1	1-affirmative	10 (16%)	Item indicates sub-optimal management and social risk.
				Item indicates poor caregiver coping.
		1-dissenting		Item not related to the medical condition of the children with medical complexity.
				Item depends on too many other factors such as the needs of the child and the preferences of the family.
FN18	1	1-affirmative	13 (20%)	Item indicates a common problem in complex care.
		1-dissenting		Depends on too many other factors involving the caregiver and healthcare provider context.
				Item not specifically related to the care or needs of children with medical complexity.
	2	1-affirmative	12 (21%)	Item interferes with the care needs of the child and puts the child at risk for poor outcomes.
		1-dissenting		Most caregivers of children with medical problems are not trained or experienced early in the child's medical course but with training this improves.
				Item is not related to the care needs of children with medical complexity.

	3	1-Clarification	6 (12%)	The "primary caregiver(s)" referred to in this item are referring to caregivers at home (ex. parents, foster parents, etc.) NOT primary care providers (ex. physicians, nurse practitioners, etc.).
FN19	1	1-affirmative	7 (11%)	Item indicates increased care burden.
				Item indicates an impact on medical management.
		1-dissenting		Item does not demonstrate impact on the child or caregivers.
FN20	1	1-affirmative	12 (19%)	Item impacts the medical management of the child.
				Item fulfills FAMILY NEEDS domain.
		1-dissenting		Item not specifically related to the care or needs of children with medical complexity.
				Depends on too many other caregiver and healthcare provider factors.
	2	1-affirmative	9 (16%)	The care needs of children with medical complexity require high level of health literacy and numeracy by the caregivers and, if lacking, puts the child at risk of poor health outcomes.
		1-dissenting		Social system difficulties and caregiver difficulties should not define children with medical complexity.
				This item depends too much on social factors such as the whether these difficulties are compensated by the caregivers' family and friends.
FN21	1	1-affirmative	17 (27%)	Item increases complexity of provider visits, social needs, and care coordination needs.
		1-dissenting		Depends on too many other caregiver, healthcare provider, and social services factors.
				Item not specifically related to the care or needs of children with medical complexity.
	2	1-affirmative	13 (24%)	Item puts the caregivers at risk of poor understanding and puts the child at risk of unmet care needs.
				Item fulfills domain as it is a common, important and overlooked concept.
		1-dissenting		Item not specific enough to children with medical complexity as it can affect any child.
				Item does not reach the threshold to fulfill family needs.
FN22	1	1-affirmative	23 (36%)	Item may increase social isolation.
		1-dissenting		Depends on too many other caregiver, healthcare provider, and/or social services factors.
				Item not specifically related to the care or needs of children with medical complexity.
	2	1-affirmative	19 (33%)	Item fulfills domain, at least in the immediate time after arrival.
		1-dissenting		Item depends on too many other factors including: a) caregiver factors, b) circumstances of the immigration, c) country of origin, and d) amount of support for the newcomers.

				2) Social determinants of health and social system difficulties should not define children with medical complexity.
	3	1-dissenting	13 (25%)	Item depends on too many other factors including: a) circumstances of the immigration and b) family's ability to integrate into the new country.
		Level 2		Quantification of this item does not matter because "time since relocation" is not overly relevant in determining how integrated or supported a new immigrant becomes in their current country.
				Answer to the follow-up item depends too much on other factors including: a) the degree of cultural and language differences, b) familial resources and support, and c) difficulties with employment.
FN23	1	1-affirmative	11 (17%)	Item indicates increase in care burden.
		1-dissenting		Depends on too many other caregiver, healthcare provider, and/or social services factors.
				Item not specifically related to the care or needs of children with medical complexity.
				Item does not demonstrate impact on the child or caregivers.
	2	1-dissenting	15 (28%)	Item depends on too many other factors including: a) caregiver factors, b) social service factors, and c) the amount of support the caregiver has.
				Social determinants of health and social system difficulties should not define children with medical complexity.
	3	1-dissenting	7 (13%)	Item depends on too many other factors including: a) caregiver abilities and b) the amount of support the caregiver has.
FN24	1	1-affirmative	13 (20%)	
		1-dissenting		Item not specifically related to the care of children with medical complexity or health impact on children with medical complexity.
				Depends on too many other caregiver, healthcare provider, and/or social services factors.
FN25	2	Level 2	7 (13%)	Participant rationale for follow-up answers included: a) the amount of time that takes away from family functioning and b) the amount of time that seemed significant per week.
				Answer to the follow-up item depends on too many other factors to estimate.
	3	Level 2	2 (4%)	Participant rationale for follow-up answers included: a) the amount of time that seemed significant per day multiplied by 7 days in a week, and b) choosing the minimum value on the scale.
FN27	1	1-affirmative	12 (19%)	Item indicates difficulty with care access.

		1-dissenting		Depends on too many other caregiver, healthcare provider, and/or social services factors.
				Item not specifically related to the care of, or health impact on, children with medical complexity.
	2	1-affirmative	12 (21%)	The item interferes with care coordination, securing adequate home care, securing adequate access to care, and adequate resources for care.
		1-dissenting		Item may complicate care but does not meet the threshold to fulfill family needs.
				Social system difficulties should not define children with medical complexity.
	3	1-affirmative	4 (8%)	The item interferes with securing adequate access to care and/or resources for care.
FN28	1	1-affirmative	10 (16%)	Item indicates difficulty with access to care.
				Item indicates increase in care burden.
		1-dissenting		Depends on too many other child and/or caregiver factors.
HU1	1	1-affirmative	18 (29%)	Item indicates significant chronicity of care.
		1-dissenting		The item is too specific as it does not account for single centers with multiple services or complex children that rarely leave home.
				Item depends on too many other factors such as the chronicity of the services, the healthcare provider context, and/or social services context.
	2	1-affirmative	14 (24%)	Item can interfere with the care of children with medical complexity due to difficulties in inter-institutional communication and care coordination.
		1-dissenting		The item depends on too many other child and systemic factors. Some self-limited, acute, or developmental issues require care from many settings but the child may not be complex. Conversely, some institutions may offer many services in one setting and the child is complex.
	3	1-affirmative	11 (20%)	Item indicates the child has multiple needs and fulfills the Healthcare Use domain IF the other domains are met.
		1-dissenting		The item is vague and depends on too many other factors such as: a) what services are being provided at these centers and b) the nature of the child's issues as some self-limited, acute issues require care from 3 or more settings but the child may not be complex.
HU2	2	Level 2	18 (31%)	Participant rationale for follow-up answers included: a) the estimated time to care for the usual needs of a children with medical complexity at school, b) the estimated amount of time that differentiates the care needs at school of children with medical complexity from children with special healthcare needs, and c) the estimated amount of time that disrupts academic success, social relationships, and participation.

				Answer to the follow-up item depends on too many other factors to estimate including: a) duration of daily schooling for the child, b) the amount of regional resources and c) regional differences in school healthcare delivery.
	3	Level 2	14 (25%)	Participant rationale for follow-up answers included: a) the estimated amount of time that demonstrates at least daily nursing needs, b) the estimated amount of time per day the hinders education and socialization, and c) the estimated amount of time that would differentiate a child with complexity from one without.
				2) Answer to the follow-up item depends on too many other factors to estimate including: a) the varying weekly schedule of the child, b) the capacity of the school system to deliver regular care, and c) the amount of time the nurse in the school system spends on care for children without complexity.
HU4	1	1-affirmative	24 (38%)	
		1-dissenting		The item is too specific as it does not account for complexity and/or duration of the interventions.
				Item depends on too many other factors such as the burden of the interventions.
	2	Level 2	21 (38%)	Participant rationale for follow-up answers included the estimated number of interventions that is beyond that experienced by the non-complex child.
				Answer to the follow-up item depends on too many other factors to estimate including: a) the complexity of administration of the interventions, b) the amount of time it takes to administer the interventions, c) and the level of dependence on the intervention.
	3	Level 2	10 (18%)	Participant rationale for follow-up answers included the estimated number of interventions that is beyond that experienced by the non-complex child.
				2) Answer to the follow-up item depends on too many other factors to estimate including: a) the complexity of administration of the interventions and b) the impact of the intervention on the family.
HU5	2	Level 2	9 (16%)	Answer to the follow-up item depends on too many other factors to estimate including: a) the complexity of administration of the interventions and b) the degree of functional limitation requiring the intervention.
	3	Level 2	5 (9%)	Answer to the follow-up item depends on too many other factors to estimate including: a) the definition of "complex administration", b) chronicity of the intervention, and c) impact on the family.
HU6	2	Level 2	21 (36%)	Participant rationale for follow-up answers included: a) the cumulative number of specialists, therapists and primary care providers that is beyond the number experienced by

				typically developing children or children with special healthcare needs and b) the number of providers beyond which a caregiver has difficulty navigating.
HU7	1	1-dissenting	22 (35%)	Item depends on too many other factors such as the nature of the visits, age of the child, and/or a variety of caregiver factors.
	2	Level 2	15 (26%)	Participant rationale for follow-up answers included: a) the number of visits every 1-2 months considered significant by the respondent, b) the number of visits per year beyond the typical amount experienced by children with medical complexity, c) and the amount of visits that interfere with life participation.
				Answer to the follow-up item depends on too many other factors to estimate including: a) the child's age, b) the child's medical conditions, c) the stage of care evolution, and d) whether the visit is multi-disciplinary.
	3	Level 2	7 (13%)	Participant rationale for follow-up answers included: a) the number of visits per year deemed substantial EXCLUDING lab appointments and therapy visits and b) the number of visits per year that would allow the primary care provider and an average number of subspecialists to have follow-up every 6 months.
				Answer to the follow-up item depends on too many other factors to estimate including: a) the type of visit and b) whether the visit is multi-disciplinary.
HU8	1	1-affirmative	17 (27%)	Item represents a common problem in complex care.
		1-dissenting		1) Item depends on too many other factors such as the appropriateness of use of the emergency room or urgent care center, capacity of the regional health care system, and/or caregiver factors.
				Item not specifically related to the care of, or health impact on, children with medical complexity.
	2	Level 2	15 (26%)	Participant rationale for follow-up answers included: a) the number of presentations beyond that experienced by chance alone or b) the usual number of presentations experienced by a child with a complex medical condition.
	3	Level 2	5 (9%)	Participant rationale for follow-up answers included: a) the number of presentations that would differentiate a child with medical complexity from one without and b) the minimum value on the scale.
HU9	2	Level 2	8 (14%)	Participant rationale for follow-up answers included: a) any admission is enough or b) the number of admissions beyond that experienced by chance alone or by common acute conditions in typically developing children.
HU10	2	Level 2	5 (9%)	Participant rationale for follow-up answers included: a) any overnight admission is enough, b) the lowest threshold

				presented in the question, and c) the number of days that would capture multiple admissions over the year.
	3	Level 2	5 (9%)	Participant rationale for follow-up answers included: a) typical length-of-stay for common reasons is < 7 days and longer than that would be more complex and b) a very low threshold considering community-sensitive conditions would not typically require admission.
				Answer to the follow-up item depends on too many other factors to estimate including: a) hospital policies, b) reimbursement policies, c) clinical practices, d) availability of community based health services, and e) type of hospitalization.
HU11	2	Level 2	6 (11%)	Participant rationale for follow-up answers included: a) any readmission suggests 2 total admissions and is enough and b) the number deemed significant by the respondent if they are unplanned re-admissions.
	3	Level 2	6 (11%)	Participant rationale for follow-up answers included: a) the number of readmissions greater than that which could happen by chance, b) any readmissions suggest fragility of the child, c) any readmission is substantial IF discharge was properly planned, and d) 50% of total admission are allowed to have readmissions before it suggests complexity.
				Answer to the follow-up item depends on too many other factors to estimate including: a) total number of hospitalizations for that child and b) what the reason for the admission and/or readmission was.
HU12	2	Level 2	7 (12%)	Participant rationale for follow-up answers included: a) the number of admissions considered significant by the respondent, b) the number of admissions beyond the number experienced by chance alone or by common acute conditions, and c) any admission is enough.
				2) Answer to the follow-up item depends on too many other factors to estimate including the institution's intensive care unit admission criteria (ex. some institutions admit all children with tracheostomies to the intensive care unit).
	3	Level 2	4 (7%)	Answer to the follow-up item depends on too many other factors to estimate including: a) the child's underlying needs/condition and 2) the institution's intensive care unit admission criteria (ex. some institutions admit all children with ventilators to the intensive care unit).
HU13	2	Level 2	8 (14%)	Participant rationale for follow-up answers included: a) the number of days that accounts for a significant number of overnight intensive care unit admissions per year and b) any day in the intensive care unit is enough.

