Children with Medical Complexity: Evaluating Reported Costs and Modelling Predictors of Hospital and ED Resource Use

by

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A thesis submitted in partial fulfillment of the requirements for the degree of

Doctor of Philosophy

in

Health Services and Policy Research

School of Public Health
University of Alberta

Abstract

Children with medical complexity (CMCs) are a vulnerable population that suffers from complex chronic conditions that require intensive health care resources. Previous studies, largely focused on hospitalizations and emergency department visits, have examined how clinical factors are associated with health care resource use for this population. However, the impacts of socioeconomic factors are less studied. There is also little information on the Alberta CMC population, beyond broad quantifications of health care resource use at the national level. The objective of this research was first to identify how health care costs for CMCs are reported in the literature and then to examine how both clinical and socioeconomic factors are jointly associated with hospital days and emergency department use after an index admission (first admission with a diagnosis of Complex Chronic Condition) among CMCs in Alberta, and how these associations change over time.

This dissertation first presents a systematic review outlining how CMCs are identified in administrative health data together with the types and methods of cost reporting for CMCs. The review identifies limitations in interpreting costing trends and summarizes recommendations from the literature on CMC health care resource costing.

The dissertation then studies a population-based cohort of CMCs in Alberta through separate inferential and predictive analyses to quantitatively assess how clinical and socioeconomic factors are associated with hospitalizations and ED visits over time. The first analysis utilizes a mixed-effect linear hurdle regression model to longitudinally estimate and perform inference for these associations. The second analysis uses a tree-based gradient-boosted regression model to predict and identify the most important predictors of resource use in the

short and long terms (i.e., 1 and 5 years after index admission, respectively). The analysis leads into a discussion of the opportunities and limitations of using administrative health data and electronic health records in predictive machine learning models in CMC-related research.

The first analysis found that initial length of stay (LOS) and number of chronic medications, both proxies for clinical complexity, have strong, positive associations with resource use. Specifically, a greater initial LOS was significantly, positively associated with more hospital days whereas more chronic medications were significantly, positively associated with more hospital days and more ED visits in the first year after initial discharge. In terms of socioeconomic factors, the analysis found that CMCs living in rural and remote rural areas had more ED visits than those living in urban/metropolitan locations. Material and social deprivation, here measured with Canadian census data, also had significant, positive associations with ED visits.

The second analysis also showed that clinical proxy measures namely initial LOS and clinical classification (single vs. multiple complex chronic conditions) were top predictors of hospital days. The top predictors of ED use were consistently socioeconomic in nature, with patient residence being an important predictor. The results highlighted the need for more-detailed clinical and socioeconomic data in electronic health records and supported further discussion of how predictive modelling can be used in the future to enhance understanding of the CMC population.

Overall, that both analyses pointed to the relationships between clinical complexity and hospital use and between socioeconomic status and ED visits is an important contribution to the CMC literature and, more specifically, to policy development efforts and health care

administrators aiming to improve care for the Alberta CMC population. Further research on the availability of health care resources for CMCs living in (remote) rural areas of Alberta is warranted. Finally, this dissertation demonstrated that predictive and inferential modelling using administrative health data can be used to identify factors associated with health care use by CMCs.

Preface

This dissertation is an original work by Michael Sidra. The research project, of which this dissertation is a part, received research ethics approval from the University of Alberta Research Ethics Board (Health care Service Utilization by Children with Medical Complexity in Alberta, Pro00103550_REN2, December 7, 2020). The research presented in this dissertation did not receive any funding.

Chapter 2 of this dissertation has been published as Sidra, M., Sebastianski, M., Ohinmaa, A., & Rahman, S. (2022). Reported costs of children with medical complexity—A systematic review. *Journal of Child Health Care*, 13674935221109683. Advance online publication. https://doi.org/10.1177/13674935221109683. Michael Sidra was responsible for the design of the review protocol, including the development of the search criteria in collaboration with a librarian, the identification of selection criteria, data collection, data analysis, and manuscript development. Dr. Meghan Sebastianski was responsible for data analysis, third-opinion review, and manuscript edits. Prof. Arto Ohinmaa was responsible for providing guidance, supervision throughout, and manuscript edits. Sholeh Rahman was a second-opinion reviewer for data analysis and study selection.

Chapter 3 of this dissertation was submitted for publication at the time of writing as and authored by Sidra, M., Pietrosanu, M., Ohinmaa, A., Zwicker, J., Round, J., & Johnson, D. W. (2023). Clinical and socioeconomic associations with health care use among medically complex children. Michael Sidra was responsible for study conceptualization, methodology, formal analysis, and drafting and editing the manuscript. Matthew Pietrosanu contributed to study conceptualization, methodological development, investigation, formal analysis, and reviewing and editing the final manuscript. Prof. Arto Ohinmaa, supervised and provided guidance on study

conceptualization, methodological development, investigation, and contributed to the formal analysis and the reviewing and editing of the manuscript. Prof. Jennifer Zwicker contributed to concept design, methodology, investigation, and reviewing and editing the manuscript. Prof. Jeff Round contributed to methodological development and reviewing and editing the manuscript. Dr. David Johnson contributed to study conceptualization, methodological design, investigation, data curation, and reviewing and editing the manuscript.

Chapter 4 of this dissertation was ready for submission at time of writing as Sidra, M., Pietrosanu, M., Ohinmaa, A., Zwicker, J., Round, J., and Johnson, D. W. (2023). Clinical and socioeconomic predictors of hospital use and ED visits among children with medical complexity in Alberta: A machine learning approach using administrative data. Michael Sidra was responsible for study conceptualization, methodology, formal analysis, and drafting and editing of the manuscript. Matthew Pietrosanu contributed to study conceptualization, methodological development, investigation, formal analysis, and reviewing and editing the final manuscript. Prof. Arto Ohinmaa, supervised and provided guidance on study conceptualization, methodological development, and investigation, and contributed to the formal analysis and the reviewing and editing of the manuscript. Prof. Jennifer Zwicker contributed to concept design, methodology, investigation, and reviewing and editing the manuscript. Prof. Jeff Round contributed to methodological development and reviewing and editing the manuscript. Dr. David Johnson contributed to study conceptualization, methodological design, investigation, data curation, and reviewing and editing the manuscript.

Dedication

Dedicated to my wife, Marie Beliveau, for her unwavering support - I am forever grateful.

To Emma, Ethan, Elizabeth and Eva, you all inspire me.

Acknowledgements

I would like to express my sincere gratitude to my supervisor professor Arto Ohinmaa for his mentorship and continued support over the course of this program. His direction and gentle prodding for ideas forced me to think creatively and exposed me to several perspectives on difficult problems. I would also like to thank the rest of my committee for their guidance and support. Prof. Jennifer Zwicker's generous guidance on the practical implications of this work helped shape my understanding and enhanced my learning. Prof. Jeff Round's perspectives challenged me to consider the wider implications of this work.

I would also like to thank Dr. David Wyatt Johnson for his patience, guidance, and support. His passion for improving care for children with medical complexity is contagious and I am thankful for the opportunity to learn from him throughout this journey.

I would especially like to thank Matthew Pietrosanu for his support in all things statistics. His expertise and professional advice as well as his ability to explain complex concepts in a simple way was crucial for my development as a researcher. I am also grateful for his critical appraisal of my writings and conclusions as it helped me clarify my thoughts and refine my ideas.

Lastly, I would like to thank my colleagues and leaders at Alberta Health Services, for their support. Specifically, I'm grateful to Dr. Don Dick, Carmella Steinke, Lynette Lutes, and Dr. Francois Belanger for their encouragement.

Table of Contents

List of Tables

List of Figures

List of Abbreviations

AHRQ	Agency for Health care Research and Quality
AHS	Alberta Health Services
AHSDR	Alberta Health Services Data Repository
AUC	Area Under Curve
CASP	Critical Appraisal Skills Program
CBPPC	Community-based Pediatric Palliative Care
CCC	Complex Chronic Conditions
CCI	Chronic Condition Indicator
CCMS	Comprehensive Case Management Service
CI	Confidence Interval
CIHI	Canadian Institute for Health Information
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CMC	Children with Medical Complexity
CYSHCN	Children and Youth with Special Health Care Needs
DA	Dissemination Area
DAD	Discharge Abstract Database
DME	Durable Medical Equipment
EBSCO	Publishing Company
ED	Emergency Department
EMBASE	Excerpta Medica Database
HH	Home Health Nursing
ICD	International Classification of Diseases
ICU	Intensive Care Unit
IOM	Institute of Medicine
LOS	Length of Stay
LTC	Long Term Care
MHS	Mental Health Service
NACRS	National ambulatory care reporting system
OR	Operating Room

PACT	Plans for Action and Care Transitions
PICU	Pediatric Intensive Care Unit
PIN	AHS Pharmaceutical Information Network
PRISMA	Preferred Reporting Items for Systematic Reviews and meta analyses
RCT	Randomized Control Trial
RD	Relative Difference
ROC	Receiver–Operator characteristic
SDH	Social Determinants of Health
SE	Standard Error
SNP	Special Needs Program
TA	Technology Assistance
US	United States
USA	United States of America
WA	Washington
WCV	Well Child Visit

Chapter 1. Introduction

1.1 Children with Medical Complexity

Children and youth with special health care needs (CYSHCN) have a wide range of developmental, physical, or psychosocial conditions that require health care and other related services beyond what would typically be needed for children. Of particular focus within the CYSHCN population are children with medical complexity (CMCs), who form a more vulnerable and more medically complex pediatric subpopulation (Kuo et al., 2016). In the context of CMCs, medical complexity refers to having one or more complex chronic conditions that may affect multiple organs and require care from numerous specialists and possibly medical technology (Gordon et al., 2007). The term “children with medical complexity” was first introduced by Cohen et al. (2011), who describe CMCs as “the most medically fragile and have the most intensive health care needs" (p. 529). Their landmark paper outlined a need for focused research on CMCs and a definitional framework to identify this subpopulation. Dewan and Cohen (2013) describe CMCs as having four distinguishing characteristics:

1. the presence of one or more complex chronic conditions that are often multisystem and severe;
2. significant functional limitations that often require a reliance on technology;
3. high health care utilization; and
4. high health care service needs in the home and from parents and caregivers.

The following is an example of a child with medical complexity given by Berry et al. (2013).

> A 9-year-old child with spina bifida and eight different co-morbid conditions…who relies on the care of 17 different health care providers and allied health professionals…who is dependent on a wheelchair for mobility and a gastrostomy tube for nutrition…and is routinely hospitalized five to ten times per year…

CMCs are the most vulnerable pediatric subpopulation. They make up a small percentage of the pediatric population, have a higher mortality rate, and utilize a substantial amount of health care resources compared to the general pediatric population. A report by the Canadian Institute of Health Information (CIHI, 2020) stated the 2015–2016 age-adjusted rate of medical complexity in Canada for CMCs to be 948 per 100,000 children. Alberta's rate was slightly higher at 999 per 100,000 children and youth (newborns to 24-year-olds). The mortality rate for CMCs is also higher than that for the general pediatric population. CIHI (2020) reported that the proportion of CMCs who die in the hospital was greater than both the proportions in the general and pediatric populations. Cohen et al. (2012), in an examination of CMCs in Ontario, reported that 2.5% of their cohort died over a two-year follow up period. The Alberta CMC cohort studied in this dissertation had a mortality rate of 4% over a five-year period. Although mortality rates for CMCs are higher than those for other pediatric groups, CMC prevalence is likely increasing due to higher survival rates for some conditions such as congenital anomalies and malignancies (Burns et al., 2010; Tennant et al., 2010). Despite increasing survival rates, CMCs remain medically fragile. For CMCs, medical fragility refers to the need for services to carry out basic life functions, whereas medical complexity refers to multiple factors that include both skilled services and technology assistance (Kuo et al., 2016). Walter et al. (2019), for example, reported that children with complex chronic conditions accessed mental health services twice as much as children without complex chronic conditions.