				Answer to the follow-up item depends on too many other factors to estimate including the institution's intensive care unit admission criteria (ex. some institutions admit all children with tracheostomies to the intensive care unit).
	3	Level 2	5 (9%)	Participant rationale for follow-up answers included: a) any day in the intensive care unit is enough as it suggests fragility or failure of management.
				Answer to the follow-up item depends on too many other factors to estimate including: a) the child's underlying needs/condition and 2) the institution's intensive care unit admission criteria (ex. some institutions admit all children with ventilators to the intensive care unit).
FL1	1	1-affirmative	25 (41%)	
		1-dissenting		Item depends on too many other factors such as the needs of the child, the capacity of the caregivers, and/or the level of the underlying therapy.
				Item not specifically related to the needs of children with medical complexity.
	2	1-dissenting	17 (33%)	The item depends on too many other factors including: a) the type of therapy, b) which functions are limited requiring the therapy, and c) therapy resource availability.
				Item is not specific enough to children with medical complexity as the therapy may be related to a singular, acute incident or a developmental condition.
		Level 2		Participant rationale for follow-up answers included: a) the amount of daily therapy if caregiver-administered therapy is counted, b) the amount of therapy per year that would differentiate significant functional limitation from minor issues, c) the amount of daily therapy that impacts daily routines, and d) the usual amount of therapy provided at school per week.
				Answer to the follow-up item depends on too many other factors to estimate including: a) the child's potential for rehabilitation, b) what therapy is being provided, c) which functions are limited requiring the therapy, and d) therapy resource availability.
	3	Level 2	6 (11%)	Answer to the follow-up item depends on too many other factors to estimate including: a) the amount of therapy funding provided by insurance.
FL9	2	1-affirmative	12 (21%)	Item indicates dependence on this device for optimal functioning.
		1-dissenting		Item depends on to many other factors such as the impact on a child's quality of life and/or activities of daily life.
				Item should include other types of technology beyond mobility devices.
DC1	1	1-affirmative	36 (57%)	Sometimes the diagnosis is not yet known but the child is still complex.

				The other 3 domains determine the need for complex care services, not the diagnosis.
		1-dissenting		If the other 3 domains are met, it seems impossible to not have a diagnosis.
				This item would only fulfill the domain if it were referring to conditions the were not yet diagnosed.
	2	1-affirmative	21 (36%)	Item fulfills domain as it allows the inclusion of undiagnosed conditions, novel conditions or changing diagnoses.
				Item fulfills domain because the other three domains are the only information needed to be considered complex.
		1-dissenting		Item alone is not enough to fulfill the domain because the children must have medical conditions of some sort even if they do not have unifying or diagnosed condition.
				Item alone is not enough to fulfill the domain because without an appropriately chronic or complex condition, fulfilling the other three domains could define a child with a minor condition as being complex.
	3	1-affirmative	18 (33%)	Item fulfills domain as it allows the inclusion of undiagnosed conditions, novel diagnoses, changing diagnoses, and/or states of fragility.
		1-dissenting		Item alone is not enough to fulfill the domain because the children must have a medical condition of some sort to meet the complexity diagnosis, even if they do not have unifying or diagnosed condition.
				Item alone is not enough to fulfill the domain because without an appropriately chronic or complex condition, fulfilling the other three domains could define a child with a minor condition or with a difficult social situation as being complex.
DC2	1	1-affirmative	22 (35%)	The other 3 domains determine the need for complex care services, not the diagnosis.
		1-dissenting		The diagnosis must be chronic and/or complex.
				Item depends on the other 3 domains and diagnosis does not matter.
	2	1-dissenting	23 (40%)	Item alone is not enough to fulfill the diagnostic conditions domain because the diagnostic condition must be chronic OR chronic and complex, otherwise children with minor conditions may be considered complex.
		Level 2		Respondent rationale for follow-up answers included: a) the number of conditions that suggests multiple organ system involvement and b) only 1 condition is needed if the other 3 domains are fulfilled.
				Answer to the follow-up item depends on too many other factors to estimate including the severity of the diagnostic

				condition(s) and the impact of the condition(s) on the child and family.
DC3	1	1-affirmative	17 (27%)	Agree if multiple chronic disease are present.
				A diagnosis and it's chronicity is important.
		1-dissenting		This item must indicate greater than 1 chronic condition to fulfill the domain.
				Item depends on too many other factors such as the duration of the condition and/or level of daily functioning.
				If the other 3 domains are met, a diagnosis is not needed.
	2	1-dissenting	13 (23%)	Item is too specific because: a) the word "medical" omits pertinent non-medical conditions and b) diagnostic conditions do not have to be chronic to be considered complex.
				Item alone is not enough to fulfill the diagnostic conditions domain because the diagnostic condition(s) must be considered complex.
		Level 2		Respondent rationale for follow-up answers included: a) the number of chronic condition(s) must be greater than one, b) only 1 chronic condition is required if the other domains are met, and c) the number of chronic conditions that separates children with special healthcare needs from children with medical complexity.
	3	1-dissenting	10 (18%)	Item alone is not enough to fulfill the diagnostic conditions domain because the diagnostic condition(s) must be considered complex.
		Level 2		Respondent rationale for follow-up answers included: a) the number of chronic condition(s) must be greater than one and b) one chronic condition(s) is enough as some chronic condition(s) are complex.
DC4	1	1-affirmative	16 (26%)	A diagnosis and it's complexity is important.
		1-dissenting		A diagnosis does not necessarily have to be complex to fulfill the domain.
				This item must indicate greater than 1 complex condition to fulfill the domain.
	2	1-affirmative	14 (27%)	The item fulfills the diagnostic conditions domain because: a) the condition must involve at least 2 body systems and b) the diagnostic condition must be complex.
		1-dissenting		Item is too specific because: a) children can be considered complex with multiple chronic conditions and no complex conditions and b) the word "medical" omits pertinent non-medical conditions.
		Level 2		Answer to the follow-up item depends on too many other factors to estimate including: a) the severity of the diagnostic condition(s) and b) the comprehensiveness of the care provider in creating the problem list of unifying diagnoses and/or comorbidities.

14. REFERENCES

1. Srivastava, R., B.L. Stone, and N.A. Murphy, *Hospitalist care of the medically complex child.* Pediatr Clin North Am, 2005. **52**(4): p. 1165-87, x.
2. Cohen, E., et al., *Patterns and costs of health care use of children with medical complexity.* Pediatrics, 2012. **130**(6): p. e1463-70.
3. Buescher, P.A., et al., *Children who are medically fragile in North Carolina: using Medicaid data to estimate prevalence and medical care costs in 2004.* Matern Child Health J, 2006. **10**(5): p. 461-6.
4. Neff, J.M., et al., *Profile of medical charges for children by health status group and severity level in a Washington State Health Plan.* Health Serv Res, 2004. **39**(1): p. 73-89.
5. *Children and Youth With Medical Complexity in Canada*. 2020, Canadian Institute for Health Information: Ottawa, ON.
6. Strickland, B.B., et al., *Assessing systems quality in a changing health care environment: the 2009-10 national survey of children with special health care needs.* Matern Child Health J, 2015. **19**(2): p. 353-61.
7. Perrin, J.M., L.E. Anderson, and J. Van Cleave, *The rise in chronic conditions among infants, children, and youth can be met with continued health system innovations.* Health Aff (Millwood), 2014. **33**(12): p. 2099-105.
8. Simon, T.D., et al., *Children with complex chronic conditions in inpatient hospital settings in the United States.* Pediatrics, 2010. **126**(4): p. 647-655.
9. Berry, J.G., et al., *Trends in resource utilization by children with neurological impairment in the United States inpatient health care system: a repeat cross-sectional study.* PLoS Med, 2012. **9**(1): p. e1001158.
10. Berry, J.G., et al., *Inpatient growth and resource use in 28 children's hospitals: a longitudinal, multi-institutional study.* JAMA Pediatr, 2013. **167**(2): p. 170-7.
11. Burns, K.H., et al., *Increasing prevalence of medically complex children in US hospitals.* Pediatrics, 2010. **126**(4): p. 638-646.
12. Berry, J.G., et al., *Hospital utilization and characteristics of patients experiencing recurrent readmissions within children's hospitals.* Jama, 2011. **305**(7): p. 682-90.
13. Matlow, A.G., et al., *Adverse events among children in Canadian hospitals: the Canadian Paediatric Adverse Events Study.* Cmaj, 2012. **184**(13): p. E709-18.
14. Msall, M.E. and M.R. Tremont, *Measuring functional outcomes after prematurity: developmental impact of very low birth weight and extremely low birth weight status on childhood disability.* Ment Retard Dev Disabil Res Rev, 2002. **8**(4): p. 258-72.
15. Tennant, P.W., et al., *20-year survival of children born with congenital anomalies: a population-based study.* Lancet, 2010. **375**(9715): p. 649-56.
16. Wong, L.Y. and L.J. Paulozzi, *Survival of infants with spina bifida: a population study, 1979-94.* Paediatr Perinat Epidemiol, 2001. **15**(4): p. 374-8.
17. Hallahan, A.R., et al., *Improved outcomes of children with malignancy admitted to a pediatric intensive care unit.* Crit Care Med, 2000. **28**(11): p. 3718-21.
18. Magnani, C., et al., *Trends in survival after childhood cancer in Europe, 1978-1997: report from the Automated Childhood Cancer Information System project (ACCIS).* Eur J Cancer, 2006. **42**(13): p. 1981-2005.
19. Brehaut, J.C., et al., *Health among caregivers of children with health problems: findings from a Canadian population-based study.* Am J Public Health, 2009. **99**(7): p. 1254-62.

20. Cohen, E., et al., *Association Between the Birth of an Infant With Major Congenital Anomalies and Subsequent Risk of Mortality in Their Mothers.* Jama, 2016. **316**(23): p. 2515-2524.
21. Cohen, E., et al., *Residential movement patterns of families of young children with chronic conditions in Ontario, Canada: a population-based cohort study.* Int J Equity Health, 2013. **12**: p. 62.
22. Epel, E.S., et al., *Accelerated telomere shortening in response to life stress.* Proc Natl Acad Sci U S A, 2004. **101**(49): p. 17312-5.
23. Hexem, K.R., A.M. Bosk, and C. Feudtner, *The dynamic system of parental work of care for children with special health care needs: a conceptual model to guide quality improvement efforts.* BMC Pediatr, 2011. **11**: p. 95.
24. Berry, J.G., et al., *Characteristics of hospitalizations for patients who use a structured clinical care program for children with medical complexity.* Journal of Pediatrics, 2011. **159**(2): p. 284-290.
25. Yantzi, N., et al., *The impacts of distance to hospital on families with a child with a chronic condition.* Soc Sci Med, 2001. **52**(12): p. 1777-91.
26. *Health Care Innovation Awards Round Two*. 2014 [cited 2020 January 7]; Available from: http://innovation.cms.gov/initiatives/Health-Care-Innovation-Awards/Round-2.html.
27. *Optimizing the Role of Complex Continuing Care and Rehabilitation in the Transformation of the Health Care Delivery System*. 2006 [cited 2020 January 7]; Available from: https://www.oha.com/CurrentIssues/keyinitiatives/eHealth/Documents/Optimizing_the_Role_of_CCCandRehab.pdf.
28. Langer, C.S., et al., *Evolving Federal and State Health Care Policy: Toward a More Integrated and Comprehensive Care-Delivery System for Children With Medical Complexity.* Pediatrics, 2018. **141**(Suppl 3): p. S259-s265.
29. Orkin, J., et al., *Complex care for kids Ontario: protocol for a mixed-methods randomised controlled trial of a population-level care coordination initiative for children with medical complexity.* BMJ Open, 2019. **9**(8): p. e028121.
30. De Jonge, P., et al., *Operationalization of biopsychosocial case complexity in general health care: the INTERMED project.* Australian & New Zealand Journal of Psychiatry, 2005. **39**(9): p. 795-799.
31. Ben-Chetrit, E., et al., *Consensus proposal for taxonomy and definition of the autoinflammatory diseases (AIDs): a Delphi study.* Ann Rheum Dis, 2018. **77**(11): p. 1558-1565.
32. Kuo, D.Z. and A.J. Houtrow, *Recognition and management of medical complexity.* Pediatrics, 2016. **138**(6): p. e20163021.
33. Zanello, E., et al., *Care coordination for children with special health care needs: A cohort study.* Italian Journal of Pediatrics, 2017. **43**.
34. Berry, J.G., et al., *Ways to Identify Children with Medical Complexity and the Importance of Why.* Journal of Pediatrics, 2015. **167**(2): p. 229-237.
35. Cohen, E., et al., *Status complexicus? The emergence of pediatric complex care.* Pediatrics, 2018. **141**(Supplement 3): p. S202-S211.
36. Cohen, E., et al., *Children with medical complexity: an emerging population for clinical and research initiatives.* Pediatrics, 2011. **127**(3): p. 529-538.
37. Pordes, E., et al., *Models of Care Delivery for Children With Medical Complexity.* Pediatrics, 2018. **141**(Suppl 3): p. S212-s223.
38. Cohen, E., et al., *Hospital-based comprehensive care programs for children with special health care needs: a systematic review.* Arch Pediatr Adolesc Med, 2011. **165**(6): p. 554-61.
39. Casey, P.H., et al., *Effect of hospital-based comprehensive care clinic on health costs for medicaid-insured medically complex children.* Archives of Pediatrics and Adolescent Medicine, 2011. **165**(5): p. 392-398.

40. Condren, M., et al., *Medication Reconciliation Across Care Transitions in the Pediatric Medical Home.* Joint Commission Journal on Quality and Patient Safety, 2019.
41. Kelly, A., et al., *Children with complex chronic medical conditions and special needs privately insured through an HMO.* Families, Systems and Health, 2002. **20**(3): p. 279-289.
42. Maynard, R., et al., *Home health care availability and discharge delays in children with medical complexity.* Pediatrics, 2019. **143**(1).
43. Nelson, B.B., et al., *How Avoidable are Hospitalizations for Children With Medical Complexity? Understanding Parent Perspectives.* Academic Pediatrics, 2016. **16**(6): p. 579-586.
44. Coquillette, M., et al., *Social Work Services Utilization by Children with Medical Complexity.* Maternal and Child Health Journal, 2015. **19**(12): p. 2707-2713.
45. Gordon, J.B., et al., *A tertiary care-primary care partnership model for medically complex and fragile children and youth with special health care needs.* Archives of Pediatrics and Adolescent Medicine, 2007. **161**(10): p. 937-944.
46. Johaningsmeir, S.A., et al., *Impact of caring for children with medical complexity and high resource use on family quality of life.* Journal of Pediatric Rehabilitation Medicine, 2015. **8**(2): p. 75-82.
47. Cohen, E., et al., *Integrated complex care coordination for children with medical complexity: A mixed-methods evaluation of tertiary care-community collaboration.* BMC Health Services Research, 2012. **12**(1).
48. Kingsnorth, S., et al., *Inter-organizational partnership for children with medical complexity: The integrated complex care model.* Child: Care, Health and Development, 2015. **41**(1): p. 57-66.
49. Nageswaran, S., et al., *An innovative collaborative model of care for undiagnosed complex medical conditions.* Pediatrics, 2017. **139**(5).
50. Homer, C.J., et al., *A review of the evidence for the medical home for children with special health care needs.* Pediatrics, 2008. **122**(4): p. e922-37.
51. Berry, J.G., et al., *Ways to Identify Children with Medical Complexity and the Importance of Why.* J Pediatr, 2015. **167**(2): p. 229-37.
52. Dewan, T. and E. Cohen, *Children with medical complexity in Canada.* Paediatrics & child health, 2013. **18**(10): p. 518-522.
53. Russell, C.J. and T.D. Simon, *Care of children with medical complexity in the hospital setting.* Pediatr Ann, 2014. **43**(7): p. e157-62.
54. Cohen, E., et al., *Hospital-Based Comprehensive Care Programs for Children With Special Health Care Needs: A Systematic Review.* Archives of Pediatrics & Adolescent Medicine, 2011. **165**(6): p. 554-561.
55. Diskin, C., et al., *Research priorities for children with neurological impairment and medical complexity in high-income countries.* Dev Med Child Neurol, 2021.
56. Cooley, W.C., *Providing a primary care medical home for children and youth with cerebral palsy.* Pediatrics, 2004. **114**(4): p. 1106-13.
57. Cooley, W.C., *Redefining primary pediatric care for children with special health care needs: the primary care medical home.* Curr Opin Pediatr, 2004. **16**(6): p. 689-92.
58. Palfrey, J.S., et al., *The Pediatric Alliance for Coordinated Care: evaluation of a medical home model.* Pediatrics, 2004. **113**(5 Suppl): p. 1507-16.
59. Cooley, W.C. and J.W. McAllister, *Building medical homes: improvement strategies in primary care for children with special health care needs.* Pediatrics, 2004. **113**(5 Suppl): p. 1499-506.
60. Farmer, J.E., et al., *Comprehensive primary care for children with special health care needs in rural areas.* Pediatrics, 2005. **116**(3): p. 649-56.

61. Mosquera, R.A., et al., *Effect of an enhanced medical home on serious illness and cost of care among high-risk children with chronic illness: a randomized clinical trial.* Jama, 2014. **312**(24): p. 2640-8.
62. Mosquera, R.A., et al., *Hospital Consultation From Outpatient Clinicians for Medically Complex Children: A Randomized Clinical Trial.* JAMA Pediatr, 2021. **175**(1): p. e205026.
63. Berman, S., et al., *Utilization and costs for children who have special health care needs and are enrolled in a hospital-based comprehensive primary care clinic.* Pediatrics, 2005. **115**(6): p. e637-42.
64. Perrin, J.M., et al., *A family-centered, community-based system of services for children and youth with special health care needs.* Arch Pediatr Adolesc Med, 2007. **161**(10): p. 933-6.
65. Simon, T.D., S. Mahant, and E. Cohen, *Pediatric hospital medicine and children with medical complexity: past, present, and future.* Curr Probl Pediatr Adolesc Health Care, 2012. **42**(5): p. 113-9.
66. Liptak, G.S., et al., *Effects of providing comprehensive ambulatory services to children with chronic conditions.* Arch Pediatr Adolesc Med, 1998. **152**(10): p. 1003-8.
67. Murphy, N.A., et al., *Children With Medical Complexity: The 10-Year Experience of a Single Center.* Hosp Pediatr, 2020. **10**(8): p. 702-708.
68. Gordon, J.B., et al., *A tertiary care-primary care partnership model for medically complex and fragile children and youth with special health care needs.* Arch Pediatr Adolesc Med, 2007. **161**(10): p. 937-44.
69. Cohen, E., et al., *Integrated complex care coordination for children with medical complexity: a mixed-methods evaluation of tertiary care-community collaboration.* BMC Health Serv Res, 2012. **12**: p. 366.
70. Simon, T.D., et al., *Pediatric hospitalist comanagement of spinal fusion surgery patients.* J Hosp Med, 2007. **2**(1): p. 23-30.
71. Homer, C.J., W.C. Cooley, and B. Strickland, *Medical home 2009: what it is, where we were, and where we are today.* Pediatr Ann, 2009. **38**(9): p. 483-90.
72. Hirsch, A.W., et al., *Characteristics of Children Hospitalized With Aspiration Pneumonia.* Hosp Pediatr, 2016. **6**(11): p. 659-666.
73. Thomson, J., et al., *Aspiration and Non-Aspiration Pneumonia in Hospitalized Children With Neurologic Impairment.* Pediatrics, 2016. **137**(2): p. e20151612.
74. Jacobson, S.J., et al., *A randomized controlled trial of penicillin vs clindamycin for the treatment of aspiration pneumonia in children.* Arch Pediatr Adolesc Med, 1997. **151**(7): p. 701-4.
75. Brook, I., *Aspiration pneumonia in institutionalized children. A retrospective comparison of treatment with penicillin G, clindamycin and carbenicillin.* Clin Pediatr (Phila), 1981. **20**(2): p. 117-22.
76. Brook, I., *Treatment of aspiration or tracheostomy-associated pneumonia in neurologically impaired children: effect of antimicrobials effective against anaerobic bacteria.* Int J Pediatr Otorhinolaryngol, 1996. **35**(2): p. 171-7.
77. Thomson, J., et al., *Antibiotics for Aspiration Pneumonia in Neurologically Impaired Children.* J Hosp Med, 2020. **15**(7): p. 395-402.
78. McPherson, M., et al., *A new definition of children with special health care needs.* Pediatrics, 1998. **102**(1 Pt 1): p. 137-40.
79. Stein, R.E., L.E. Westbrook, and L.J. Bauman, *The Questionnaire for Identifying Children with Chronic Conditions: a measure based on a noncategorical approach.* Pediatrics, 1997. **99**(4): p. 513-21.
80. Bramlett, M.D., et al., *Differentiating subgroups of children with special health care needs by health status and complexity of health care needs.* Matern Child Health J, 2009. **13**(2): p. 151-63.

81. Davis, M.M. and J.P. Brosco, *Being specific about being special: defining children's conditions and special health care needs.* Arch Pediatr Adolesc Med, 2007. **161**(10): p. 1003-5.
82. Stein, R.E., et al., *Framework for identifying children who have chronic conditions: the case for a new definition.* J Pediatr, 1993. **122**(3): p. 342-7.
83. Cooley, W.C., et al., *Improved outcomes associated with medical home implementation in pediatric primary care.* Pediatrics, 2009. **124**(1): p. 358-64.
84. Gadomski, A., P. Jenkins, and M. Nichols, *Impact of a Medicaid primary care provider and preventive care on pediatric hospitalization.* Pediatrics, 1998. **101**(3): p. E1.
85. Todd, J., et al., *Increased rates of morbidity, mortality, and charges for hospitalized children with public or no health insurance as compared with children with private insurance in Colorado and the United States.* Pediatrics, 2006. **118**(2): p. 577-85.
86. Berry, J.G., et al., *Characteristics of hospitalizations for patients who use a structured clinical care program for children with medical complexity.* J Pediatr, 2011. **159**(2): p. 284-90.
87. Feudtner, C., D.A. Christakis, and F.A. Connell, *Pediatric deaths attributable to complex chronic conditions: A population-based study of Washington State, 1980-1997.* Pediatrics, 2000. **106**(1 II): p. 205-209.
88. Feudtner, C., et al., *Pediatric complex chronic conditions classification system version 2: updated for ICD-10 and complex medical technology dependence and transplantation.* BMC Pediatr, 2014. **14**: p. 199.
89. Feudtner, C., et al., *How well can hospital readmission be predicted in a cohort of hospitalized children? A retrospective, multicenter study.* Pediatrics, 2009. **123**(1): p. 286-93.
90. Gay, J.C., et al., *Epidemiology of 15-Day Readmissions to a Children's Hospital.* Pediatrics, 2011. **127**(6): p. e1505-12.
91. Peltz, A., et al., *Characteristics of Children Enrolled in Medicaid With High-Frequency Emergency Department Use.* Pediatrics, 2017. **140**(3).
92. Lu, S. and D.Z. Kuo, *Hospital charges of potentially preventable pediatric hospitalizations.* Acad Pediatr, 2012. **12**(5): p. 436-44.
93. Simon, T.D., et al., *Pediatric medical complexity algorithm: a new method to stratify children by medical complexity.* Pediatrics, 2014. **133**(6): p. e1647-54.
94. Chan, T., et al., *Pediatric Critical Care Resource Use by Children with Medical Complexity.* J Pediatr, 2016. **177**: p. 197-203.e1.
95. *International Classification of Functioning, Disability, And health (Version 1.0)*. 2001, World Health Organization (WHO): Geneva.
96. *3M Clinical Risk Grouping Software (TM)*. 2021 October 28, 2021]; Available from: https://www.3m.com/3M/en_US/health-information-systems-us/drive-value-based-care/patient-classification-methodologies/crgs/.
97. Neff, J.M., et al., *Identifying and classifying children with chronic conditions using administrative data with the clinical risk group classification system.* Ambul Pediatr, 2002. **2**(1): p. 71-9.
98. Neff, J.M., et al., *Stratification of children by medical complexity.* Acad Pediatr, 2015. **15**(2): p. 191-6.
99. Morley, J.E., et al., *Frailty Consensus: A Call to Action.* Journal of the American Medical Directors Association, 2013. **14**(6): p. 392-397.
100. Berry, J.G., et al., *Pediatric readmission prevalence and variability across hospitals.* Jama, 2013. **309**(4): p. 372-80.
101. Berry, J.G., et al., *Contributions of Children With Multiple Chronic Conditions to Pediatric Hospitalizations in the United States: A Retrospective Cohort Analysis.* Hosp Pediatr, 2017. **7**(7): p. 365-372.