Identifying CMCs through administrative health data is challenging despite the group's high health care use because indications of medical complexity are multifaceted. In terms of administrative data, these indications include the presence of multiple complex chronic conditions, the need for medical technology assistance, and high resource needs. However, other indicators of medical complexity such as functional limitations are harder to observe through administrative health data. At the patient level, physicians may be able to identify functional limitations brought on by a CMC's medical condition and affecting the child's quality of life. For example, if a child is unable to perform daily tasks such as walking or dressing due to their medical condition, they will be limited in their ability to participate in school or community activities (Kuo et al., 2016). However, this type of patient-level information is not readily available in administrative health data. There is currently no universally adopted method for identifying CMCs. The four most common techniques used in the literature (Berry et al., 2015) are:

1. Complex Chronic Conditions classification system (Feudtner et al., 2000),
2. Clinical Risk Groups (3M, 2020),
3. Chronic Condition Indicators (Agency for Healthcare Research and Quality, 2020), and
4. Patient Medical Complexity algorithm (Simon et al., 2014).

Each of the four methods have strengths and limitations in identifying CMCs in administrative data. For example, the Complex Chronic Conditions classification system is publicly available (unlike the proprietary 3M Clinical Risk Groups) and is based on ICD-10 codes which are readily used to identify diseases in health care literature. However, the Complex Chronic Conditions classifications system does not include certain chronic conditions such as asthma and obesity which, in combination with other chronic conditions, results in medical complexity in

children (Berry et al., 2015). The systematic review in Chapter 2 of this dissertation discusses these approaches in more detail and identifies the methods most commonly used in the literature for quantifying CMC health care resource use. Although these methods identify CMCs in administrative health data, a common limitation among all of them is the inability to identify functional limitations stemming from chronic conditions. This drawback limits CMCs' access to health care resources and adds to financial and psychosocial stresses on families and caregivers.

1.2 CMCs and Family Burden

Beyond having extensive health care needs, CMCs also require significant caregiver resources and present many challenges in this regard. These children often require care from multiple specialists in various medical disciplines and have the highest risk among the pediatric population for adverse developmental and psychosocial outcomes (Cohen et al., 2012; Berry et al., 2015). For example, Cohen et al. (2012) found that CMCs in Ontario saw on average about 13 physicians from six disciplines. In addition to managing CMCs' chronic medical conditions, families also struggle to ensure that their children can participate in everyday activities such as additional training from teachers or coaches (Ray, 2002). In a Canadian study, Brehaut et al. (2009) demonstrated a positive correlation among caregivers between caring for children with health problems and the likelihood of chronic medical conditions and mental health issues. According to CIHI (2020), "37% [of primary caregivers of CMCs] expressed anger, distress or depression, or felt they were unable to continue in their caring activities" (p. 20). Parents of CMCs can also endure medically traumatic stress as a result of a traumatic incident or ongoing stress, which can be exacerbated by a lack of trust in their child's health care provider (Dewan et al., 2023).

Studies have also shown that the families and caregivers of CMCs are at higher risks of medical, financial, marital, and employment difficulties (Kuo et al., 2014; Edelstein et al., 2017; Orkin et al., 2019). A U.S. study by Walter et al. (2019) reported that the families of CMCs were almost four times more likely to spend over $1,000 out of pocket for medical care per year than the families of non-CMCs. These out-of-pocket expenses did not include lost income due to the reduction in working hours necessary to provide care for their medically complex child. In Canada, the families of CMCs also report needing to purchase additional private insurance plans or needing to pay out of pocket for medications (CIHI, 2020). These families often experience dramatic lifestyle changes as a result of these costs. For example, one parent may be required to reduce their working hours in order to care for their child while the other may need to work more by taking up another part-time job in order to balance additional medical expenses. The families of CMCs also face a number of other challenges such as marital stress and the need for additional financial and caregiving support from extended family members. The siblings of CMCs are also affected; in some cases, siblings are required to restrict social activities in order to prevent passing an infectious illness to their medically complex sibling (Connor et al., 2010).

Numerous studies have examined ways to support the families and caregivers of CMCs. Support interventions generally fall under six domains: respite, telemedicine, peer and emotional support programs, insurance and employment benefits, health and related supports, and care coordination, with the latter being the most common type (Edelstein et al., 2017). The American Society for Pediatrics describes care coordination as a "patient- and family-centered, assessment-driven, team-based activity designed to meet the needs of children and youth while enhancing the caregiving capabilities of families" (Council on Children with Disabilities and Medical Home Implementation Project Advisory Committee, 2014).

While care coordination is not standardized and differs both between and within health systems, evidence exists in favour of such programs. Care coordination interventions have been reported to improve the quality of care for patients, reduce caregiver burden, and reduce health care costs such as those related to hospital use (Cohen et al., 2012; Berry et al., 2014; Goldhagen et al., 2016; Sidra et al., 2022). Despite the reported advantages of the majority of care coordination interventions, there remains a substantial gap in the literature on the characteristics of CMCs that are the most likely to benefit from care coordination (Edelstein et al., 2017). Several studies have recommended a needs-based approach where care coordination and other family-support programs and resources are directed to the CMCs who are most likely to benefit (Bekmezian et al., 2008; Gay et al., 2016; Gold et al., 2016; Simon et al., 2017). The clinical heterogeneity of CMCs makes it plausible that care needs differ across CMC subpopulations.

1.3 CMCs and Health Care Resource Use

There are several interrelated factors that contribute to health care resource use among CMCs and the general population. Theoretical frameworks such as the Andersen behavioral model of health service use, the Grossman utility model, and the Institute of Medicine's Access to Healthcare Services and Healthcare Disparities and Cultural Competency Model describe the interplay between patient and health system characteristics that influence health services use (Meade et al., 2015). Other frameworks, such as the Penchansky model, focus on client satisfaction and define access to health care in terms of availability, accessibility, accommodation, affordability, and acceptability (Karikari-Martin, 2010). However, the Penchansky model necessitates primary data collection as not all elements are typically captured in administrative health data. Within these frameworks, CMC characteristics such as lower

socioeconomic status, clinical need, and other physical or psychosocial limitations are associated with higher health care resource use (Cohen et al., 2012).

While disproportionately higher health care spending by CMCs relative to other pediatric populations is well documented, empirical evidence of how patient characteristics such as socioeconomic factors impact health care resource use is limited. Complicating this issue is the difficulty of comparing reported health care costs across jurisdictions. Health care costing studies utilize various administrative databases in which cost may be reported at a micro level (e.g., hospital costs) or at a macro level (e.g., via case-mix methods that account for all costs, including primary care and diagnostics). Despite the variability in reported costs for CMCs, studies consistently report hospital and emergency department (ED) utilization (e.g., in 89% and 37%, respectively, of the studies considered in Chapter 2) through numbers of hospital days and ED visits or through associated costs. Measuring these resources is important from a health system perspective because both are costly services and avoidable hospitalization, and ED visits cause additional financial and psychosocial stress on the family and may indicate unmanaged care for the child.

1.3.1 Hospital Use

Berry et al. (2017) calculated that children with multiple chronic conditions accounted for over one-fourth of acute care hospitalizations and one-half of all pediatric hospital dollars in the U.S., while Srivastava et al. (2016) reported that CMCs accounted for one-third of total hospital costs in Australia. CIHI (2020) reported an $866 million cost associated with hospital care for children and youth with medical complexity in Canada during 2015–2016. Despite CMCs being only about 1% of children and youth in Canada, they accounted for 57% of hospital care costs, 37% of all hospital admissions, and 54% of total hospital days among the pediatric population.

The CIHI report also found that CMCs had more health care visits relative to children without medical complexity (i.e., six vs. three). In Ontario, Cohen et al. (2012) reported that CMCs accounted for approximately one-third of pediatric health care resources, of which 79% was attributable to index hospitalizations and subsequent rehospitalizations.

Hospitalization rates for CMCs vary across jurisdictions and even within countries (Gold et al., 2016). In a retrospective, population-based study, Ralston et al. (2015) found that the numbers of inpatient and intensive care days varied more than two-fold between three U.S. states. This variation in health care resource use, which included hospitalizations and ED visits, could not be explained by population differences in the cohort.

Hospital use is increasingly being used as an indicator of care quality and is the target of quality improvement efforts in reducing hospital costs (Coye, 2008; Berry et al., 2011). Studies have shown that hospitalization among CMCs is associated with both clinical and socioeconomic factors (Garcia-Altes et al., 2018; Coller et al., 2018). Understanding relationships between these factors and resource use is thus important for care delivery and the development of health care policy. The variation in reported hospital use also reinforces the need for local investigations, beyond what is currently available in the literature. The systematic review in Chapter 2 found no studies examining hospital or ED use in Alberta. While CIHI (2020) reported high-level statistics on health care use by province, they did not conduct further analyses of trends in or correlates of utilization.

1.3.2 Emergency Department Use

Several studies have shown that CMCs also utilize EDs substantially more than the general pediatric population. At least two-thirds of CMCs had at least one ED visit during the

2015–2016 and 2017–2018 periods in Ontario, Alberta, and the Yukon (CIHI, 2020). CMCs are also more likely to be admitted to an ED and more likely to return after being discharged (Akenroye et al., 2014). Moreover, CMCs are at a higher risk of medical errors that may result in a need for urgent care. For example, Feinstein et al. (2014) found that ED visits related to adverse drug events were more likely in children with complex chronic conditions.

Access to urgent medical care through EDs is critical. However, going to an ED can be stressful for the families and caregivers of CMCs (CIHI, 2020), especially for those in lower socioeconomic positions (Berry et al., 2010). Particularly in rural or remote locations where specialized expertise and support might not be readily available, EDs may be the only source of after-hours care for CMC families. In a cross-sectional, nation-wide sampling study, Murtagh et al. (2014) reported that up to 70% of children with complex chronic conditions and 92% of transition-aged patients were treated in general EDs rather than at a pediatric ED. General ED providers might not be as comfortable providing care for CMCs due to a lack of training, time constraints, or a lack of reimbursement for care coordination (Pulcini & Rubin, 2019).

Identifying factors that drive ED utilization by CMCs is complex and requires both clinical and socioeconomic considerations, but relevant quantitative analyses are limited for the Alberta CMC population and in the CMC literature more generally. Given the cost to health care systems and the impact on CMCs and their families, there is a need to enhance understanding of the relationship between clinical/socioeconomic factors and ED utilization.

1.4 CMCs and Socioeconomic Status

Social determinants of health (SDHs) play an important role in children's health generally (Ungar, 2009), including long-term health (Halfon et al., 2010; Barnert et al., 2017).

Newacheck et al. (2003), based on data from the U.S. National Household Survey, reported a strong, negative association between income level and unmet health needs. In a Spanish study, Carrilero et al. (2020) found that a high proportion of CMCs under 15 years of age were in lower socioeconomic positions (defined as no household members employed or one or more members employed with a combined income of less than €18,000 per year). A U.S.-based, qualitative study by Barnert et al. (2017) identified five main themes in interviews about the CMC population with health experts. Two of the five themes centred on SHDs and the health of the family unit. The study reported that SDHs such as economic position, race, and geographic location all had strong effects on health outcomes for CMCs through access to care and parental stress. The study also noted that differences in access to care due to SDHs would have a more acute impact on CMCs because they need more-intensive care relative to non-CMCs.

Low socioeconomic status and the inaccessibility of specialized care (e.g., through residence rurality) may present a barrier to access to otherwise available health services, place increased stress on caregivers, and lower quality of care (Barnert et al., 2017). A systematic review by Spencer et al. (2015) found a negative association between socioeconomic status and disabling childhood chronic conditions in high-income countries. While the review included 10 studies from Canada, none were focused solely on CMCs or Alberta. In their Spanish study of socioeconomic parameters and CMC comorbidity, Carrilero et al. (2020) reported that children in lower socioeconomic positions had greater representation in all identified CMC groups relative to non-CMCs. Both studies note that, while an association exists, it is not known if disabling chronic conditions are part of a causal pathway to poor social and environmental conditions in early childhood or vice versa (i.e., disabling chronic conditions in children lead to low socioeconomic status). Caring for CMCs is known to have negative financial impacts on

families that may also affect the CMC (Thomson et al., 2016, Barnert et al., 2017). On the other hand, disadvantageous socioeconomic conditions may contribute to the development of chronic conditions such as asthma or congenital abnormalities (Correa-Villaseñor et al., 1991; Halfon & Newacheck, 1993; Spencer et al., 2015). Regardless of the possible causal pathway, an understanding of associations between health care utilization, health outcomes, and SDHs for the CMC population are useful in informing policy and operational decision-making. Research on associations between socioeconomic status and health care utilization patterns for CMCs in Alberta (and Canada more generally) is lacking in the academic literature.