102. Berry, J.G., et al., *Predictors of clinical outcomes and hospital resource use of children after tracheotomy.* Pediatrics, 2009. **124**(2): p. 563-72.
103. Shah, S.S., et al., *A multicenter study of factors influencing cerebrospinal fluid shunt survival in infants and children.* Neurosurgery, 2008. **62**(5): p. 1095-102; discussion 1102-3.
104. *Improving the Lives of Children and Youth with Special Health Care Needs (CYSHCN)*. 2021 November 1, 2021]; Available from: https://mchb.hrsa.gov/maternal-child-health-topics/children-and-youth-special-health-needs.
105. Bethell, C.D., et al., *Taking stock of the CSHCN screener: a review of common questions and current reflections.* Acad Pediatr, 2015. **15**(2): p. 165-76.
106. Kuo, D.Z., et al., *Variation in child health care utilization by medical complexity.* Matern Child Health J, 2015. **19**(1): p. 40-8.
107. Berry, J.G., et al., *Children with medical complexity and Medicaid: spending and cost savings.* Health Aff (Millwood), 2014. **33**(12): p. 2199-206.
108. van der Lee, J.H., et al., *Definitions and measurement of chronic health conditions in childhood: a systematic review.* Jama, 2007. **297**(24): p. 2741-51.
109. Feudtner, C., D.A. Christakis, and F.A. Connell, *Pediatric deaths attributable to complex chronic conditions: a population-based study of Washington State, 1980–1997.* Pediatrics, 2000. **106**(Supplement 1): p. 205-209.
110. McPherson, M., et al., *A new definition of children with special health care needs.* Pediatrics, 1998. **102**(1): p. 137-139.
111. Stein, R. and D.J. Jessop, *A noncategorical approach to chronic childhood illness.* Public health reports, 1982. **97**(4): p. 354.
112. Law, M., et al., *Service coordination for children and youth with complex needs.* Hamilton, ON, Canada: CanChild Centre for Childhood Disability Research, McMaster University, 2004.
113. Neff, J.M., et al., *Profile of medical charges for children by health status group and severity level in a Washington State Health Plan.* Health services research, 2004. **39**(1): p. 73-90.
114. Neff, J.M., et al., *Identifying and classifying children with chronic conditions using administrative data with the clinical risk group classification system.* Ambulatory Pediatrics, 2002. **2**(1): p. 71-79.
115. Neff, J.M., et al., *Using medical billing data to evaluate chronically ill children over time.* The Journal of ambulatory care management, 2006. **29**(4): p. 283-290.
116. Meehan, E., et al., *Comprehensive care programmes for children with medical complexity.* Cochrane Database of Systematic Reviews, 2019(5).
117. Looman, W.S., et al., *Outcomes research on children with medical complexity: A scoping review of gaps and opportunities.* Child: care, health, and development., 2019.
118. Berry, J.G., et al., *Ways to identify children with medical complexity and the importance of why.* The Journal of pediatrics, 2015. **167**(2): p. 229-237.
119. Artiga;, S. and E. Hinton *Beyond Health Care: The Role of Social Determinants in Promoting Health and Health Equity*. May 10, 2018.
120. Hsu, C.-C. and B.A. Sandford, *The Delphi technique: making sense of consensus.* Practical Assessment, Research, and Evaluation, 2007. **12**(1): p. 10.
121. Powell, C., *The Delphi technique: myths and realities.* Journal of advanced nursing, 2003. **41**(4): p. 376-382.
122. Murphy, M.K., et al., *Consensus development methods, and their use in clinical guideline development.* Health Technol Assess, 1998. **2**(3): p. i-iv, 1-88.
123. Dawson, S. and J. Barker, *Hospice and palliative care: A Delphi survey of occupational therapists' roles and training needs.* Australian Occupational Therapy Journal, 1995. **42**(3): p. 119-127.

124. Delbecq, A.L., A.H. Van de Ven, and D.H. Gustafson, *Group techniques for program planning: A guide to nominal group and Delphi processes*. 1975: Scott, Foresman.
125. McPherson, S., C. Reese, and M.C. Wendler, *Methodology Update: Delphi Studies.* Nurs Res, 2018. **67**(5): p. 404-410.
126. Hasson, F., S. Keeney, and H. McKenna, *Research guidelines for the Delphi survey technique.* J Adv Nurs, 2000. **32**(4): p. 1008-15.
127. Green, B., et al., *Applying the Delphi technique in a study of GPs' information requirements.* Health & social care in the community, 1999. **7**(3): p. 198-205.
128. Humphrey-Murto, S., et al., *The Use of the Delphi and Other Consensus Group Methods in Medical Education Research: A Review.* Acad Med, 2017. **92**(10): p. 1491-1498.
129. Fitch, K., et al., *The RAND/UCLA Appropriateness Method User's Manual*. 2001: RAND Corporation.
130. McMillan, S.S., M. King, and M.P. Tully, *How to use the nominal group and Delphi techniques.* Int J Clin Pharm, 2016. **38**(3): p. 655-62.
131. Keeney, S., F. Hasson, and H. McKenna, *Consulting the oracle: ten lessons from using the Delphi technique in nursing research.* J Adv Nurs, 2006. **53**(2): p. 205-12.
132. Beech, B., *Studying the future: a Delphi survey of how multi-disciplinary clinical staff view the likely development of two community mental health centres over the course of the next two years.* J Adv Nurs, 1997. **25**(2): p. 331-8.
133. Butterworth, T. and V. Bishop, *Identifying the characteristics of optimum practice: findings from a survey of practice experts in nursing, midwifery and health visiting.* J Adv Nurs, 1995. **22**(1): p. 24-32.
134. Gibson, J.M., *Using the Delphi technique to identify the content and context of nurses' continuing professional development needs.* J Clin Nurs, 1998. **7**(5): p. 451-9.
135. Hartley, M.E., *The development of module evaluation: a Delphi approach.* Nurse Educ Today, 1995. **15**(4): p. 267-73.
136. Leyenaar, J.K., et al., *Importance and Feasibility of Transitional Care for Children With Medical Complexity: Results of a Multistakeholder Delphi Process.* Academic pediatrics, 2018. **18**(1): p. 94-101.
137. Fayed, N., et al., *Family-provider consensus outcomes for children with medical complexity.* Developmental Medicine & Child Neurology, 2018.
138. Barnert, E.S., et al., *A healthy life for a child with medical complexity: 10 domains for conceptualizing health.* Pediatrics, 2018. **142**(3): p. e20180779.
139. Diskin, C., et al., *Research priorities for children with neurological impairment and medical complexity in high-income countries.* Developmental Medicine & Child Neurology, 2022. **64**(2): p. 200-208.
140. Morin, L., et al., *Refractory septic shock in children: a European Society of Paediatric and Neonatal Intensive Care definition.* Intensive Care Med, 2016. **42**(12): p. 1948-1957.
141. Tait, A.R. and M.E. Geisser, *Development of a consensus operational definition of child assent for research.* BMC Med Ethics, 2017. **18**(1): p. 41.
142. Vanoni, F., et al., *An international delphi survey for the definition of the variables for the development of new classification criteria for periodic fever aphtous stomatitis pharingitis cervical adenitis (PFAPA).* Pediatr Rheumatol Online J, 2018. **16**(1): p. 27.
143. Gibson, F., et al., *Developing a national 'low risk' febrile neutropenia framework for use in children and young people's cancer care.* Support Care Cancer, 2013. **21**(5): p. 1241-51.
144. Roth, J., et al., *Preliminary Definitions for the Sonographic Features of Synovitis in Children.* Arthritis Care Res (Hoboken), 2017. **69**(8): p. 1217-1223.

145. van Ganzewinkel, C.J., et al., *Chronic pain in the newborn: toward a definition.* Clin J Pain, 2014. **30**(11): p. 970-7.
146. Lux, A.L. and J.P. Osborne, *A proposal for case definitions and outcome measures in studies of infantile spasms and West syndrome: consensus statement of the West Delphi group.* Epilepsia, 2004. **45**(11): p. 1416-28.
147. Ludwig, B., *Predicting the future: Have you considered using the Delphi methodology.* Journal of extension, 1997. **35**(5): p. 1-4.
148. Asselin, M. and M. Harper, *Revisiting the Delphi technique: implications for nursing professional development.* J Nurses Prof Dev, 2014. **30**(1): p. 11-5.
149. Keeney, S., F. Hasson, and H. McKenna, *Consulting the oracle: ten lessons from using the Delphi technique in nursing research.* Journal of Advanced Nursing, 2006. **53**(2): p. 205-212.
150. Jairath, N. and J. Weinstein, *The Delphi methodology (Part one): A useful administrative approach.* Can J Nurs Adm, 1994. **7**(3): p. 29-42.
151. Toronto, C., *Considerations when conducting e-Delphi research: a case study.* Nurse Res, 2017. **25**(1): p. 10-15.
152. Jones, J., C. Sanderson, and N. Black, *What Will Happen to the Quality of Care with Fewer Junior Doctors?: A Delphi Study of Consultant Physicians' Views.* Journal of the Royal College of Physicians of London, 1992. **26**(1): p. 36.
153. Cunliffe, S., *Forecasting risks in the tourism industry using the Delphi technique.* Tourism, 2002. **50**(1): p. 31-41.
154. Altschuld, J.W. and P.M. Thomas, *Considerations in the application of a modified scree test for Delphi survey data.* Evaluation Review, 1991. **15**(2): p. 179-188.
155. Marchant, E., *Methodological problems associated with the use of the Delphi technique—some comments.* Fire Technology, 1988. **24**(1): p. 59-62.
156. Goodman, C.M., *The Delphi technique: a critique.* Journal of advanced nursing, 1987. **12**(6): p. 729-734.
157. Cyphert, F.R. and W.L. Gant, *The Delphi technique: A case study. .* Phi Delta Kappan, 1971. **52**: p. 272-273.
158. Dalkey, N., *An experimental study of group opinion: the Delphi method.* Futures, 1969. **1**(5): p. 408-426.
159. McKenna, H.P., *The Delphi technique: a worthwhile research approach for nursing?* Journal of advanced nursing, 1994. **19**(6): p. 1221-1225.
160. Pill, J., *The Delphi method: substance, context, a critique and an annotated bibliography.* Socio-economic planning sciences, 1971. **5**(1): p. 57-71.
161. Fan, W. and Z. Yan, *Factors affecting response rates of the web survey: A systematic review.* Comput Human Behav, 2010. **26**(2): p. 132–139.
162. Asch, D.A., M.K. Jedrziewski, and N.A. Christakis, *Response rates to mail surveys published in medical journals.* J Clin Epidemiol, 1997. **50**(10): p. 1129-36.
163. Griffin, J.M., et al., *A comparison of small monetary incentives to convert survey non-respondents: a randomized control trial.* BMC Med Res Methodol, 2011. **11**: p. 81.
164. Young, J.M., et al., *Unconditional and conditional incentives differentially improved general practitioners' participation in an online survey: randomized controlled trial.* J Clin Epidemiol, 2015. **68**(6): p. 693-7.
165. Ulrich, C.M., et al., *Does it pay to pay? A randomized trial of prepaid financial incentives and lottery incentives in surveys of nonphysician healthcare professionals.* Nurs Res, 2005. **54**(3): p. 178-83.