1.5 Research Gaps

1.5.1 Clinical Research Gaps

With the clinical fragility of CMCs, associated costs to health care systems, and impacts on families and caregivers in mind, an understanding of factors that drive health care use for this heterogeneous population is essential in designing effective care programs that efficiently improve patient outcomes and the experiences of caregivers.

While CIHI (2020) has reported on the demographic characteristics of the Canadian CMC population, their report neither explores relationships between utilization trends and clinical factors nor discusses in any detail the impact of SDHs on CMCs. In another vein, most of the peer-reviewed literature on Canadian CMCs focuses on Ontario and works on resource utilization outside of Canada typically originate from jurisdictions with different delivery models. A more-nuanced understanding of Albertan CMCs is thus necessary to correctly inform decision makers. At the same time, an understanding of the strengths and limitations of studies reporting from privately funded health care organizations (e.g., the U.S.) can further

contextualize the conclusions and applicability of these works relative to publicly funded health care systems (e.g., Canada and Australia).

There is also a significant gap in the general literature (not specific to Alberta) on the impact of socioeconomic factors on health care utilization by CMCs. A meta-analysis and systematic review conducted by Spencer et al. (2015) on disabling childhood chronic conditions and socioeconomic status did not list any studies focusing specifically on CMCs even though CMCs are the most clinically complex patient population. Recently, two Spanish studies (Garcia-Altes et al., 2018; Carrilero et al., 2020) examined socioeconomic inequalities and the use of health care services among CMCs. Their results suggest significant associations between socioeconomic factors and health care use, which confirms the importance of socioeconomic factors with respect to CMC outcomes.

1.5.2 Methodological Research Gaps

Most of the research on CMCs involves retrospective, cross-sectional cohort studies. Longitudinal studies of CMC populations are limited in the literature. While a few U.S. studies consider longitudinal CMC cohorts, no analogous work exists from Canada. The existing longitudinal research from the U.S. does not describe relationships between hospital use and clinical or socioeconomic factors, but instead focuses on how clinical characteristics of the population change over time (Berry, Hall, et al., 2013). While the crude time trends presented in these studies are useful, they do not account for clinical or socioeconomic factors that other works have found to be important (Berry et al., 2011; Carrilero et al., 2020), nor do they address how associations with these factors change over time. More-detailed analyses of time trends (e.g., resource use over time) would provide important details and insights to researchers and administrators who create or modify policies for the CMC population.

Existing studies predominantly utilize inferential methods (e.g., regression analysis) to quantify associations between resource use and factors readily available in administrative health care data. While inferential models are useful for assessing the statistical significance of broad trends in a population, they are not suited for predictive purposes (e.g., forecasting or making patient-level predictions based on new data). The latter have become increasingly attractive in recent years due to proliferation of electronic health records and technological advancements in data storage technologies. Instead, these objectives are better met by more-modern machine learning methods, which typically boast far greater model capacity through their ability to analyze large numbers of variables and account for more-complex interactions than inferential models (King & Strumpf, 2022).

While not new, machine learning methods have only recently seen wide-scale adoption in certain fields of research. Applications of machine learning within health care are limited primarily to diagnostics and screening for a small number of conditions such as cancers and nervous system diseases (Jiang et al., 2017), and applications to administrative data are sparse and remain in an exploratory, developmental stage (King & Strumpf, 2022). Predictive machine learning models, such as tree-based models and neural networks, are well-positioned to take advantage of routinely collected administrative data. Well-performing predictive models could, for example, identify important predictors (as opposed to just correlates) of resource utilization and make individualized predictions regarding future resource use. While these are significant advantages for heterogeneous populations such as CMCs, machine learning models are at present not established in this area of the literature.

1.6 Research Questions and Objectives

This dissertation focuses on the Alberta CMC population and addresses several important, general gaps in CMC research. As high health care resource use is a defining characteristic of CMCs, understanding the context of how health care costs are reported for this population is essential in understanding the applicability and limitations of existing work.

Research Question 1: What and how are costs reported in the existing literature for CMC populations?

The systematic review in Chapter 2 explores this question, reports on how CMCs are identified in the literature where health care costs are reported, examines the ways costs are reported, and explores authors' recommendations pertaining to cost reporting. This systematic review provides essential context on interpretations and limitations of cost-reporting for CMCs.

Since hospital admissions and ED service utilization are the most commonly reported measures of resource use, the next research question explores how clinical and socioeconomic factors are associated with these measures over time.

Research Question 2: How are clinical and socioeconomic factors, in the five years following initial hospitalization, associated with hospital and ED use among the CMC population in Alberta, Canada?

The composition of Alberta CMC data (namely, the longitudinal nature of the data and the large proportion of CMCs with no resources use) precludes standard modelling approaches such as linear regression. These complications, which have the potential to invalidate statistical inference, are not considered in the existing literature. Chapter 3 of this dissertation uses a linear

mixed-effect hurdle regression model to address these issues and perform valid inference for the CMC population in Alberta.

However, as discussed previously, inferential models are not suitable for predictive tasks despite the insights the former permits into population-level associations. Research Question 3 explores predictive modelling under a different paradigm, here with the main goal of identifying important predictors (rather than just correlates).

Research Question 3: What are clinical and socioeconomic predictors of short- and long-term hospital use and ED visits by CMCs in Alberta, Canada?

Unlike the previous hurdle model, the boosted regression tree model used in Chapter 4 to address Research Question 3 can account for complex interactions between clinical and socioeconomic factors. This analysis also permits a preliminary examination of the utility of routinely collected administrative health care data in the development of predictive machine learning models of CMC outcomes and naturally addresses this dissertation's final research question.

Research Question 4: What are the opportunities and limitations of how administrative health data is currently being collected in electronic health records with regard to predictive machine learning models?

The discussion in Chapter 4 considers the opportunities and limitations of leveraging electronic health records and administrative health data for predictive modelling. This discussion puts forward recommendations for improving electronic health records to support more-robust predictive modelling endeavours in the future.

Broadly, this dissertation identifies important correlates and predictors of health care resource use among CMCs in Alberta and comments on the future integration of electronic health records and machine learning models in CMC-related research. This dissertation can empower health care providers, researchers, and policy makers through a more-nuanced understanding of the relationships between clinical/socioeconomic factors and CMC resource use. Methodologically, this work uses both inferential and predictive modelling techniques to describe and better understand the CMC population in a longitudinal context. These results add to the general knowledge on CMCs, provide critical information about the Alberta CMC population, and facilitate discussion on the future of administrative health care data in CMC research.

1.7 MyChild Dataset

Chapters 3–4 of this dissertation use the MyChild dataset (constructed by the Alberta Children's Hospital in 2018) to address Research Questions 2–4. Since Alberta has a single integrated provincial health system, the analyses presented incorporate data on all CMCs in the province. The dataset includes both clinical and socioeconomic variables, which makes the data ideal for the proposed work. This section gives an overview of important features of the dataset but see Chapters 3–4 for further details (e.g., cohort demographic summaries).

1.7.1 Inclusion Criteria

MyChild defines a CMC as a child (at most 18 years of age) who was first admitted and discharged from an acute care site in Alberta between 2010 and 2013 with a diagnosis of neurological impairment, multiple complex chronic conditions, or a single complex chronic condition (index admission), as defined by the ICD-10-based coding scheme in Feudtner et al.

(2000) and Cohen et al. (2012). The number of CMCs identified for each discharge year is presented in Table 1.

Table 1.1 Initial discharge date by year in the MyChild dataset.

Discharge Year	**Patients**
*2010	3,645
2011	4,003
2012	3,988
**2013	985

* 2010 only includes patients from April to December

**2013 only includes patients from January to March

1.7.2 Outcome Variables

Yearly numbers of hospital days and ED visits were derived from commonly used administrative health data. Provincial hospital discharge data was extracted from the Discharge Abstract Database (CIHI, 2023a) and ED visit data from the National Ambulatory Care Reporting System (NACRS) (CIHI, 2023b). Cumulative counts for each response were extracted yearly for each of the five years following each CMC's initial discharge.

1.7.3 Clinical Variables

CMCs were placed into one or more of nine nondisjoint clinical categories based on ICD-10 codes (Cohen et al., 2012): cardiology, congenital/genetic, gastrointestinal, hematology/immunodeficiency, malignancy, metabolic, neurology, renal, and respiratory. Only binary indicators (and not specific diagnoses or ICD-10 codes) were included in the dataset. Each CMC was marked as having either a single chronic condition or multiple chronic

conditions based on these clinical indicators. A binary indicator of the presence of technology assistance was also included.

Initial length of stay was calculated as the difference between the admission and discharge dates for each CMC's initial hospitalization. Seven- and 30-day readmission indicators for the initial hospitalization were also included. The Alberta Health Services (AHS) Pharmaceutical Information Network (Government of Alberta, 2012) was used to identify number of chronic medications.

Deaths in the cohort were identified through Alberta Vital Statistics (Government of Alberta, 2017).

1.7.4 Socioeconomic Variables

Socioeconomic data were linked to clinical data via residential postal code (at initial discharge). Residential postal code was also used to identify the AHS zone where each CMC was living. AHS divides the integrated health system in Alberta into five distinct zones, each of which has its own health care resources. For example, the Edmonton and Calgary zones are the most populous and have dedicated pediatric acute care centres whereas the other, less populous zones do not.

MyChild's socioeconomic data includes deprivation scores from the Pampalon index, which measure deprivation through census data at the dissemination-area (DA) level. DAs (each typically including 400–700 people) are the smallest geographic unit in the Canadian census and are socioeconomically homogeneous (Predy et al., 2008, Pampalon et al., 2009). Specifically, the Pampalon index uses six DA-level measures to measure two types of deprivation: material deprivation and social deprivation. The material score is computed using,

1. the proportion of individuals aged 15 years or older who are without a high school education,
2. the employment ratio of people 15 years or older, and
3. the average income of people 15 years or older.

The social deprivation score is computed from,

1. the proportion of people 15 years or older who are living alone;
2. the proportion of people aged 15 years or older who are separated, divorced, or widowed; and
3. the proportion of single-parent families.

Scores for each component were generated through principal component analysis. The scores were further discretized into quintiles (i.e., groups of 20%), with the first and fifth quintiles representing low and high deprivation, respectively.

Lastly, the use of mental health resources by the mothers of CMCs prior to index admission was also collected from DAD and NACRS. This categorical variable indicates if the mother had a mental health encounter in an inpatient facility or an ED in the 12 months prior to their child's admission, or if no data was available.

1.8 Dissertation Structure

This dissertation is structured in a paper-based format that follows the progression of its overall analysis and discussion. Chapter 2 is a systematic review that has been published in the Journal of Child Health Care (Sidra et al., 2022). The review addresses Research Question 1 through an analysis of reported costs and health care resource use by CMCs.

Chapter 3 addresses Research Question 2 through a quantitative, inferential analysis of the CMC population in Alberta with a mixed-effect hurdle regression model. This chapter presents a longitudinal analysis of CMCs that outlines time-varying trends in resource use (hospital days and ED visits) and associations with clinical and socioeconomic factors.

Chapter 4 presents a predictive analysis of short- and long-term CMC resource use in Alberta through boosted, tree-based regression models. To address Research Questions 3–4, this chapter identifies predictors of hospital days and ED visits over one- and five-year time frames and ends with a discussion of the potential of administrative data and electronic health records to support machine-learning-based analyses of CMCs in the future.

Lastly, Chapter 5 summarizes the major findings of this dissertation and discusses the contributions of this work together with its strengths and limitations. This chapter also outlines recommendations for future work and knowledge translation based on this dissertation's main findings. Specifically, these recommendations highlight how the results herein can enhance CMC research and care.

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Chapter 2. Reported Costs of Children with Medical Complexity - A Systematic Review

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This has been published as Sidra, M., Sebastianski, M., Ohinmaa, A., & Rahman, S. (2022). Reported costs of children with medical complexity—A systematic review. Journal of Child Health Care, 13674935221109683. Advance online publication. https://doi.org/10.1177/13674935221109683

2.1 Abstract

Examining reported costs for Children with Medical Complexity (CMCs) is essential because costing and resource utilization studies influence policy and operational decisions. Our objectives were to (1) examine how authors identified CMCs in administrative databases, (2) compare reported costs for the CMC population in different study settings, and (3) analyze author recommendations related to reported costs. We undertook a systematic search of the following databases: Medical Literature Analysis and Retrieval System Online, Excerpta Medica database, Cumulative Index to Nursing and Allied Health Literature, and Cochrane Library with a focus on CMCs as a heterogeneous group. The most common method used n = 11 (41%) to identify the CMC population in administrative data was the Complex Chronic Conditions methodology. The majority of included studies reported on health care service costs n = 24 (89%). Only n = 3 (11%) of the studies included costs from the family perspective. Author recommendations included standardizing how costs are reported and including the family perspective when making care delivery or policy decisions. Health system administrators and

policymakers must consider the limitations of reported costs when assessing local costing studies or comparing costs across jurisdictions.

2.2 Introduction

Children with Medical Complexity (CMCs) are a heterogeneous pediatric population that span a wide range of clinical diagnoses such as Neuromuscular and Cardiovascular chronic conditions and Congenital/genetic defects (Cohen et al., 2011). CMCs require considerable health care resources, have poor health outcomes, and place a substantial financial and psychological burden on caregivers (Berry et al., 2015; Cohen et al., 2011). While CMCs make up roughly 1% of the pediatric population, they account for 30% of pediatric health care resource use (Berry et al., 2015; Cohen et al., 2012a). Understanding health care and family costs are essential for clinicians, researchers, and policymakers to improve health care delivery and support for CMCs and their families.

Children with Medical Complexity share four key characteristics; the presence of one or more complex chronic conditions, reliance on technology for survival (e.g., feeding tubes or tracheostomies), high health care utilization (e.g., requiring several specialist care visits and frequent hospitalizations), and significant caregiver needs (e.g., care coordination, financial and social needs) (Cohen et al., 2011; Dewan and Cohen 2013). While identifying CMCs at an individual level is accomplished through clinical assessment, identifying CMCs at a population level can be challenging (Berry et al., 2015). According to Berry et al. (2015), the three main challenges to identifying CMCs at a population level are (1) how medical complexity is defined, (2) discrepancies in health care data collection and reporting, and (3) CMCs, unlike adults with medical complexity, tend to have multiple chronic conditions without any being dominant or

prevalent across the population. In addition to the heterogeneity of CMCs, these challenges make it difficult to report on health care costs for this population accurately.

This review examined the type and methods of reporting costs for this population. Such a review is essential as cost information can shape health care policy, care service delivery and research priorities. To our knowledge, this is the first review of this kind.

2.3 Aims

This systematic review had three aims: (1) to determine how CMCs are identified, (2) to analyze and compare reported costs for the CMC population, and (3) to examine author recommendations related to the costs reported in each included study.

2.4 Methods

A protocol was developed (not published), using the Cochrane Handbook for Systematic Reviews of Interventions and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) method for systematic reviews (Moher et al., 2009).

2.4.1 Search strategy

We developed the search strategy in consultation with a research librarian and peer-reviewed by a second librarian. We searched for peer-reviewed English language articles in the following databases: Medical Literature Analysis and Retrieval System Online (Medline) (1946–April 22, 2019), Excerpta Medica dataBase (EMBASE) (1974–April 22, 2019), Cumulative Index to Nursing and Allied Health Literature (CINAHL) (inception to April 22, 2019), and Cochrane Library (inception to Current). An online search of non-peer-reviewed articles was also conducted using Google search engine but did not yield additional studies.

2.4.2 Selection criteria

Studies were included if they reported costs for a pediatric population with Neurological Impairment, Complex Chronic Conditions (CCCs), use for Technology Assistance, high resource use, or high family care needs as described in the conceptual framework for CMCs reported by Cohen et al., (2011). Studies not examining CMCs as a heterogeneous group were excluded.

Study designs included were primary studies and literature reviews; letters, commentaries and opinion pieces were excluded. The outcomes of interest were to examine how CMCs were identified, compare what and how cost estimates were reported and examine author recommendations related to the measurement of CMC-related costs.

2.4.3 Study selection

The selection of studies was a two-step process. The first screening involved examining study titles and abstracts against pre-determined selection criteria using Microsoft Excel© 2016 (Microsoft Corporation, Redmond, WA, USA) by two independent reviewers (MSi, MR). The second screening stage involved examining the full text of studies identified in stage one and was completed by the same reviewers (MSi, MR). Any differences underwent discussion and were resolved by consensus. A third reviewer was available in case the two reviewers could not reconcile differences but was not needed.

2.4.4 Data collection and analysis

A piloted data collection tool Microsoft Excel© 2016 (Microsoft Corporation, Redmond, WA, USA), was used to extract the following data: study details (e.g., author, year, country, study design, and objective), population characteristics (e.g., inclusion and exclusion criteria,

sample size, participant age), and study costs and related outcomes (e.g., the definition used to identify CMCs, method used for evaluation of an intervention, costs reported and related author recommendations). One reviewer (MSi) extracted data, and a second reviewer (MSe) verified a random selection of studies.

Data were then organized by type of study topic. For example, some studies used descriptive analysis of health care administrative data, whereas others conducted an economic evaluation of a specified intervention. The types of costs analyzed included large-scale macro-costing, local level micro-costing, and economic assessment studies reporting costs for the population. Studies were then analyzed for similarities and differences in (1) how the study defined the CMC population, (2) type of costs reported, and (3) cost-related author recommendations.

Quality appraisal of the included studies was conducted in duplicate by two reviewers (MSi and MSe), and conflict was resolved through consensus. The Critical Appraisal Skills Program (CASP) (Critical Appraisal Skills Programme, 2020) for cohort studies (CASP Cohort Study Checklist) was used to assess 26 (96%) of the studies, while the CASP Economic Evaluation Checklist was used for one (3%) study (Fields et al., 2018).

The three domains of quality assessment in the CASP tool are validity of results, study precision and applicability. Results are included in Table 2.1. Each included study was rated using the tool's assessment questions as follows:

1. Are results valid? This domain was summarized as either high, medium, or low validity based on clear objectives, appropriate cohort recruitment and accounting for outcome bias and confounding measures.

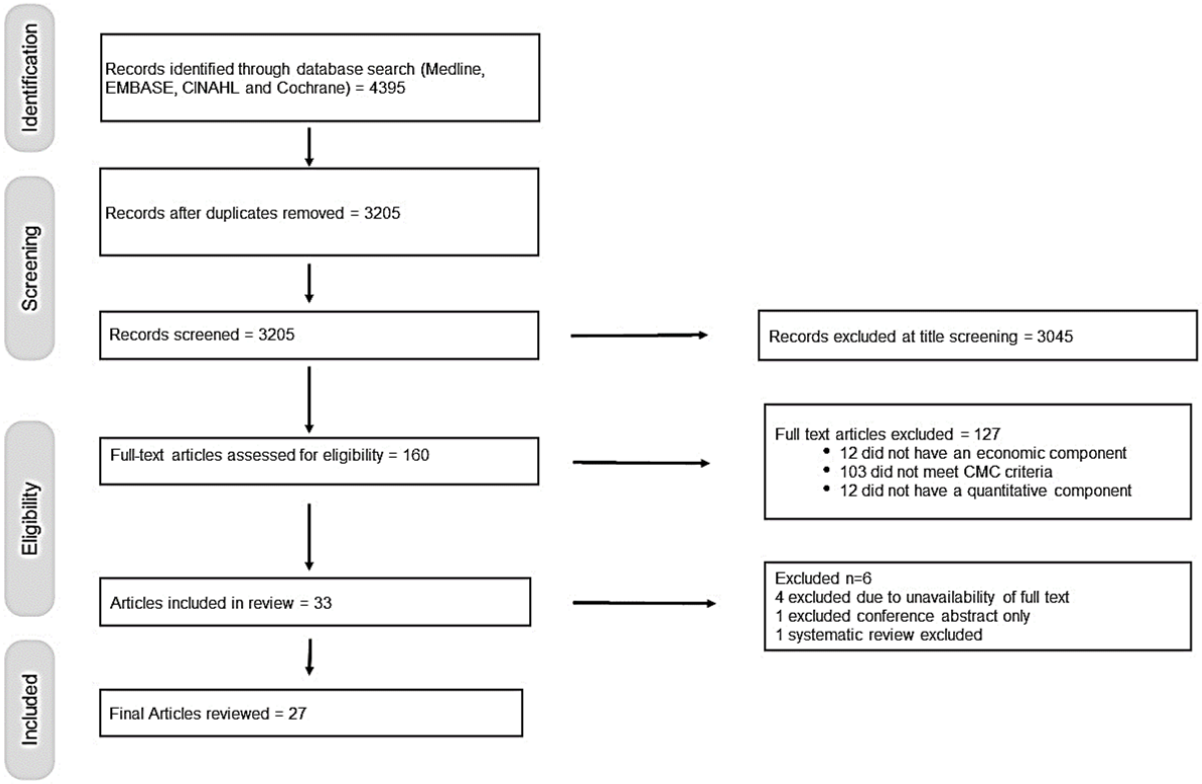
2. Are results precise? This domain was summarized as either precise, somewhat precise and not precise/unsure. We determined precision based on sample size and spread of the confidence intervals or inter-quartile.
3. Are results applicable? This domain category was summarized as either high, medium or low based on the generalizability of study results, the applicability of results to local CMC populations, and how similar they were to other evidence in the literature.

The CASP Economic Review Checklist was used to examine one process improvement study as it was not a cohort study.

2.5 Results

The initial search identified 4395 studies in total, and 3205 studies remained after removing duplicates, as illustrated in Figure 2.1. Following initial title and abstract screening, 160 titles remained for full-text screening. The full-text review identified 33 papers for data extraction; however, six papers were later excluded because they were conference presentations or full text was not available. A systematic review was excluded to prevent bias as it included similar studies to those in this review.

Figure 2.1. Study Selection Flow Diagram



Of the n = 27 included studies, the majority n = 24 (89%) were from the United States; two were from Canada and one from Australia (Table 2.1). The earliest study was from 2007 (Gordon et al., 2007), and the most recent was published in 2019 (Chirico et al., 2019; Ronis et al., 2019). The ages of participants in the studies varied from birth to 18 years, while three studies included older participants up to 24 years old (Chirico et al., 2019; Goldhagen et al., 2016; Murtagh Kurowski et al., 2014). These studies were included in this review despite having participants older than 18 years as they reported on CMC's transition from pediatric to adult care, an important consideration for this population. Table 2.1 outlines the included studies, summarizing how CMCs were identified, reported costs and data sources, author recommendations related to costs and qualitative appraisal results.