166. Pit, S.W., T. Vo, and S. Pyakurel, *The effectiveness of recruitment strategies on general practitioner's survey response rates - a systematic review.* BMC Med Res Methodol, 2014. **14**: p. 76.
167. Wilson, P.M., et al., *Effects of a financial incentive on health researchers' response to an online survey: a randomized controlled trial.* J Med Internet Res, 2010. **12**(2): p. e13.
168. Glidewell, L., et al., *Do incentives, reminders or reduced burden improve healthcare professional response rates in postal questionnaires? two randomised controlled trials.* BMC Health Serv Res, 2012. **12**: p. 250.
169. Macfarlane, L., G. Owens, and P. Cruz Bdel, *Identifying the features of an exercise addiction: A Delphi study.* J Behav Addict, 2016. **5**(3): p. 474-84.
170. Judd, R.C., *Use of Delphi methods in higher education.* Technological Forecasting and Social Change, 1972. **4**(2): p. 173-186.
171. Cantrill JA, S.B., Buetow S., *The Delphi and Nominal Group Techniques in Health Services Research. .* International Journal of Pharmacy Practice, 1996. **4**: p. 67-74.
172. Grimshaw, J. and I. Russell, *Achieving health gain through clinical guidelines. I: Developing scientifically valid guidelines.* Qual Health Care, 1993. **2**(4): p. 243-8.
173. Miller, G., *The development of indicators for sustainable tourism: results of a Delphi survey of tourism researchers.* Tourism management, 2001. **22**(4): p. 351-362.
174. Anderson, D.H., *Using the Delphi process to identify significant recreation research-based innovations.* Journal of Park and Recreation Administration, 1993. **11**(1): p. 25-36.
175. Jünger, S., et al., *Guidance on Conducting and REporting DElphi Studies (CREDES) in palliative care: Recommendations based on a methodological systematic review.* Palliat Med, 2017. **31**(8): p. 684-706.
176. Black, N., et al., *Consensus development methods: a review of best practice in creating clinical guidelines.* J Health Serv Res Policy, 1999. **4**(4): p. 236-48.
177. Akins, R.B., H. Tolson, and B.R. Cole, *Stability of response characteristics of a Delphi panel: application of bootstrap data expansion.* BMC Med Res Methodol, 2005. **5**: p. 37.
178. Linstone, H.A. and M. Turoff, *The delphi method*. 1975: Addison-Wesley Reading, MA.
179. Jones, J., C. Sanderson, and N. Black, *What will happen to the quality of care with fewer junior doctors? A Delphi study of consultant physicians' views.* J R Coll Physicians Lond, 1992. **26**(1): p. 36-40.
180. Brooks, K.W., *Delphi technique: Expanding applications.* North Central Association Quarterly, 1979. **53**(3): p. 377-85.
181. Custer, R.L., J.A. Scarcella, and B.R. Stewart, *The modified Delphi technique-A rotational modification.* 1999.
182. Diamond, I.R., et al., *Defining consensus: A systematic review recommends methodologic criteria for reporting of Delphi studies.* Journal of Clinical Epidemiology, 2014. **67**(4): p. 401-409.
183. Weaver, W.T., *The Delphi forecasting method.* The Phi Delta Kappan, 1971. **52**(5): p. 267-271.
184. Hasson, F., S. Keeney, and H. McKenna, *Research guidelines for the Delphi survey technique.* Journal of advanced nursing, 2000. **32**(4): p. 1008-1015.
185. Pill, J., *The Delphi method: substance, context, a critique and an annotated bibliography. .* Socioecon Plan Sci, 1971. **5**: p. 57-71.
186. Duffield, C., *The Delphi technique: a comparison of results obtained using two expert panels.* Int J Nurs Stud, 1993. **30**(3): p. 227-37.
187. Lemmer, B., *Successive surveys of an expert panel: research in decision-making with health visitors.* J Adv Nurs, 1998. **27**(3): p. 538-45.
188. Bond, S. and J. Bond, *A Delphi survey of clinical nursing research priorities.* J Adv Nurs, 1982. **7**(6): p. 565-75.

189. Boulkedid, R., et al., *Using and reporting the Delphi method for selecting healthcare quality indicators: a systematic review.* PLoS One, 2011. **6**(6): p. e20476.
190. Sinha, I.P., R.L. Smyth, and P.R. Williamson, *Using the Delphi technique to determine which outcomes to measure in clinical trials: recommendations for the future based on a systematic review of existing studies.* PLoS Med, 2011. **8**(1): p. e1000393.
191. Diamond, I.R., et al., *Defining consensus: a systematic review recommends methodologic criteria for reporting of Delphi studies.* J Clin Epidemiol, 2014. **67**(4): p. 401-9.
192. Foth, T., et al., *The use of Delphi and Nominal Group Technique in nursing education: A review.* Int J Nurs Stud, 2016. **60**: p. 112-20.
193. Waggoner, J., J.D. Carline, and S.J. Durning, *Is There a Consensus on Consensus Methodology? Descriptions and Recommendations for Future Consensus Research.* Acad Med, 2016. **91**(5): p. 663-8.
194. Rietjens, J.A.C., et al., *Definition and recommendations for advance care planning: an international consensus supported by the European Association for Palliative Care.* Lancet Oncol, 2017. **18**(9): p. e543-e551.
195. De Vet, E., et al., *Determinants of forward stage transitions: a Delphi study.* Health Educ Res, 2005. **20**(2): p. 195-205.
196. Biondo, P.D., et al., *Applying the Delphi process to palliative care tool development: lessons learned.* Support Care Cancer, 2008. **16**(8): p. 935-42.
197. Rahaghi, F.F., et al., *Delphi consensus recommendations for a treatment algorithm in pulmonary sarcoidosis.* Eur Respir Rev, 2020. **29**(155).
198. Distler, O., et al., *Defining appropriate outcome measures in pulmonary arterial hypertension related to systemic sclerosis: a Delphi consensus study with cluster analysis.* Arthritis Rheum, 2008. **59**(6): p. 867-75.
199. Huscher, D., et al., *Interactions between rheumatologists and cardio-/pulmonologists in the assessment and use of outcome measures in pulmonary arterial hypertension related to systemic sclerosis.* Clin Exp Rheumatol, 2010. **28**(2 Suppl 58): p. S47-52.
200. Saketkoo, L.A., et al., *Connective tissue disease related interstitial lung diseases and idiopathic pulmonary fibrosis: provisional core sets of domains and instruments for use in clinical trials.* Thorax, 2014. **69**(5): p. 428-36.
201. Rahaghi, F.F., et al., *Recommendations for the use of oral treprostinil in clinical practice: a Delphi consensus project pulmonary circulation.* Pulmonary circulation, 2017. **7**(1): p. 167-174.
202. Rahaghi, F.F., et al., *Recommendations for the clinical management of patients receiving macitentan for pulmonary arterial hypertension (PAH): a Delphi consensus document.* Pulmonary circulation, 2017. **7**(3): p. 702-711.
203. Woudenberg, F., *An evaluation of Delphi.* Technological forecasting and social change, 1991. **40**(2): p. 131-150.
204. Graham, B., G. Regehr, and J.G. Wright, *Delphi as a method to establish consensus for diagnostic criteria.* Journal of clinical epidemiology, 2003. **56**(12): p. 1150-1156.
205. Norman, G., *Likert scales, levels of measurement and the "laws" of statistics.* Adv Health Sci Educ Theory Pract, 2010. **15**(5): p. 625-32.
206. Bould, M.D. and M.R. Sury, *Defining awakening from anesthesia in neonates: a consensus study.* Paediatr Anaesth, 2011. **21**(4): p. 359-63.
207. Loughlin, K.G. and L.F. Moore, *Using Delphi to achieve congruent objectives and activities in a pediatrics department.* J Med Educ, 1979. **54**(2): p. 101-6.
208. Melnyk, B.M. and E. Fineout-Overholt, *Evidence-based practice in nursing & healthcare: A guide to best practice*. 2011: Lippincott Williams & Wilkins.

209. Arksey, H. and L. O'Malley, *Scoping studies: towards a methodological framework.* International journal of social research methodology, 2005. **8**(1): p. 19-32.
210. Simes, R.J., *An improved Bonferroni procedure for multiple tests of significance.* Biometrika, 1986. **73**(3): p. 751-754.
211. StataCorp, *Stata Statistical Software: Release 17*. 2021, StataCorp LLC: College Station, TX.
212. Melloul, E., et al., *Guidelines for Perioperative Care for Liver Surgery: Enhanced Recovery After Surgery (ERAS) Society Recommendations.* World J Surg, 2016. **40**(10): p. 2425-40.
213. Guckenberger, M., et al., *Characterisation and classification of oligometastatic disease: a European Society for Radiotherapy and Oncology and European Organisation for Research and Treatment of Cancer consensus recommendation.* Lancet Oncol, 2020. **21**(1): p. e18-e28.
214. Gordijn, S.J., et al., *Consensus definition of fetal growth restriction: a Delphi procedure.* Ultrasound Obstet Gynecol, 2016. **48**(3): p. 333-9.
215. Kanate, A.S., et al., *Maintenance Therapies for Hodgkin and Non-Hodgkin Lymphomas After Autologous Transplantation: A Consensus Project of ASBMT, CIBMTR, and the Lymphoma Working Party of EBMT.* JAMA Oncol, 2019. **5**(5): p. 715-722.
216. Merlo, A., et al., *Appropriateness of admission to rehabilitation: definition of a set of criteria and rules through the application of the Delphi method.* Eur J Phys Rehabil Med, 2020. **56**(5): p. 537-546.
217. Rodríguez-Mañas, L., et al., *Searching for an operational definition of frailty: a Delphi method based consensus statement: the frailty operative definition-consensus conference project.* J Gerontol A Biol Sci Med Sci, 2013. **68**(1): p. 62-7.
218. Shankar-Hari, M., et al., *Developing a New Definition and Assessing New Clinical Criteria for Septic Shock: For the Third International Consensus Definitions for Sepsis and Septic Shock (Sepsis-3).* Jama, 2016. **315**(8): p. 775-87.
219. Tawfik, E.A., et al., *Guidelines for neuromuscular ultrasound training.* Muscle Nerve, 2019. **60**(4): p. 361-366.
220. Beune, I.M., et al., *Consensus Based Definition of Growth Restriction in the Newborn.* J Pediatr, 2018. **196**: p. 71-76.e1.
221. Galindo-Zavala, R., et al., *Expert panel consensus recommendations for diagnosis and treatment of secondary osteoporosis in children.* Pediatr Rheumatol Online J, 2020. **18**(1): p. 20.
222. Tieu, J., et al., *Rituximab for maintenance of remission in ANCA-associated vasculitis: expert consensus guidelines.* Rheumatology (Oxford), 2020. **59**(4): p. e24-e32.
223. Diermeier, T., et al., *Treatment After Anterior Cruciate Ligament Injury: Panther Symposium ACL Treatment Consensus Group.* Orthop J Sports Med, 2020. **8**(6): p. 2325967120931097.
224. Papamichael, K., et al., *Appropriate Therapeutic Drug Monitoring of Biologic Agents for Patients With Inflammatory Bowel Diseases.* Clin Gastroenterol Hepatol, 2019. **17**(9): p. 1655-1668.e3.
225. Tucci, D.L., et al., *Clinical Consensus Statement: Balloon Dilation of the Eustachian Tube.* Otolaryngol Head Neck Surg, 2019. **161**(1): p. 6-17.
226. Pittiruti, M., et al., *Evidence-based criteria for the choice and the clinical use of the most appropriate lock solutions for central venous catheters (excluding dialysis catheters): a GAVeCeLT consensus.* J Vasc Access, 2016. **17**(6): p. 453-464.
227. Lau, C.S., et al., *2018 update of the APLAR recommendations for treatment of rheumatoid arthritis.* Int J Rheum Dis, 2019. **22**(3): p. 357-375.
228. Abu Hilal, M., et al., *The Southampton Consensus Guidelines for Laparoscopic Liver Surgery: From Indication to Implementation.* Ann Surg, 2018. **268**(1): p. 11-18.
229. Sano, Y., et al., *Narrow-band imaging (NBI) magnifying endoscopic classification of colorectal tumors proposed by the Japan NBI Expert Team.* Dig Endosc, 2016. **28**(5): p. 526-33.