Table 2.1. Characteristics of Included Studies and Quality Appraisal Results

First Author (Year) / Country	Study Design / Sample Size	Study Focus	Method used to identify CMCs	Reported Costs and Data Sources	Main Findings	CASP* Evaluation Results		
			Focus of this review			Valid	Precise	Applicable
Descriptive Studies								
Agrawal, R., et.al., (2016), USA	Retrospective Cohort, sample size = 48743	Describes characteristics and trends of pediatric high resource users in Medicaid	Three groups; (1) children whose health system costs fell below the top 5% and did not return to the top 5% (2) children whose spending fell below the top 5% but returned to the top 5% (3) children who's spending consistently remained in the top 5%.	Utilization rates reported - CMCs relative to other pediatric populations. Administrative and claims data from 10 state Medicaid programs	One half of the top 5% of children resource utilizers in Medicaid will not continue to be in the top 5% while the other half will either stay in the top 5% or intermittently fall off and return to the top 5%.	High	Precise	Medium
Ananth, P., et.al., (2015), USA	Retrospective Cohort, sample size = 1252	Describes characteristics of children who died in 2012 and hospital use of type and number of life-threatening complex chronic conditions	Feudtner et. al, definition of complex chronic conditions (CCCs).	Costs were derived by converting charges to costs and adjusting for inflation. Hospital costs for last year of life by type and number of Complex Chronic Conditions. National administrative	Hospital use was greatest among children with hematologic/immunologic conditions and children with > 3 life threatening complex chronic conditions (LTC-CCC). Hospital use for children with LTC-CCCs in the last year of life varies significantly across the type and number of conditions.	High	Precise	Medium

				data from 40 US Children's Hospitals				
Bekmezian, A., et.al., (2008), USA	Retrospective Cohort, sample size = 816	Compares costs of patients with subspecialty needs in a staff only general pediatric hospitalist group with subspecialty/faculty /house staff gastroenterology, hematology and oncology groups.	3M's Clinical Risk Group categories.	Reported actual direct hospital costs. Local administrative data and direct costing supplied by the study hospital.	Patients in a staff-only hospitalist group experienced a 38% reduction in LOS and 29% reduction in direct costs compared to patients admitted to a faculty/house staff system.	High	Precise	Medium
Berry, JG., et.al., (2011), USA	Retrospective Cohort, sample size = 1083	Describes characteristics of hospitalizations for children with multiple, chronic conditions.	(1) Feudtner et. al, Complex Chronic Condition, (2) Neurologic impairment and (3) Technology assistance.	Costs reported as utilization rates. Administrative Data from 4 pediatric hospitals.	Better provider communication during transition from hospital to home, can prevent hospital readmissions. High readmission rates in children with technology	Medium	Precise	Medium
Berry, JG., et.al., (2017), USA	Retrospective Cohort, 2.26 million hospital discharges	Identifies which specific chronic conditions of children with multiple chronic conditions are associated with high hospital resource use	Health care Research and Quality's Chronic Condition Indicators.	Hospital costs derived by converting charges to costs and adjusting for inflation. National administrative data from all USA acute care hospital discharges.	CMCC accounted for over one-fourth of acute-care hospitalizations and one-half of all hospital dollars for US pediatric care in 2012.	Medium	Precise	High

Berry, JG., et.al., (2014), USA	Retrospective Cohort, sample size = 214765	Describes expenditures of CMCs and present a business case for reducing cost and reinvesting in outpatient and community care management.	Health care Research and Quality's Chronic Condition Indicators.	Reported costs derived using standard costing and administrative data. Multiple administrative databases including public payer direct billing data (Medicaid).	Cost savings from a small reduction in hospital days could pay for more expensive care management for a small proportion (most acute) of CMC. While, less intensive care management could be delivered to a larger portion of CMC. Business case did not report how much families of CMC spend on care management activities in their home.	Medium	No / unsure	Medium
Chan, T., et.al., (2016), USA	Cross-sectional, sample size = 136133	Describes the use of critical care resources for children with differing medical complexity in pediatric ICUs in tertiary-care children's hospital.	Critical care defined as admission to a pediatric ICU or pediatric cardiac ICU. Medical complexity categories i.e., no chronic condition, noncomplex chronic conditions, complex chronic conditions	Hospital costs derived by converting charges to costs and adjusting for inflation. National administrative data from 40 pediatric hospitals.	ICU resource use was greatest among patients with complex chronic disease compared with children with no complex chronic disease or no chronic disease.	Medium	No / unsure	Medium
Chirico, J., et.al., (2019), USA	Retrospective Cohort, sample size = 224	Describes characteristics of patients who died in a community-based palliative program and examine cost	All children who were enrolled in Compassion Net (community based palliative care program) between 2008 and 2015.	Reported costs derived using standard costing and administrative data.	Location of death, malignancy diagnosis and participation in Medicaid all were significantly related to	High	Precise	Low

		differences between location of death		Local and national administrative databases linked with insurance claims data to derive costs.	total cost in final year of life.			
Cohen, E., et.al., (2012), Canada	Retrospective Cohort, sample size = 15771	Evaluates health care utilization and costs in a population-based sample of CMC in Ontario, Canada	Complex Chronic Conditions All children aged newborn to 16 years who were hospitalized between Apr 1, 2005 and Mar 31, 2007. ICD-10 diagnostic codes within 3 clinical categories relevant to CMC: NI, complex chronic conditions (CCCs), and TA (Technology Assistance).	Reported costs derived using standard costing and administrative data. Provincial linked administrative data with public payer costs.	Hospitals wanting to discharge patients sooner without increasing risk of readmission should consider targeting care coordination for this high user group.	High	Precise	High
Coller, RJ., et.al., (2018), USA	Randomized Control trial, sample size = 147	Examines the effect of a caregiver coaching intervention, Plans for Action and Care Transitions (PACT), on hospital use among CMCs within a complex care medical home at an urban tertiary medical center.	Complex Chronic conditions (CCCs) as defined by Feudtner et al. and included need for technology assistance.	Costs reported as charges for hospital and other interventions. Institutional administrative database.	Those enrolled in PACT had fewer all-cause 30-day readmissions and were less likely to experience at least 1 readmission.	High	No / unsure	Low

Gold, JM., et.al., (2016), USA	Retrospective Cohort, sample size = 954,018CMC at 44 children's hospitals	Assesses the impact of, risk factors for, and variation across children's hospitals regarding long LOS (10 days) hospitalizations in CMC	3M's Clinical Risk Group categories.	Hospital costs reported by converting charges to costs and adjusting for inflation. National administrative data	The characteristics most strongly associated with LOS 10 days were use of the ICU, respiratory complex chronic condition, and transfer from another medical facility at admission. Efforts to reduce hospital costs in CMC might benefit from a focus on prolonged LOS.	High	Precise	Medium
Murtagh K., et.al., (2014), USA	Cross-sectional, sample size = 0-17 years = 577507 patients, 18-24 years = 482785 patients	Generates nationwide estimates for Emergency Department (ED) use by children and young adults with complex chronic conditions. Evaluate if being of the age for transition to adult care significantly affects the site of care and likelihood of hospital admission.	Patients with one or more complex chronic conditions, between the ages of 0 and 24 years, who visited an ED at least once during 2008.	Hospital costs reported as charges. National administrative database.	The majority of visits by pediatric- and transition aged patients with chronic complex conditions are into general EDs.	High	Precise	Medium
Ralston, SL., et.al., (2015) USA	Retrospective cohort, sample size = 11804	Describes patterns of care for a population-based cohort of children with medical complexity in northern New England	Complex Chronic conditions (CCCs) as defined by Feudtner et al.	Reported costs derived using standard costing and administrative data.	Hospital-specific resource utilization in children with medical complexity exhibits a high degree of variation, particularly around high cost, discretionary services.	High	Precise	Medium

				National administrative data.	Need for identifying best practices for this growing patient population.			
Srivastava, R., et.al., (2016) Australia	Retrospective Cohort, sample size = 48758	Describes the hospital costs, hospital types and differences across states and territories for CMCs cared for in Australian public hospitals	Children with complex chronic conditions, neurological impairment, and technology assistance	Reported costs derived using standard costing and utilization metrics. National administrative databases.	CMCs accounted for 32.1% of pediatric health care costs for a total cost of $620,948,769.	Medium	Precise	High
Evaluation								
Avritscher, E.B.C., et.al., (2018), USA	Pre-post and 2-phase stepped wedge design, sample size = 280	Assesses sustainability and scalability of medical home for high-risk children with medical complexity	>2 hospitalizations or >1 pediatric intensive care unit (PICU) admission in the year before enrollment in the medical home intervention	Costs reported by converting charges to costs and adjusting for inflation. Local hospital administrative data.	Findings support scalability of medical home type model of pediatric complex care. Such models are not adequately reimbursed. New alternative reimbursement mechanisms are needed.	High	Precise	Medium
Casey, P.H., et.al., (2011), USA	Pre-post without control, sample size = 225	Evaluates the effect of care coordination multidisciplinary clinic on Medicaid costs for CMC.	Children with at least 2 chronic medical conditions and who were followed by at least 2 pediatric subspecialists.	Local clinic costs and Medicaid data. Local and state (Medicaid) databases.	The program showed a decrease in total Medicaid costs for CMC.	High	No / unsure	Medium

Cohen, E., et.al., (2012), Canada	Pre-post without control, sample size = 81	Compares, pre- and post-implementation of integrated complex care coordination for CMC.	• Children (<16 years) with a known and/or suspected diagnosis of a complex chronic condition that is associated with medical fragility; (B) technology assistance and, (C) involvement of multiple specialists.	Reported costs derived using standard costing and administrative data. Family costs derived through administered surveys. Provincial administrative databases.	Formal partnerships between children's hospitals, community hospitals, families and primary care providers, in care coordination is a promising model for complex care delivery. Improvements in health care utilization and family-centeredness of care are possible with little to no changes in parental perceptions of the child's health.	Medium	No / unsure	Medium
Gay, J.C., et.al., (2016), USA	Retrospective cohort, sample size = 2783 hospitalized children receiving post discharge HH services across 19 states and 7361 matched controls not discharged to HH services.	Assesses the relationship between home health nursing care (HH) services and hospital use in Children	3 or more Complex Chronic Conditions Chronic condition defined as conditions expected to last >12 months and involve either several different organ systems or 1 organ system severely enough to require specialty pediatric care Medical technology to maintain a child's health status (e.g., gastrostomy, tracheostomy).	Costs estimated using charges to cost ratios adjusted for inflation. Local Case Mix database and National administrative data.	Children discharged to HH care experienced less hospital use than children with similar characteristics who did not use HH care.	Medium	No / unsure	Medium
Goldhagen, J., et.al., (2016), USA	Multi-method with primary data collection and retrospective cohort, sample size =	Evaluates the results from a 2007 study to demonstrate the impact of a community PedsCare program	Children with chronic life-limiting conditions including children already enrolled in hospice.	Costs reported using local institutional costs. Local institutional database.	Parents-caregivers reported overall positive perceptions with impaired emotional health, decision-making, social support, interaction and	Low	No / unsure	Low

	48 Children 53 Caregivers	(CBPPC) on Health-Related Quality of Life (HRQoL) and hospital utilization and costs.			communication, child health and self-efficacy in caring for their children, with higher HRQoL scores associated with longer periods of enrollment The utilization analysis showed reductions in hospital services during the pre-enrollment quarters compared to the quarters following their enrollment			
Gordon, J.B., et.al., (2007), USA	Pre-post without control, sample size = 227	Evaluates the impact of a tertiary care center special needs program (SNP).	Children with complex chronic conditions that involve several organ systems and require multiple specialists, technological supports, and community services.	Hospital costs reported as charges. Local institutional costs.	An investment of $400,000 per year was associated with more than 50% decrease in hospital days and $10.7M decrease in tertiary care center payments.	Low	No / unsure	Low
Ronis, S.D., et.al., (2019), USA	Time study, sample size = 208	Quantifies the time required to perform non-reimbursed care coordination activities by a multidisciplinary care coordination program for CMC and to estimate the direct salary costs of that time.	3 or more affected body systems Dependence on technology and/or reliance on caregivers for instrumental activities of daily living. Demographics Family characteristics Activities performed by staff that are not reimbursed and time spent on those activities.	Costs were reported by multiplying non-reimbursed worked hours by staff salary costs. Staff hourly costs were based on national employment statistics.	In the context of a children's hospital that is generally well staffed to care for these children, care coordination activities required conservative estimate of $145 to $210 in unreimbursed salary per child per month, not including fringe benefits, management or overhead costs.	High	No / unsure	Medium