230. Aringer, M., et al., *2019 European League Against Rheumatism/American College of Rheumatology Classification Criteria for Systemic Lupus Erythematosus.* Arthritis Rheumatol, 2019. **71**(9): p. 1400-1412.
231. Hill, A.T., et al., *Pulmonary exacerbation in adults with bronchiectasis: a consensus definition for clinical research.* Eur Respir J, 2017. **49**(6).
232. Mimura, T., et al., *Evidence-based clinical practice guideline for adult Still's disease.* Mod Rheumatol, 2018. **28**(5): p. 736-757.
233. Antonini, A., et al., *Developing consensus among movement disorder specialists on clinical indicators for identification and management of advanced Parkinson's disease: a multi-country Delphi-panel approach.* Curr Med Res Opin, 2018. **34**(12): p. 2063-2073.
234. Allen, J., et al., *Severe Neurological Impairment: A delphi consensus-based definition.* Eur J Paediatr Neurol, 2020. **29**: p. 81-86.
235. Ludlow, C.L., et al., *Consensus-Based Attributes for Identifying Patients With Spasmodic Dysphonia and Other Voice Disorders.* JAMA Otolaryngol Head Neck Surg, 2018. **144**(8): p. 657-665.
236. Liyanage, H., et al., *Artificial Intelligence in Primary Health Care: Perceptions, Issues, and Challenges.* Yearb Med Inform, 2019. **28**(1): p. 41-46.
237. Haesler, E., et al., *Clinical indicators of wound infection and biofilm: reaching international consensus.* J Wound Care, 2019. **28**(Sup3b): p. s4-s12.
238. Singer, M., et al., *The Third International Consensus Definitions for Sepsis and Septic Shock (Sepsis-3).* Jama, 2016. **315**(8): p. 801-10.
239. Gomes, J.A., et al., *Global consensus on keratoconus and ectatic diseases.* Cornea, 2015. **34**(4): p. 359-69.
240. Khalil, A., et al., *Consensus definition and essential reporting parameters of selective fetal growth restriction in twin pregnancy: a Delphi procedure.* Ultrasound Obstet Gynecol, 2019. **53**(1): p. 47-54.
241. D'Souza, N., et al., *Definition of the Rectum: An International, Expert-based Delphi Consensus.* Ann Surg, 2019. **270**(6): p. 955-959.
242. Maverakis, E., et al., *Diagnostic Criteria of Ulcerative Pyoderma Gangrenosum: A Delphi Consensus of International Experts.* JAMA Dermatol, 2018. **154**(4): p. 461-466.
243. Zanker, J., et al., *Establishing an Operational Definition of Sarcopenia in Australia and New Zealand: Delphi Method Based Consensus Statement.* J Nutr Health Aging, 2019. **23**(1): p. 105-110.
244. Strober, B., et al., *Recategorization of psoriasis severity: Delphi consensus from the International Psoriasis Council.* J Am Acad Dermatol, 2020. **82**(1): p. 117-122.
245. Fernández-de-Las-Peñas, C. and J. Dommerholt, *International Consensus on Diagnostic Criteria and Clinical Considerations of Myofascial Trigger Points: A Delphi Study.* Pain Med, 2018. **19**(1): p. 142-150.
246. Barrios, M., et al., *Consensus in the delphi method: What makes a decision change?* Technological Forecasting and Social Change, 2021. **163**: p. 120484.
247. Baron-Epel, O., et al., *Extreme and acquiescence bias in a bi-ethnic population.* Eur J Public Health, 2010. **20**(5): p. 543-8.
248. Krosnick, J.A., *Survey research.* Annu Rev Psychol, 1999. **50**: p. 537-67.
249. Kuru, O. and J. Pasek, *Improving social media measurement in surveys: Avoiding acquiescence bias in Facebook research.* Computers in Human Behavior, 2016. **57**: p. 82-92.
250. Smith, P.B., *Acquiescent Response Bias as an Aspect of Cultural Communication Style.* Journal of Cross-Cultural Psychology, 2004. **35**(1): p. 50-61.

251. Bachman, J.G. and P.M. O'Malley, *Yea-Saying, Nay-Saying, and Going to Extremes: Black-White Differences in Response Styles.* Public Opinion Quarterly, 1984. **48**(2): p. 491-509.
252. Pasek, J. and J.A. Krosnick, *Optimizing survey questionnaire design in political science: Insights from psychology.* Oxford handbook of American elections and political behavior, 2010: p. 27-50.
253. Green, S.M., et al., *An international multidisciplinary consensus statement on fasting before procedural sedation in adults and children.* Anaesthesia, 2020. **75**(3): p. 374-385.
254. Hassett, M.J., M.R. Somerfield, and S.H. Giordano, *Management of Male Breast Cancer: ASCO Guideline Summary.* JCO Oncol Pract, 2020. **16**(8): p. e839-e843.
255. Klinger, J.R., et al., *Therapy for Pulmonary Arterial Hypertension in Adults: Update of the CHEST Guideline and Expert Panel Report.* Chest, 2019. **155**(3): p. 565-586.
256. Hussler, C., P. Muller, and P. Rondé, *Is diversity in Delphi panelist groups useful? Evidence from a French forecasting exercise on the future of nuclear energy.* Technological Forecasting and Social Change, 2011. **78**(9): p. 1642-1653.
257. Makkonen, M., T. Hujala, and J. Uusivuori, *Policy experts' propensity to change their opinion along Delphi rounds.* Technological Forecasting and Social Change, 2016. **109**: p. 61-68.
258. Bolger, F., et al., *Does the Delphi process lead to increased accuracy in group-based judgmental forecasts or does it simply induce consensus amongst judgmental forecasters?* Technological Forecasting and Social Change, 2011. **78**(9): p. 1671-1680.
259. Meijering, J.V. and H. Tobi, *The effects of feeding back experts' own initial ratings in Delphi studies: A randomized trial.* International Journal of Forecasting, 2018. **34**(2): p. 216-224.
260. Rowe, G., G. Wright, and A. McColl, *Judgment change during Delphi-like procedures: The role of majority influence, expertise, and confidence.* Technological Forecasting and Social Change, 2005. **72**(4): p. 377-399.
261. Brookes, S.T., et al., *Three nested randomized controlled trials of peer-only or multiple stakeholder group feedback within Delphi surveys during core outcome and information set development.* Trials, 2016. **17**(1): p. 1-14.
262. Harman, N.L., et al., *The importance of integration of stakeholder views in core outcome set development: otitis media with effusion in children with cleft palate.* PloS one, 2015. **10**(6): p. e0129514.
263. Turnbull, A.E., et al., *A survey of Delphi panelists after core outcome set development revealed positive feedback and methods to facilitate panel member participation.* Journal of clinical epidemiology, 2018. **102**: p. 99-106.
264. Rowe, G. and G. Wright, *The impact of task characteristics on the performance of structured group forecasting techniques.* International Journal of Forecasting, 1996. **12**(1): p. 73-89.
265. Toor, M., *What is acquiescence bias and how can you stop it?*, in *QualtricsXM*. 2020.
266. *2009-2010 National Survey of Children with Special Healthcare Needs*, Bureau of Maternal and Child Health. 2011, Agency of Health Resources and Service Administration.
267. *Complex Care for Kids Ontario Standard Operation Definition*. 2019, Provincial Council for Maternal and Child Health.
268. Berry, J.G., et al., *Hospital Utilization and Characteristics of Patients Experiencing Recurrent Readmissions Within Children's Hospitals.* JAMA, 2011. **305**(7): p. 682-690.
269. Cohen, E., et al., *Patterns and costs of health care use of children with medical complexity.* Pediatrics, 2012. **130**(6): p. e1463-e1470.
270. Kuo, D.Z. and A.J. Houtrow, *Recognition and Management of Medical Complexity.* Pediatrics, 2016. **138**(6).
271. Rehm, R.S. and J.F. Bradley, *Normalization in families raising a child who is medically fragile/technology dependent and developmentally delayed.* Qual Health Res, 2005. **15**(6): p. 807-20.

272. Rehm, R.S. and J.F. Bradley, *The search for social safety and comfort in families raising children with complex chronic conditions.* J Fam Nurs, 2005. **11**(1): p. 59-78.
273. *Children and Youth With Medical Complexity in Canada.* Canadian Institute for Health Information. 2020: Ottawa, ON.
274. Perrin, E.C., et al., *Issues involved in the definition and classification of chronic health conditions.* Pediatrics, 1993. **91**(4): p. 787-93.
275. Hughes, D.C., et al., *Improving children's access to health care: the role of decategorization.* Bull N Y Acad Med, 1996. **73**(2): p. 237-54.
276. Simon, T.D., et al., *Pediatric medical complexity algorithm: a new method to stratify children by medical complexity.* Pediatrics, 2014. **133**(6): p. e1647-e1654.
277. Klitzner, T.S., L.A. Rabbitt, and R.K.R. Chang, *Benefits of Care Coordination for Children with Complex Disease: A Pilot Medical Home Project in a Resident Teaching Clinic.* Journal of Pediatrics, 2010. **156**(6): p. 1006-1010.
278. Foster, C.C., et al., *Social Determinants of Health and Emergency and Hospital Use by Children With Chronic Disease.* Hosp Pediatr, 2020. **10**(6): p. 471-480.
279. Council on Community Pediatrics. *Poverty and Child Health in the United States.* Pediatrics, 2016. **137**(4).
280. Fleming, J., et al., *Impact on the family of children who are technology dependent and cared for in the home.* Pediatr Nurs, 1994. **20**(4): p. 379-88.
281. O'Brien, M.E., *Living in a house of cards: family experiences with long-term childhood technology dependence.* J Pediatr Nurs, 2001. **16**(1): p. 13-22.
282. Clegg, A., et al., *Frailty in elderly people.* Lancet, 2013. **381**(9868): p. 752-62.
283. Fried, L.P., et al., *Frailty in older adults: evidence for a phenotype.* J Gerontol A Biol Sci Med Sci, 2001. **56**(3): p. M146-56.
284. Abbasi, M., et al., *Identification and management of frailty in the primary care setting.* Cmaj, 2018. **190**(38): p. E1134-e1140.
285. Rockwood, K., et al., *A brief clinical instrument to classify frailty in elderly people.* Lancet, 1999. **353**(9148): p. 205-6.
286. Winograd, C.H., et al., *Screening for frailty: criteria and predictors of outcomes.* J Am Geriatr Soc, 1991. **39**(8): p. 778-84.
287. Campbell, A.J. and D.M. Buchner, *Unstable disability and the fluctuations of frailty.* Age Ageing, 1997. **26**(4): p. 315-8.
288. Buchner, D.M. and E.H. Wagner, *Preventing frail health.* Clin Geriatr Med, 1992. **8**(1): p. 1-17.
289. Chin, A.P.M.J., et al., *How to select a frail elderly population? A comparison of three working definitions.* J Clin Epidemiol, 1999. **52**(11): p. 1015-21.
290. Evans, W.J., *What is sarcopenia?* J Gerontol A Biol Sci Med Sci, 1995. **50 Spec No**: p. 5-8.
291. Chandler, J.M. and E.C. Hadley, *Exercise to improve physiologic and functional performance in old age.* Clin Geriatr Med, 1996. **12**(4): p. 761-84.
292. Cerreta, F., H.G. Eichler, and G. Rasi, *Drug policy for an aging population--the European Medicines Agency's geriatric medicines strategy.* N Engl J Med, 2012. **367**(21): p. 1972-4.
293. *The 2012 Ageing Report: Economic and budgetary projections for the 27 EU Member States (2010-2060)*. 2012, European Commission: Brussels, Belgium.
294. Di Pollina, L., et al., *Integrated care at home reduces unnecessary hospitalizations of community-dwelling frail older adults: a prospective controlled trial.* BMC Geriatr, 2017. **17**(1): p. 53.
295. Landi, F., et al., *Physical activity and mortality in frail, community-living elderly patients.* J Gerontol A Biol Sci Med Sci, 2004. **59**(8): p. 833-7.