				Hours of work recorded by participants.				
Shumskiy, I., et.al., (2018), USA	Retrospective Cohort, sample size = 93121	Assesses correlations between years of Well Child Visits (WCV) with hospitalizations for CMC.	Feudtner pediatric Chronic Complex Conditions Agency for Health care Research and Quality's Chronic Condition Indicators (CCI)	Health service utilization rates. State Medicaid database.	Most Medicaid-insured CMC do not receive annual WCVs consistently over time. Children with fewer annual WCVs have a higher likelihood of hospitalization.	High	Precise	Medium
Simon, T.D., et.al., (2017), USA	Cluster randomized, sample size = Control group n=150. Intervention group n=181. Primary Care Physicians = 200	Assesses whether children with medical complexity (CMC) exposed to a hospital-based comprehensive case management service (CCMS) experience improved health care quality, improved functional status, reduced hospital-based utilization, and/or reduced overall health care costs.	3M Clinical Risk Group categories Identification by participating Primary Care Physicians.	Reported costs derived using standard costing and administrative data. Program implementation costs; net cost after accounting for clinic revenues. Local institutional database and third-party payers.	Exposure to a hospital-based CCMS focused on care coordination for CMC generally improved quality of care, did not change functional status or hospital-based utilization, and increased overall health care costs. Tiered care providing care coordination services commensurate with disease severity may be a more cost-effective approach for CMC.	Medium	Precise	Medium
Van Orne, J., et.al., (2018), USA	Quasi-experimental, sample size = 38 patients, 34 primary caregivers plus a similar size control group.	Determines the effectiveness of a structured boot camp–style pre-discharge training program for caregivers of pediatric	Patients with complex medical conditions who are dependent on technology and have newly placed tracheostomies.	Savings reported are derived from local administrative data.	Statistically significant decreases were found in; Mean unit length of stay (92 vs 60 days), mean discharge training days (60 vs 16 days; and	High	Precise	Low

		patients with complex medical conditions		All costs were not reported; however, cost savings were reported and calculated based on median Length of Stay for intervention group versus control group. Local institutional costs.	median total parental stress score. More than 90% of caregiver participants were very satisfied with the program. Cost savings were estimated at between $53 300 and $69 900 per patient.			
Walter, A.W., et.al., (2018), USA	Retrospective Cohort, sample size = 7286570 -Nonchronic (76.8%) - Noncomplex chronic (16.6%) - Complex chronic (6.6%)	Assesses health care utilization and costs for children enrolled in private insurance plans in the United States	The Pediatric Medical Complexity Algorithm, a modification of the Chronic Disability Payment System, was used to identify and stratify children aged 0–17 years by levels of medical complexity.	Reported costs derived using standard costing and administrative data. Commercial claims data (employer sponsored health insurance).	Families of children with medical complexity are 1.9–3.8 times as likely as those without chronically ill children to spend over US$1000 out of pocket per year on children's medical services. Medical spending on children is not the only source of financial burdens for these families.	High	Precise	Low
Other								

Thomson, J., et.al. (2016), USA	Survey design, sample size = 167 CMCs (111 responded to survey)	Describes hardships experienced by families of children with medical complexity (CMC) and compare them with those experienced by families of children with asthma	Severe, chronic disease, and receive care from more than or equal to 3 clinical subspecialists.	Costing using local administrative costing. Survey data.	Families of CMC may face greater challenges than previously recognized. Families of CMC frequently report financial and social hardships, often at rates higher than families with asthma who were generally of lower socioeconomic status	Medium	Precise	Low
Fields, E., et.al. (2018), USA	Pre-Post without control. sample size = NA	Uses Lean methodologies to standardize processes, and eliminate waste in the provision of medical equipment.	All children needing to access medical equipment supplies	Cost assumptions were based on median local staff salaries. Data collected at implementation of intervention.	The median lead time for processing DME/supply requests decreased from 50 days (range 42–80 days) to 3 days (range 1–17 days; $p<0.0001$). The annual cost of completing DME/supply requests decreased from an estimated $29,000 to $18,000.	A different toll, the CASP economic Review, was used to evaluate this study. Although the intervention showed a positive economic benefit the results are not applicable outside of the study area.		

*Critical Appraisal Skills Program (CASP) tool for cohort studies was used for every study except for Fields, E (2018) where the CASP tool for Economic Evaluation was used.

We summarized the CASP Cohort evaluation tool findings as high, medium or low based on clear objectives, appropriately recruited cohort, and adequately accounting for outcome bias and confounding measures. Precision was summarized as precise, somewhat precise and not precise/unsure based on sample size and spread of confidence intervals or inter-quartile range reported. Finally, applicability of results was summarized as high, medium or low based on study design and how similar the results were to other evidence.

Cost Methods and data sources are further summarized in Table 2 – Cost Variable Summary.

Studies were grouped under Descriptive Studies category if they identified CMCs costs and characteristics but were not an evaluation of an intervention.

Studies were grouped under Evaluative Studies category if they included an evaluation of an intervention.

Studies were grouped under other category if they did not fit with Descriptive or Evaluative Studies.

2.5.1 Identifying the children with medical complexity population

The majority n = 18 (67%) of studies included in this review used one of four CMC identification methods (Table 2.1) as described by Berry et al. (2015). The most common method used n = 11 (41%) was Feudtner et al. (2000) Complex Chronic Conditions. 3M Clinical Risk Groups 3M Science (2020), were used by n = 3 (11%), Chronic Condition Indicators Agency for Health care Research and Quality (2020), was used by n = 2 (7%), and Patient Medical Complexity Mangione-Smith (2014), was used by n = 1 (3%) of the studies. Technology assistance was identified in seven (25%) studies as part of their CMC definition.

2.5.2 Costing methods

Type of reported health care costs varied and included standard costing methodology, such as case costing or standard hospital costs *n* = 8 (30%), local direct care costs *n* = 7 (26%), costs converted from hospital charges *n* = 6 (22%), hospital charges not converted to costs *n* =3 (15%), and utilization rates including relative utilization rates *n* = 3 (15%), as outlined in Table 2.1. Costs to CMC families was reported in *n* = 3 (11%) of studies. This information was captured in family surveys. Data sources varied and included national administrative databases, claims data, provincial or state-wide databases, private payer data, and local administrative and costing data.

A majority of studies *n* = 24 (89%) included hospital-related costs while only *n* = 3 (11%) included both health system and family costs, the most under-reported cost variable shown in this review. Ten (37%) studies reported on outpatient costs, the second most utilized cost variable. Cost variables are summarized in Table 2.2.

Children with medical complexity costs at a hospital, geographic or national level

accounted for n = 14 (52%) descriptive studies. In evaluative studies, n = 11 (40%) included costing of a given intervention. Types of interventions included care coordination, medical home-like support models and CMC family/caregiver educational programs. The remaining two studies, n = 2 (7%), did not fit into the descriptive or evaluative category; one compared CMC family experience and financial hardship with families of asthmatic children, and the other reported a process improvement intervention to improve access to technology for CMCs (Table 2.1).

A review by Gold et al. (2016) found the prevalence of lengthy hospitalizations for CMC varied significantly by type of chronic condition and that no dominant chronic condition was evident. This lack of a dominant chronic condition presents a challenge in effectively measuring the burden of the disease as each specific ailment has different acuities and, therefore, different financial and social burdens on patients, families and the health system (Berry et al., 2011; Cohen et al., 2012a; Kuo and Houtrow, 2016).

All three (11%) studies that included families' costs identified a significant financial impact to CMC families/caregivers. After examining 7.2 million children's records, including CMCs enrolled in private insurance plans in the United States, Walter et al. (2018) found that families of CMCs are almost four times more likely to spend over $1000 out of pocket per year than non-CMCs. The authors also noted that this did not include any lost income due to reduced working hours to care for a CMC.

Only two studies (7%) in this review included a health state assessment such as cost-utility analysis or Patient Reported Outcome measures for families/caregivers. Families and caregivers of CMCs are more likely to score their health lower than parents of healthy children or children with single chronic diseases (Dewan and Cohen, 2013).

Cohen et al. (2012b) and Goldhagen et al. (2016) included Health State Quality of Life

measurement as part of the evaluation of care coordination. Both studies suggested that improving care delivery models such as partnerships between children's hospitals and care coordination programs can positively affect family caregiver quality of life and have a positive impact on health care costs for the child and the caregiver.

Table 2.2. Cost Variable Summary

Cost Variable Considered	# of Studies Included Cost Variable
Hospital costs including Admissions, hospital days, hospitalization rates, inpatient costs, Length of Stay, ICU days, post ICU days, total hospital costs, readmissions and physician compensation. (Agrawal et al., 2016; Ananth et al., 2015; Avritscher et al., 2019; Bekmezian et al., 2008; Berry et al., 2011; Berry et al., 2017; Berry et al., 2014; Casey et al., 2011; Chan et al., 2016; Chirico et al., 2019; Cohen et al., 2012a; Cohen et al., 2012b; Coller et al., 2018; Gay et al., 2016; Gold et al., 2016; Goldhagen et al., 2016; Gordon et al., 2007; Murtagh Kurowski et al., 2014; Ralston et al., 2015; Shumskiy et al., 2018; Simon et al., 2017; Srivastava et al., 2016; Thomson et al., 2016; Van Orne et al., 2018; Walter et al., 2018)	n=24 (89%)
Outpatient costs including specialist visits, day surgery visits, clinic costs. (Agrawal et al., 2016; Avritscher et al., 2019; Berry et al., 2014; Casey et al., 2011; Cohen et al., 2012a; Cohen et al., 2012b; Ralston et al., 2015; Shumskiy et al., 2018; Simon et al., 2017; Thomson et al., 2016; Walter et al., 2018)	n =11 (40%)
Primary Care (Agrawal et al., 2016; Avritscher et al., 2019; Berry et al., 2014; Casey et al., 2011; Cohen et al., 2012a; Cohen et al., 2012b; Gordon et al., 2007; Shumskiy et al., 2018; Simon et al., 2017; Van Orne et al., 2018)	n =10 (37%)
Emergency department (ED) costs including ED visits, type of ED (pediatric vs. adult), and admission or transfers from ED. (Berry et al., 2014; Casey et al., 2011; Chan et al., 2016; Cohen et al., 2012a; Murtagh Kurowski et al., 2014; Ralston et al., 2015; Shumskiy et al., 2018; Srivastava et al., 2016; Walter et al., 2018)	n =10 (37%)
Pharmaceutical (inpatient or outpatient) excluding out of pocket family costs. (Agrawal et al., 2016; Bekmezian et al., 2008; Berry et al., 2014; Casey et al., 2011; Chan et al., 2016; Cohen et al., 2012a; Goldhagen et al., 2016; Simon et al., 2017; Srivastava et al., 2016; Walter et al., 2018)	n =10 (37%)
Diagnostic testing including medical imaging and/or laboratory (Avritscher et al., 2019; Bekmezian et al., 2008; Berry et al., 2014; Cohen et al., 2012a; Goldhagen et al., 2016; Ralston et al., 2015; Simon et al., 2017; Srivastava et al., 2016)	n =9 (33%)
Home Health Care (does not include family or other in-kind home care contributions) (Agrawal et al., 2016; Berry et al., 2014; Chirico et al., 2019; Cohen et al., 2012a; Cohen et al., 2012b; Gay et al., 2016; Gold et al., 2016; Simon et al., 2017)	n =8 (29%)
Therapeutic care including physical therapy, occupational therapy and/or speech therapy. (Agrawal et al., 2016; Berry et al., 2014; Casey et al., 2011; Cohen et al., 2012a; Goldhagen et al., 2016; Simon et al., 2017; Van Orne et al., 2018; Walter et al., 2018)	n =8 (29%)

Mental health Services (inpatient and outpatient) (Agrawal et al., 2016; Berry et al., 2017; Chan et al., 2016; Cohen et al., 2012a; Srivastava et al., 2016; Walter et al., 2018)	n =6 (22%)
Care Coordination activities including family training programs (excluding home care). (Cohen et al., 2012b; Simon et al., 2017; Thomson et al., 2016; Van Orne et al., 2018)	n =5 (18%)
Family costs including any out-of-pocket expenses and opportunity costs. (Cohen et al., 2012b; Thomson et al., 2016; Walter et al., 2018)	n =3 (11%)

Cost variables were reported differently throughout the included studies. For example, some hospital costs were reported as charges, converted to costs or reported as hospital days.

2.5.3 Author recommendations

Table 2.3 summarizes the author recommendations and outlines four high-level themes; health spending and resource utilization, care coordination, service delivery and family perspective. Care coordination and a medical home-like model for CMCs post-acute care discharge was the focus of eight (30%) (Avritscher et al., 2019; Casey et al., 2011; Cohen et al., 2012b; Gay et al., 2016; Goldhagen et al., 2016; Gordon et al., 2007; Simon et al., 2017; Van Orne et al., 2018) included studies. All but one of the care coordination evaluation studies reported cost savings to the health system through reduced length of stay, hospital admissions, and readmissions. Unlike the rest of the intervention studies, Simon et al. (2017) found that while hospital-based comprehensive case management service improved quality of care for the CMC population, it did not change hospital-based utilization and increased overall health care costs.

Another set of recommendations addressed clinical service delivery for CMCs. A key finding was the need for standardized protocols for clinical practice for CMCs as a means of enhancing care for CMCs and reducing cost variation (Chan et al., 2016; Ralston et al., 2015; Srivastava et al., 2016). Srivastava et al. (2016) suggested that measuring variation in medical and surgical pediatric conditions costs might provide more insight into cost variation. Similarly, Berry et al. (2017) found that better provider communication during transition from hospital to home could prevent hospital

readmissions. Cohen et al. (2012b) reported formal partnerships between children's hospitals, community hospitals, and primary care are beneficial in reducing CMC care costs. Alternative reimbursement methods for programs supporting CMCs were also recommended (Avritscher et al., 2019; Gordon et al., 2007; Ronis et al., 2019)

Table 2.3. Summary of Author Recommendations

Topic Theme	Summary of Author Recommendations
Health Spending and resource utilization patterns for CMCs	• Efforts to reduce hospital costs for CMC may benefit from focusing on long LOS. (Bekmezian et al., 2008; Gold et al., 2016) • Measuring variation in costs of medical and surgical paediatric conditions across hospital types might provide more context for cost variation. (Srivastava et al., 2016) • A publicly available database of health care costs and utilization to assess longitudinal trends in spending and health outcomes for CMC is needed. (Berry et al., 2014) • New alternative reimbursement methods for programs to support high-risk medically complex children are needed. (Avritscher et al., 2019; Gordon et al., 2007; Ronis et al., 2019)
Care Coordination	• Consider policies or programs that enhance and promote consistent utilization of primary care services for CMCs (Agrawal et al., 2016; Shumskiy et al., 2018) • Identifying CMC populations that are more likely to benefit from care coordination programs is beneficial for health system costs and families. It's also important to assess the effectiveness of different approaches to care. (Berry et al., 2014; Casey et al., 2011; Cohen et al., 2012; Gay et al., 2016; Simon et al., 2017; Van Orne et al., 2018) • Further studies, examining care coordination type programs, should include more rigorous designs such as RCTs and studies that examine the population for a longer time frame.(Gay et al., 2016) • Families, health systems and payers may all benefit from community-based pediatric palliative care programs for CMCs. (Chirico et al., 2019; Goldhagen et al., 2016)
Service Delivery	• Hospitals might benefit from exploring the efficiency and effectiveness of inpatient care delivery for CMCs with a chronic mental health condition. (Berry et al., 2017) • Consider recruiting clinicians and staff with the clinical competency to care for CMCs with the most common underlying conditions e.g., neurological impairment. (Berry et al., 2011) • Standardized care protocols for clinical practice including for common admission diagnoses to the ICU may be useful for CMCs. (Chan et al., 2016; Ralston et al., 2015)

	• Using Lean methods can be beneficial to improving equipment supply processing time and is beneficial to patients and staff. (Fields et al., 2018) • Future research for CMC population should include young adults who are cared for outside of pediatric-specific care center. (Murtagh Kurowski et al., 2014)
Family Perspective	• Need to identify key outcomes most important to patients, families, health care providers and payers. (Chirico et al., 2019; Goldhagen et al., 2016; Thomson et al., 2016; Walter et al., 2018) • Need to better understand the relationship between family-experienced hardships and resource utilization of CMC, as well as to identify characteristics of CMC (e.g., CCC category, type of technology dependence, receipt of home health care) associated with the most hardships. (Cohen et al., 2012; Thomson et al., 2016)

Themes from the included studies are summarized.
Further research on health economic assessment recommendations, especially those considering the family perspective are needed.

2.5.4 Quality of included studies

We assessed the quality of included studies using the CASP tool to examine strength of study designs and applicability of author recommendations. Table 2.1 includes a summary of the quality appraisal results.

Using the Cohort CASP tool, 15 (56%) of the studies rated as high validity, 9 (33%) medium validity, and two (7%) were low validity. For precision, 17 (63%) were rated as precise, and 9 (33%) were rated as not precise or unsure. In applicability, seven (26%) studies were rated as highly applicable, 16 (59%) were medium applicability, and seven (26%) were rated as low applicability. Finally, using the economic evaluation CASP tool, one (3%) study showed favorable results in reduced operating costs; the savings are specific to the local operational context and may not be applicable in other areas due to different operational structures and processes.

2.6 Discussion

This review aimed to examine reported costs for the CMC population, including methods used to identify CMCs and related recommendations. Results indicate that most reported costs are from the health care system's perspective, with significant variation in types and methods used to represent these costs for CMCs. Cost-related author recommendations covered health care spending, resource utilization, care coordination, service delivery and the need to include family perspective in re- porting costs for this population.

2.6.1 Identifying children with medical complexity

Identifying CMCs in administrative data can be challenging and may affect reported costs for the population (Berry et al., 2015). CMCs are identified in health administrative data using International Classification of Diseases (ICD) ICD-9 and 10 diagnostic codes using various methods.

Our review included studies using all four CMC identification methods based on Berry et al. (2015). While all these methods are capable of identifying CMCs, they may yield different cohorts. For example, Feudtner et al. (2000) would not identify noncomplex Chronic Conditions such as behavioral/mental health conditions, whereas the other three methods would. While Complex Chronic Conditions are clearly defined in Feudtner et al. (2000) and Patient Medical Complexity Algorithm methods, there is no clear definition for Complex Chronic Conditions in the Clinical Risk Groups and Chronic Condition Indicator methods (Berry et al., 2015).

Another limitation of all four methods is the dependence on administrative data, as they do not typically include patient and family-related costs or outcomes. Berry et al. (2015) describes strengths and limitations of each method in detail. While it may be possible to compensate for limitations of these methods through study design, it makes it more challenging to compare costs

and outcomes between studies. Future studies need to carefully consider challenges and limitations of using administrative data to identify CMCs and choose the most appropriate option for their study.

2.6.2 Reported costs—health system perspective

Variation in health resource utilization and other types of reported costs in the included studies was partially caused by differences in how data is collected and stored in administrative databases. While administrative databases are a reliable and accessible source of information, they also have well-documented limitations. Riley (2009) described several limitations of claims databases, such as the potential for claims data to contain biases as providers are inherently incentivized to maximize payments, limiting generalizability to a larger population. Limitations with claims data were especially applicable to for-profit health systems, such as the case with many of the studies included in this review.

Health care costs were reported as local costs converted by hospital-specific ratios ((AHRQ), 2020) or reported as hospital charges, making it difficult to compare costs across studies. This variation, along with variation in type of health care service cost included in costing data, provides an incomplete picture of CMC resource use. For example, only six (22%) of included studies reported on costs or utilization of mental health resources, even though according to Berry et al. (2017), mental health conditions were the most prevalent chronic conditions in acute-care hospitalizations and are associated with a significant number of pediatric hospital days.

Costs were also reported as a component of evaluating a given intervention, namely care co-ordination or patient/family education type programs. Consistent with a systematic review by Cohen et al. (2011), we found that most evaluations in the included studies are quasi-experimental

and did not include control groups. Nonetheless, intervention studies indicated that examining characteristics of the local CMC population is recommended as some CMCs may benefit from more intensive and often costlier care coordination services than other CMCs. This differentiation in CMC cohort needs is an essential finding for jurisdictions looking to design or improve their CMC care coordination programs.

Standardizing clinical care where appropriate was also identified as an important quality improvement activity that can reduce health system costs and improve care for CMCs and their families (Chan et al., 2016; Ralston et al., 2015). Chan et al. (2016) reported that CMCs make up the largest pediatric Intensive Care Unit (ICU) population, and that standardized care protocols for common admissions, and standard use of palliative care services can improve care for this population. A systematic review by Coller et al. (2014) found that discharge points, transfer from another medical facility, home care, technology supports, and post-discharge follow-up were important in reducing preventable hospitalizations for CMCs (Coller et al., 2014). Our study showed that many of these service categories were not consistently reported in included studies.

Health care activities not reimbursed by funding agencies were another area that was not widely considered. Imperfect measurement of reimbursed care activities is a potential barrier to effective and sustainable care coordination efforts (Avritscher et al., 2019; Gordon et al., 2007; Ronis et al., 2019). Ronis et al. (2019) estimated that health care staff provided approximately $2400 worth of unreimbursed care coordination time per CMC per year in undocumented activities such as review of medical records, paperwork, and multidisciplinary team meetings. Considering these costs in the design of CMC care programs ensures adequate funding and long-term sustainability.

The context of how health care is funded is also an important consideration. For example, in

the United States, while the Medicare program provides public health insurance for some, the private patient funded third-party insurance is the predominant model. Canada and Australia have more publicly funded health care services than the U.S. These important differences should also be considered when using health care costing data for policy decisions.

2.6.3 Family/caregiver perspective

Evidence from this review suggests a substantial gap in reporting of costs incurred by families and caregivers of CMCs. Included studies that reported CMC family costs showed that families of CMCs endure a substantial financial burden. For example, Walter et al. (2018), after examining 7.2 million children's records including CMCs that were enrolled in private insurance plans in the United States, found that families of CMCs are almost four times more likely to spend over $1000 out of pocket per year than non-CMCs. The authors also noted that this did not include any lost income due to reduction in working hours to care for a child with medical complexity. Another study by Thomson et al. (2016) showed that families of CMCs experienced more financial and social hardships.

Measuring and reporting family costs can be more arduous as it often involves survey design and implementation. Excluding family costs from program evaluation not only provides incomplete information but risks transferring the cost burden from the health system to individual CMC families, causing harm to patients and families. Prudent policy makers and funders would carefully examine the implications of funding decisions given the complexity of accurately measuring the patient and family burden for this population. It is critical to consider funding decisions from a larger system perspective that include social and health care system supports for this population.

2.6.4 Limitations

This study has several limitations that must be considered. We examined CMCs as a heterogeneous cohort, so costing evaluations of individual complex chronic diseases were not included. As a result, disease-specific costing assessments that were excluded may provide further insight into the financial burden of CMCs. A lack of European studies, with most of the included papers being from the United States n=24 (89%) is another limitation. This result may be due to the use of the term Children with Medical Complexity, the definition used for CMCs in this search, or because non- English papers were excluded. More research is needed to ascertain differences between North American and other countries' definitions of CMCs. Only peer-reviewed studies were included in this review, and unpublished studies may have different results or provide more insights into the gaps identified.

2.6.5 Implications for practice

While CMCs make up only a small fraction of all children (usually less than 1%), they are consistently identified as the highest utilizers of health care resources (Agrawal et al., 2016; Berry et al., 2017; Chan et al., 2016; Cohen et al., 2012a; Srivastava et al., 2016). Calculating family/ caregiver costs alongside health system costs may provide more insight on policy or operational gaps (Berry et al., 2014).

Health care administrators, policymakers, and researchers can benefit from the findings of this review by considering limitations of costing data when making policy or operational decisions. The type of data used for cost analysis, including claims data, charge data, or publicly funded system data, impacts how costing information is used and compared across jurisdictions. Variables such as unfunded care coordination time and examining trends over time would also help provide a clearer picture of costs for the local CMC population and inform local care policy or delivery decisions. (Agrawal et al., 2016; Berry et al., 2014; Shumskiy et al., 2018; Srivastava

et al., 2016).

2.7 Conclusion

Caring for CMCs requires a large, disproportionate amount of health care resources and exerts financial and psychosocial strain on families. This systematic review identified essential issues, including inconsistent reporting of health care costs and a need to consider the family perspective. Finally, identifying standardized methodologies for reporting costs for this population will improve the comparability and applicability of results across different jurisdictions.