296. Poli, S., et al., *Frailty is associated with socioeconomic and lifestyle factors in community-dwelling older subjects.* Aging Clin Exp Res, 2017. **29**(4): p. 721-728.
297. Hoover, M., et al., *Validation of an index to estimate the prevalence of frailty among community-dwelling seniors.* Health Rep, 2013. **24**(9): p. 10-7.
298. Gill, T.M., et al., *Trajectories of disability in the last year of life.* N Engl J Med, 2010. **362**(13): p. 1173-80.
299. Covinsky, K.E., et al., *The last 2 years of life: functional trajectories of frail older people.* J Am Geriatr Soc, 2003. **51**(4): p. 492-8.
300. Morley, J.E., et al., *Frailty consensus: a call to action.* J Am Med Dir Assoc, 2013. **14**(6): p. 392-7.
301. Dent, E., P. Kowal, and E.O. Hoogendijk, *Frailty measurement in research and clinical practice: A review.* Eur J Intern Med, 2016. **31**: p. 3-10.
302. Bergman, H., et al., *Frailty: an emerging research and clinical paradigm--issues and controversies.* J Gerontol A Biol Sci Med Sci, 2007. **62**(7): p. 731-7.
303. de Vries, N.M., et al., *Outcome instruments to measure frailty: a systematic review.* Ageing Res Rev, 2011. **10**(1): p. 104-14.
304. Clegg, A., L. Rogers, and J. Young, *Diagnostic test accuracy of simple instruments for identifying frailty in community-dwelling older people: a systematic review.* Age Ageing, 2015. **44**(1): p. 148-52.
305. Pialoux, T., J. Goyard, and B. Lesourd, *Screening tools for frailty in primary health care: a systematic review.* Geriatr Gerontol Int, 2012. **12**(2): p. 189-97.
306. Bergman, H., D. Hogan, and S. Karunananthan, *Frailty: A clinically relevant concept?* Canadian Journal of Geriatrics, 2008. **11**: p. 124-128.
307. Sourial, N., et al., *Contribution of frailty markers in explaining differences among individuals in five samples of older persons.* J Gerontol A Biol Sci Med Sci, 2012. **67**(11): p. 1197-204.
308. Drubbel, I., et al., *Screening for frailty in primary care: a systematic review of the psychometric properties of the frailty index in community-dwelling older people.* BMC Geriatr, 2014. **14**: p. 27.
309. Rockwood, K. and A. Mitnitski, *Frailty defined by deficit accumulation and geriatric medicine defined by frailty.* Clin Geriatr Med, 2011. **27**(1): p. 17-26.
310. Allen, J., E. Molloy, and D. McDonald, *Severe neurological impairment: a review of the definition.* Dev Med Child Neurol, 2020. **62**(3): p. 277-282.
311. Morris, C., et al., *Towards a definition of neurodisability: a Delphi survey.* Dev Med Child Neurol, 2013. **55**(12): p. 1103-8.
312. Agrawal, R., et al., *Trends in health care spending for children in medicaid with high resource use.* Pediatrics, 2016. **138**(4).
313. de Wit, M., C. Cooper, and J.Y. Reginster, *Practical guidance for patient-centred health research.* Lancet, 2019. **393**(10176): p. 1095-1096.
314. Edwards, P., et al., *Identification of randomized controlled trials in systematic reviews: accuracy and reliability of screening records.* Stat Med, 2002. **21**(11): p. 1635-40.
315. Waffenschmidt, S., et al., *Single screening versus conventional double screening for study selection in systematic reviews: a methodological systematic review.* BMC Medical Research Methodology, 2019. **19**(1): p. 132.
316. Buscemi, N., et al., *Single data extraction generated more errors than double data extraction in systematic reviews.* Journal of Clinical Epidemiology, 2006. **59**(7): p. 697-703.
317. Elliott, V., *Thinking about the Coding Process in Qualitative Data Analysis.* The Qualitative Report, 2018. **23**(11): p. 2850-2861.
318. Knoll, M., et al., *The use of incentives in vulnerable populations for a telephone survey: a randomized controlled trial.* BMC Res Notes, 2012. **5**: p. 572.

319. Shen, S., et al., *How and why should we engage parents as co-researchers in health research? A scoping review of current practices.* Health Expect, 2017. **20**(4): p. 543-554.
320. U.S. Congress, Office of Technology Assessment, *Technology-Dependent Children: Hospital v. Home Care—A Technical Memorandum*. 1987, US Government Printing Office Washington, DC.
321. *The Child and Adolescent Health Measurement Initiative.* June 19, 2019]; Available from: http://www.childhealthdata.org/learn/NS-CSHCN.
322. Aboneh, E.A. and M.A. Chui, *Care coordination, medical complexity, and unmet need for prescription medications among children with special health care needs.* Research in Social and Administrative Pharmacy, 2017. **13**(3): p. 524-529.
323. Abbott, L., R. McConkey, and M. Dobbins, *Key players in inclusion: Are we meeting the professional needs of learning support assistants for pupils with complex needs?* European Journal of Special Needs Education, 2011. **26**(2): p. 215-231.
324. Adams, S., et al., *Exploring the usefulness of comprehensive care plans for children with medical complexity (CMC): A qualitative study.* BMC Pediatrics, 2013. **13**(1).
325. Adams, S., et al., *Care maps for children with medical complexity.* Developmental Medicine and Child Neurology, 2017. **59**(12): p. 1299-1306.
326. Afolabi, T.M. and K.A. Fairman, *Pediatric Home Parenteral Nutrition: Indications and Short-Term Outcomes in a Large National Sample of Commercially Insured Children and Adolescents.* Nutrition in Clinical Practice, 2019. **34**(2): p. 242-249.
327. Agrawal, R., et al., *Rate of spending on chronic conditions among medicaid and CHIP recipients.* Pediatrics, 2014. **134**(1): p. e80-e87.
328. Agarwal, A., et al., *Transition of respiratory technology dependent patients from pediatric to adult pulmonology care.* Pediatric Pulmonology, 2015. **50**(12): p. 1294-1300.
329. Altman, L., et al., *A qualitative study of health care providers' perceptions and experiences of working together to care for children with medical complexity (CMC).* BMC Health Services Research, 2018. **18**(1).
330. Amin, R., et al., *Pediatric long-term home mechanical ventilation: Twenty years of follow-up from one Canadian center.* Pediatric Pulmonology, 2014. **49**(8): p. 816-824.
331. An, R., *Unmet mental health care needs in U.S. children with medical complexity, 2005-2010.* Journal of Psychosomatic Research, 2016. **82**: p. 1-3.
332. Ananth, P., et al., *Hospital use in the last year of life for children with life-threatening complex chronic conditions.* Pediatrics, 2015. **136**(5): p. 938-946.
333. Arthur, K.C., et al., *Quality of Care for Children With Medical Complexity: An Analysis of Continuity of Care as a Potential Quality Indicator.* Academic Pediatrics, 2018. **18**(6): p. 669-676.
334. Aruda, M.M., M. Kelly, and K. Newinsky, *Unmet needs of children with special health care needs in a specialized day school setting.* Journal of School Nursing, 2011. **27**(3): p. 209-218.
335. Avritscher, E.B.C., et al., *Post-Trial Sustainability and Scalability of the Benefits of a Medical Home for High-Risk Children with Medical Complexity.* Journal of Pediatrics, 2019. **206**: p. 232-239.e3.
336. Balistreri, K.S., *Food insufficiency and children with special healthcare needs.* Public Health, 2019. **167**: p. 55-61.
337. Barnert, E.S., et al., *Experts' Perspectives Toward a Population Health Approach for Children With Medical Complexity.* Academic Pediatrics, 2017. **17**(6): p. 672-677.
338. Benneyworth, B.D., et al., *Inpatient health care utilization for children dependent on long-term mechanical ventilation.* Pediatrics, 2011. **127**(6): p. e1533-e1541.
339. Berry, J.G., et al., *Inpatient growth and resource use in 28 children's hospitals: A longitudinal, multi-institutional study.* JAMA Pediatrics, 2013. **167**(2): p. 170-177.

340. Berry, J.G., et al., *Children with medical complexity and medicaid: Spending and cost savings.* Health Affairs, 2014. **33**(12): p. 2199-2206.
341. Berry, J.G., et al., *Pediatric hospital discharges to home health and postacute facility care: A national study.* JAMA Pediatrics, 2016. **170**(4): p. 326-333.
342. Berry, J.G., et al., *Predicting postoperative physiologic decline after surgery.* Pediatrics, 2019. **143**(4).
343. Berry, J.G., et al., *Association of Home Respiratory Equipment and Supply Use with Health Care Resource Utilization in Children.* Journal of Pediatrics, 2019. **207**: p. 169-175.e2.
344. Bowles, J.D., et al., *Infants with technology dependence.* Advances in Neonatal Care, 2016. **16**(6): p. 424-429.
345. Brady, P.W., et al., *Prevalence and nature of adverse medical device events in hospitalized children.* Journal of Hospital Medicine, 2013. **8**(7): p. 390-393.
346. Breen, C., et al., *Significant reductions in tertiary hospital encounters and less travel for families after implementation of Paediatric Care Coordination in Australia 11 Medical and Health Sciences 1117 Public Health and Health Services.* BMC Health Services Research, 2018. **18**(1).
347. Cady, R.G., et al., *Meeting the Needs of Children with Medical Complexity Using a Telehealth Advanced Practice Registered Nurse Care Coordination Model.* Maternal and Child Health Journal, 2015. **19**(7): p. 1497-1506.
348. Caicedo, C., *Families With Special Needs Children: Family Health, Functioning, and Care Burden.* Journal of the American Psychiatric Nurses Association, 2014. **20**(6): p. 398-407.
349. Caicedo, C., *Health and functioning of families of children with special health care needs cared for in home care, long-term care, and medical day care settings.* Journal of Developmental and Behavioral Pediatrics, 2015. **36**(5): p. 352-361.
350. Caicedo, C., *Children With Special Health Care Needs: Child Health and Functioning Outcomes and Health Care Service Use.* Journal of Pediatric Health Care, 2016. **30**(6): p. 590-598.
351. Carnevale, F.A., et al., *Daily living with distress and enrichment: The moral experience of families with ventilator-assisted children at home.* Pediatrics, 2006. **117**(1): p. e48-e60.
352. Castro-Codesal, M.L., et al., *Longitudinal changes in clinical characteristics and outcomes for children using long-term non-invasive ventilation.* PLoS ONE, 2018. **13**(1).
353. Chan, T., et al., *Pediatric Critical Care Resource Use by Children with Medical Complexity.* Journal of Pediatrics, 2016. **177**: p. 197-203.e1.
354. Chien, A.T., et al., *Care Quality and Spending Among Commercially Insured Children With Disabilities.* Academic Pediatrics, 2019. **19**(3): p. 291-299.
355. Cohen, E., et al., *The impact of a complex care clinic in a children's hospital.* Child: Care, Health and Development, 2010. **36**(4): p. 574-582.
356. Cohen, E., et al., *Residential movement patterns of families of young children with chronic conditions in Ontario, Canada: A population-based cohort study.* International Journal for Equity in Health, 2013. **12**(1).
357. Coller, R.J., et al., *Medical Complexity among Children with Special Health Care Needs: A Two-Dimensional View.* Health Services Research, 2016. **51**(4): p. 1644-1669.
358. Coller, R.J., et al., *Hospitalizations for Ambulatory Care-Sensitive Conditions among Children with Chronic and Complex Diseases.* Journal of Pediatrics, 2018. **194**: p. 218-224.
359. Coller, R.J., et al., *Complex care hospital use and postdischarge coaching: A randomized controlled trial.* Pediatrics, 2018. **142**(2).
360. Donohue, P.K., et al., *"it's Relentless": Providers' experience of pediatric chronic critical illness.* Journal of Palliative Medicine, 2018. **21**(7): p. 940-946.
361. Desai, A.D., et al., *Caregiver Perceptions of Hospital to Home Transitions According to Medical Complexity: A Qualitative Study.* Academic Pediatrics, 2016. **16**(2): p. 136-144.

362. Dodds, C.B., et al., *The effect of supported physical activity on parental-reported health-related quality of life in children with medical complexity.* Journal of Pediatric Rehabilitation Medicine, 2015. **8**(2): p. 83-95.
363. Dodds, C.B., et al., *The effect of supported physical activity on parental-reported sleep qualities and pain severity in children with medical complexity.* Journal of Pediatric Rehabilitation Medicine, 2016. **9**(3): p. 195-206.
364. Edwards, J.D., et al., *Repeated critical illness and unplanned readmissions within 1 year to PICUs.* Critical Care Medicine, 2017. **45**(8): p. 1276-1284.
365. Elias, E.R., et al., *Home care of children and youth with complex health care needs and technology dependencies.* Pediatrics, 2012. **129**(5): p. 996-1005.
366. Ellzey, A., et al., *Parent perceptions of quality of life and healthcare satisfaction for children with medical complexity.* Journal of Pediatric Rehabilitation Medicine, 2015. **8**(2): p. 97-104.
367. Fayed, N., et al., *Family-provider consensus outcomes for children with medical complexity.* Developmental Medicine and Child Neurology, 2018.
368. Fortin, K., S. Kwon, and M.C. Pierce, *Characteristics of Children Reported to Child Protective Services for Medical Neglect.* Hospital pediatrics, 2016. **6**(4): p. 204-210.
369. Foster, C.C., R. Mangione-Smith, and T.D. Simon, *Caring for Children with Medical Complexity: Perspectives of Primary Care Providers.* Journal of Pediatrics, 2017. **182**: p. 275-282.e4.
370. Friedman, S.L., et al., *Out-of-home placement for children and adolescents with disabilities-Addendum: Care options for Children and adolescents with disabilities and medical complexity.* Pediatrics, 2016. **138**(6).
371. Feudtner, C., D.L. DiGiuseppe, and J.M. Neff, *Hospital care for children and young adults in the last year of life: A population-based study.* BMC Medicine, 2003. **1**.
372. Feudtner, C., et al., *Technology-dependency among patients discharged from a children's hospital: A retrospective cohort study.* BMC Pediatrics, 2005. **5**.
373. Feudtner, C., et al., *Pediatric complex chronic conditions classification system version 2: Updated for ICD-10 and complex medical technology dependence and transplantation.* BMC Pediatrics, 2014. **14**(1).
374. Gay, J.C., et al., *Home health nursing care and hospital use for medically complex children.* Pediatrics, 2016. **138**(5).
375. Geremia, C., et al., *Totally implantable venous access devices in children with medical complexity: Preliminary data from a tertiary care hospital.* Journal of Vascular Access, 2017. **18**(5): p. 426-429.
376. Giambra, B.K., et al., *Identification of hospitalized tracheostomy and ventilator dependent patients in administrative data.* Pediatric Pulmonology, 2018. **53**(7): p. 973-978.
377. Gidengil, C., et al., *Development and implementation of the family experiences with coordination of care survey quality measures.* Academic pediatrics, 2017. **17**(8): p. 863-870.
378. Golden, S.L. and S. Nageswaran, *Caregiver voices: Coordinating care for children with complex chronic conditions.* Clinical Pediatrics, 2012. **51**(8): p. 723-729.
379. Gold, J.M., et al., *Long length of hospital stay in children with medical complexity.* Journal of Hospital Medicine, 2016. **11**(11): p. 750-756.
380. Goossens, E., et al., *Predictors of care gaps in adolescents with complex chronic condition transitioning to adulthood.* Pediatrics, 2016. **137**(4).
381. Graham, R.J., et al., *Pediatric specialty care model for management of chronic respiratory failure: Cost and savings implications and misalignment with payment models.* Pediatric Critical Care Medicine, 2018. **19**(5): p. 412-420.
382. Haarbauer-Krupa, J., et al., *Readiness for transition and health-care satisfaction in adolescents with complex medical conditions.* Child: Care, Health and Development, 2019. **45**(3): p. 463-471.

383. Hessels, A.J., et al., *National Testing of the Nursing-Kids Intensity of Care Survey for Pediatric Long-term Care.* Journal of Pediatric Nursing, 2017. **37**: p. 86-90.
384. Hoffman, A. and I.A. Larson, *Primary care preparation for children with medical complexity for emergency encounters in the community.* Pediatric Annals, 2018. **47**(3): p. e102-e105.
385. Hudson, S.M., et al., *Factors influencing hospital admissions and emergency department visits among children with complex chronic conditions: A qualitative study of parents' and providers' perspectives.* Comprehensive Child and Adolescent Nursing, 2014. **37**(1): p. 61-80.
386. Joly, E., *Access to services for young adults with medical complexity.* Nursing Ethics, 2017. **24**(3): p. 329-336.
387. Keilty, K., et al., *Sleep disturbance in family caregivers of children who depend on medical technology.* Archives of Disease in Childhood, 2018. **103**(2): p. 137-142.
388. Kun, S.S., et al., *Hospital readmissions for newly discharged pediatric home mechanical ventilation patients.* Pediatric Pulmonology, 2012. **47**(4): p. 409-414.
389. Kuo, D.Z., et al., *Recognition and management of medical complexity.* Pediatrics, 2016. **138**(6).
390. Kuo, D.Z., et al., *A national profile of caregiver challenges among more medically complex children with special health care needs.* Archives of Pediatrics and Adolescent Medicine, 2011. **165**(11): p. 1020-1026.
391. Kuo, D.Z., et al., *Inequities in health care needs for children with medical complexity.* Health Affairs, 2014. **33**(12): p. 2190-2198.
392. Kuo, D.Z., et al., *Variation in Child Health Care Utilization by Medical Complexity.* Maternal and Child Health Journal, 2014. **19**(1): p. 40-48.
393. Liberman, D.B., et al., *Early introduction of palliative care and advanced care planning for children with complex chronic medical conditions: A pilot study.* Child: Care, Health and Development, 2016. **42**(3): p. 439-449.
394. Lichstein, J.C., R.M. Ghandour, and M.Y. Mann, *Access to the medical home among children with and without special health care needs.* Pediatrics, 2018. **142**(6).
395. Lin, J.L., E. Cohen, and L.M. Sanders, *Shared Decision Making among Children with Medical Complexity: Results from a Population-Based Survey.* Journal of Pediatrics, 2018. **192**: p. 216-222.
396. Looman, W.S., et al., *Health-Related Quality of Life Outcomes of a Telehealth Care Coordination Intervention for Children With Medical Complexity: A Randomized Controlled Trial.* Journal of Pediatric Health Care, 2018. **32**(1): p. 63-75.
397. Mandic, C.G., et al., *Impact of caring for children with medical complexity on parents' employment and time.* Community, Work and Family, 2017. **20**(4): p. 444-458.
398. Marshall, J., et al., *Intervention for feeding difficulties in children with a complex medical history: A randomized clinical trial.* Journal of Pediatric Gastroenterology and Nutrition, 2018. **66**(1): p. 152-158.
399. Matiz, L.A., L. Robbins-Milne, and J.A. Rausch, *EMR Adaptations to Support the Identification and Risk Stratification of Children with Special Health Care Needs in the Medical Home.* Maternal and Child Health Journal, 2019.
400. McKissick, H.D., et al., *The Impact of Telehealth and Care Coordination on the Number and Type of Clinical Visits for Children With Medical Complexity.* Journal of Pediatric Health Care, 2017. **31**(4): p. 452-458.
401. Meier, J.D., et al., *Emergency department use among children with tracheostomies: Avoidable visits.* Journal of Pediatric Rehabilitation Medicine, 2015. **8**(2): p. 105-111.
402. Mesman, G.R., et al., *The Impact of Technology Dependence on Children and Their Families.* Journal of Pediatric Health Care, 2013. **27**(6): p. 451-459.

403. Miller, A.R., et al., *Continuity of care for children with complex chronic health conditions: Parents' perspectives.* BMC Health Services Research, 2009. **9**.
404. Ming, D.Y., et al., *Mobile Complex Care Plans to Enhance Parental Engagement for Children With Medical Complexity.* Clinical Pediatrics, 2019. **58**(1): p. 34-41.
405. Murphy, K.L., et al., *Rural and nonrural differences in providing care for children with complex chronic conditions.* Clinical Pediatrics, 2012. **51**(5): p. 498-503.
406. Nageswaran, S. and S.L. Golden, *Home Health Nurse Collaboration in the Medical Neighborhood of Children with Medical Complexity.* Home healthcare now, 2016. **34**(9): p. 507-518.
407. Nageswaran, S. and S.L. Golden, *Improving the Quality of Home Health Care for Children With Medical Complexity.* Academic Pediatrics, 2017. **17**(6): p. 665-671.
408. Nageswaran, S. and S.L. Golden, *Establishing relationships and navigating boundaries when: Caring for children with medical complexity at home.* Home Healthcare Now, 2018. **36**(2): p. 93-102.
409. Nageswaran, S., et al., *Caregiver Perceptions about their Decision to Pursue Tracheostomy for Children with Medical Complexity.* Journal of Pediatrics, 2018. **203**: p. 354-360.e1.
410. Nageswaran, S., A.I. Rosado, and M.S. Beveridge, *Challenges Faced by Latino Caregivers in Transportation of Children with Medical Complexity.* North Carolina medical journal, 2018. **79**(6): p. 358-364.
411. Nasir, A., et al., *Complexity in pediatric primary care.* Primary Health Care Research and Development, 2018: p. 1-7.
412. Nkoy, F.L., et al., *Information needs for designing a home monitoring system for children with medical complexity.* International Journal of Medical Informatics, 2019. **122**: p. 7-12.
413. Noritz, G., et al., *A population intervention to improve outcomes in children with medical complexity.* Pediatrics, 2017. **139**(1).
414. Parmeter, J., D. Tzioumi, and S. Woolfenden, *Medical neglect at a tertiary paediatric hospital.* Child Abuse and Neglect, 2018. **77**: p. 134-143.
415. Peltz, A., et al., *Characteristics of rural children admitted to pediatric hospitals.* Pediatrics, 2016. **137**(5).
416. Perros, I., et al., *Temporal phenotyping of medically complex children via PARAFAC2 tensor factorization.* Journal of Biomedical Informatics, 2019. **93**.
417. Petitgout, J.M., *The Financial Impact of a Hospital-Based Care Coordination Program for Children With Special Health Care Needs.* Journal of Pediatric Health Care, 2018. **32**(1): p. 3-9.
418. Petruik, C., et al., *Talking with caregivers of children living in the community with ventricular assist devices.* Pediatric Transplantation, 2017. **21**(7).
419. Phillips, C.D., et al., *Post-acute care for children with special health care needs.* Disability and Health Journal, 2018. **11**(1): p. 49-57.
420. Pizur-Barnekow, K., *Maternal health after the birth of a medically complex infant: Setting the context for evaluation of co-occupational performance.* American Journal of Occupational Therapy, 2010. **64**(4): p. 642-649.
421. Puls, H.T., et al., *Failure to Thrive Hospitalizations and Risk Factors for Readmission to Children's Hospitals.* Hospital pediatrics, 2016. **6**(8): p. 468-475.
422. Quigley, L., et al., *A qualitative analysis of information sharing for children with medical complexity within and across health care organizations.* BMC Health Services Research, 2014. **14**(1).
423. Quinn, B.L., et al., *CE: Original research: Pain in nonverbal children with medical complexity: A two-year retrospective study.* American Journal of Nursing, 2018. **118**(8): p. 28-37.

424. Rafferty, K.A. and S.L. Sullivan, *"You Know the Medicine, I Know My Kid": How Parents Advocate for Their Children Living With Complex Chronic Conditions.* Health Communication, 2017. **32**(9): p. 1151-1160.
425. Ranade-Kharkar, P., et al., *Information needs of physicians, care coordinators, and families to support care coordination of children and youth with special health care needs (CYSHCN).* Journal of the American Medical Informatics Association, 2017. **24**(5): p. 933-941.
426. Ralston, S.L., et al., *Hospital variation in health care utilization by children with medical complexity.* Pediatrics, 2015. **136**(5): p. 860-867.
427. Raymond, J.A., *The integration of children dependent on medical technology into public schools.* Journal of School Nursing, 2009. **25**(3): p. 186-194.
428. Rehm, R.S. and J.F. Bradley, *The Search for Social Safety and Comfort in Families Raising Children With Complex Chronic Conditions.* Journal of Family Nursing, 2005. **11**(1): p. 59-78.
429. Ronis, S.D., et al., *Estimated nonreimbursed costs for care coordination for children with medical complexity.* Pediatrics, 2019. **143**(1).
430. Schrager, S.M., et al., *Development and validation of a method to identify children with social complexity risk factors.* Pediatrics, 2016. **138**(3).
431. Sellers, D., et al., *Development and reliability of a system to classify the eating and drinking ability of people with cerebral palsy.* Developmental Medicine & Child Neurology, 2014. **56**(3): p. 245-251.
432. Walter, A.W., R.P. Ellis, and Y. Yuan, *Health care utilization and spending among privately insured children with medical complexity.* Journal of Child Health Care, 2018.