2.8 Acknowledgments

The authors would like to thank Ms Linda G. Slatter for her support in development and execution of the search strategy and Dr David Wyatt Johnson for his review and recommendations for the manuscript. Dr David Wyatt Johnson, MD Senior Medical Director Maternal, Newborn, Child and Youth Strategic Clinical Network at Alberta Health Services. Linda G. Slatter BA, BEd, MLIS, John W. Scott Health Sciences Library (retired).

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Chapter 3. Clinical and Socioeconomic Associations with Hospital Days and Emergency Department Visits Among Medically Complex Children: A Retrospective Cohort Study

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This paper was submitted to Pediatrics Journal at time of writing.

3.1 Abstract

Objective. To estimate associations between clinical/socioeconomic variables and hospital days and emergency department (ED) visits for children with medical complexity (CMCs) for 5 years following initial admission.

Study design. Retrospective, longitudinal, population-based cohort study of CMCs in Alberta, Canada (n = 12 621) diagnosed 2010–2013 using administrative data (Alberta Health Services data repository) linked to socioeconomic data (Canadian census). The primary outcomes were annual cumulative numbers of hospital days and ED visits for 5 years following a CMC's initial admission. Data were analyzed using mixed-effect hurdle regression models.

Results. Among CMCs utilizing resources, those with more chronic medications had more hospital days (RD 3.331 for ≥5 vs 0 medications in year 1, SE 0.347, P < .001) and ED visits (RD 1.836 for 0 vs ≥5 medications in year 1, SE 0.133, P < .001). Among these CMCs, initial LOS had significant, positive associations with hospital days (RD 1.960–5.097, SE 0.161–0.610, *P* < .001 outside of the gastrointestinal and hematology/immunodeficiency groups). Those residing in rural/remote areas had more ED visits than those in urban/metropolitan locations (RD

1.727 for rural vs urban, SE 0.075, $P < .001$). Material and social deprivation had significant, positive associations with number of ED visits.

Conclusions. Clinical factors are more strongly associated with hospitalizations, and socioeconomic factors with ED visits. Policy administrators and researchers aiming to optimize resource use and improve outcomes for these children should engage with families and consider supports that address both clinical and socioeconomic needs.

3.2 Introduction

Children with medical complexity (CMCs) are a heterogeneous population characterized by multiple complex chronic medical conditions that often require a disproportionate amount of health care resources for daily living (Berry et al., 2015, Cohen et al., 2012, Dewan et al., 2013). Berry et al., 2017 reported that children with multiple chronic health conditions account for over one-fourth of acute care hospitalizations and one-half of all pediatric hospital dollars in the United States. The Canadian Institute for Health Information (CIHI 2020) attributes 57% of hospital care costs, 37% of hospital admissions, and 54% of hospital days among children in Canada to CMCs, while Srivastava et al., 2016 ascribes one-third of hospital costs for children in Australia's public hospitals to CMCs. An understanding of subgroups within this population and their resource utilization over time is needed to improve the efficiency of health care delivery and appropriate supports for patients and families.

While associations between clinical complexity and health service use are well established, (Dewan et al., 2013, Berry et al., 2017, Srivastava et al., 2016), the role of socioeconomic factors in how CMCs use health care resources is less understood. A high proportion of CMCs are in lower socioeconomic positions (Carrilero et al., 2020), where they have 3–7 times higher mental

health service use, hospitalizations, prescription drug use, and mortality (Garciá-Altés et al., 2018). Furthermore, the children of mothers with depressive symptoms have increased acute care resource use in the first 2–4 months of infancy (Minkovitz et al., 2005). Understanding how clinical and socioeconomic factors are associated with health care resource use, especially hospitalizations and emergency department (ED) visits, is essential to designing effective supports for CMCs (Hudson et al., 2013). While much of the research to date on CMC hospital admissions and ED visits has focused on readily available clinical data, relatively few studies have considered socioeconomic factors (Carrilero et al., 2020), and none have considered both simultaneously.

This population-based, longitudinal, retrospective cohort study examines associations between clinical and socioeconomic factors at initial discharge and subsequent hospitalization and ED use for the CMCs cared for in a publicly funded, integrated provincial health system in Canada.

3.3 Methods

3.3.1 Study Data

We extracted data for this study from 4 standardized Canadian, Alberta-specific health administrative data sets housed in the Alberta Health Services data repository (AHSDR): the Discharge Abstract Database (DAD), (CIHI, 2023) which contains hospital discharge data; the National Ambulatory Care Reporting System (NACRS), (CIHI, 2023) which contains data on hospital ED visits; Alberta Vital Statistics, which lists all births and deaths in Alberta; and the Pharmaceutical Information Network (PIN), which captures all pharmaceuticals dispensed in a

community setting. The AHSDR provides a unique identifier that links individuals across these data sets.

3.3.2 Study Population and Primary Outcome Variables

We identified CMCs through DAD using the ICD-10-based definition established by Cohen et al., (2012), and Feudtner et al., (2000) (ICD-based classification criteria appear commonly in the literature (Sidra et al., 2022)) and only considered CMCs who were Alberta residents, had an initial discharge and diagnosis between 2010 and 2013, and were at most 18 years old at initial admission (first admission with a Complex Chronic Condition diagnosis). For each eligible CMC, we obtained annual cumulative numbers of hospital days and ED visits from DAD and NACRS for the 5 years following initial hospital discharge; these serve as our primary outcome variables of interest.

3.3.3 Study Variables

Demographic and clinical data included age, sex, initial hospitalization length (in days), number of chronic medications, 9 indicators for chronic disease involving different body systems, the presence of technology assistance (e.g., hemodialysis), and 7-day and 30-day readmission following initial discharge. The technology assistance indicator was based on procedure codes established by Cohen et al., (2012). Number of chronic medications was extracted from PIN data for the 12 months following initial discharge. All variables were measured at initial admission unless otherwise specified.

Socioeconomic variables were derived from residential postal code at initial discharge. Alberta Health Services classifies regions of Alberta into 6 rurality categories ranging from metropolitan to remote rural and classifies residents into socioeconomic groups using the

Pampalon Deprivation Index (Predy et al.,2008). The Pampalon index incorporates 6 variables from Canadian census data at the dissemination area (DA) level into 2 factor scores that respectively measure material and social deprivation. A DA is the smallest unit of the Canadian census, is socioeconomically homogeneous, and contains approximately 400–700 people. CMCs' mothers' use of mental health services in the 12 months prior to initial admission (with missing data treated as a separate categorical level, "no data available") were extracted from DAD and NACRS.

3.3.4 Statistical Analysis

We used a multivariable mixed-effect hurdle model to measure associations between clinical/socioeconomic factors and each outcome variable. This type of model has previously been used to examine health care utilization and expenditure (Deb et al., 2018) and is useful when an outcome contains disproportionately many zeros, which is a key characteristic of our study data (e.g., 58.2% and 43.1% of CMCs have no hospital days and ED visits, respectively, in the first year).

Each hurdle model consists of 2 submodels. A binary (logistic regression) submodel estimates the distribution of responses of 0. A conditional submodel estimates the distribution of strictly positive response values as a truncated negative binomial distribution (with the canonical log link) and includes random intercepts by patient to account for repeated (i.e., annual) measurements; the conditional submodel for hospital days also models conditional overdispersion. We used a simulation-based approach to examine statistical fit. Data simulated from the model over multiple simulations was compared to the original data with respect to the proportion of zero responses as well as the conditional distribution of each response.

We included time, sex, age group (with categories following Cohen et al., 2012), clinical conditions (following the ICD-based classification scheme noted previously), technology assistance, number of chronic medications (with ≥5 medications corresponding to polypharmacy and an established higher risk of adverse drug effects to CMCs (Feinstein et al., 2023), readmission indicators (conditional submodels only), residence rurality, and mental health service use as categorical predictors. See Table 3.1 for categorical levels and summaries. We included log-transformed initial length of stay (LOS) and factor scores for material and social deprivation as continuous predictors. Log transformation was applied to (nonzero) responses in the conditional submodels to reduce the effect of outliers on the fitted model. Previous studies have demonstrated differences in resource use between age (Gold et al., 2016) and clinical groups (Berry et al., 2017), and have suggested links between chronic medications and overmedicalization (Pordes et al., 2020). We include the other clinical factors as proxies for clinical severity and quality of care (Cohen et al., 2012, Coller et al.,2014). As noted above, previous studies have found associations between resource use and socioeconomic factors, (Garciá-Altés et al., 2018, Spencer et al., 2015) which here we study in conjunction with clinical factors.

We included the following potential interaction effects after an assessment of clinical plausibility: between clinical indicators, between clinical indicators and time, between clinical groups and initial LOS, between mental health service use and the deprivation scores, between number of chronic medications and time, and between age and time. We removed the latter 2 interaction effects from the binary submodel for ED visits for computational reasons and did not include any interaction effects in the dispersion model.

We performed all analyses with R (version 3.6.3) (R Core Team, 2023) using glmmTMB (version 1.1.3) (Brooks et al., 2017). We conducted tests of overall covariate significance as type-II Wald chi-square tests with a Benjamini–Hochberg correction for false discovery and comparisons between groups with Holm corrections via multcomp (version 1.4-17) (Hothorn et al., 2008). All formal tests had 2-sided alternative hypotheses and used a .05 significance threshold.

3.3.5 Ethics Statement

Research ethics approval for this study was granted by the University of Alberta Research Ethics Board (Pro00103550_REN2). This study was conducted in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki).

3.4 Results

3.4.1 Cohort Characteristics

We identified 12 621 CMCs, which suggests that in 2013, CMCs diagnosed within the last 4 years accounted for approximately 1.3% of children aged 0–18 in Alberta. Over half of this CMC population were newborns (<1 year old). Of the cohort, 82.3% had 1 complex chronic condition, 13.8% had 2, and 3.9% had 3 or more. Number of chronic medications was most commonly 0 (64.9%), 1 (15.8%), or 2–4 (15.8%). The majority of CMCs lived in metropolitan areas (58.7%) and a minority in rural (18.7%) or remote rural (3.8%) areas. The relatively uniform factor score distributions suggest similarities in socioeconomic conditions relative to the sample used to develop the Pampalon Index (Predy et al., 2008). See Table 3.1 for detailed summary statistics. In the first year after diagnosis, 1.5% of CMCs accounted for 28.0% of all CMC hospital days, while 1.7% of CMCs accounted for 15.7% of ED visits. In total, 809 (6.4%)

CMCs died during initial admission or the subsequent 5-year observation window. CMCs who died were not excluded from the analysis, however post-hoc analysis did not reveal any significant differences in model results or interpretation.

We excluded 1516 CMCs (12.0%) who were missing socioeconomic status measures (1515) or had a sex of "other" (1). The excluded CMCs were comparable to the remaining 11 105 CMCs in the analytic cohort on the basis of sex, age, clinical group, number of chronic medications, initial LOS, and mothers' prior use of mental health services.

Table 3.1. CMC Cohort Characteristics (n = 12 621)

	No. (%)
Sex	
Female	5821 (46.1)
Male	6799 (53.9)
Other	1 (0.0)
Age[a], years	
<1 (newborn)	6465 (51.2)
1–4	1763 (14.0)
5–9	1274 (10.1)
10–13	1086 (8.6)
14–18	2033 (16.1)
Clinical group[a,b]	
Cardiology	3926 (31.1)
Neurology	3606 (28.6)
Malignancy	1715 (13.6)
Congenital/genetic	1524 (12.1)
Respiratory	1277 (10.1)
Renal	1142 (9.0)
Gastrointestinal	723 (5.7)
Metabolic	681 (5.4)
Hematology/immunodeficiency	335 (2.7)
Technology assistance	530 (4.2)
Number of chronic medications one year after initial admission	
0	8186 (64.9)
1	1990 (15.8)

2–4	1985 (15.7)
≥5	460 (3.6)
Initial LOS, days	
1–10	10 003 (79.3)
11–30	1510 (12.0)
31–50	499 (4.0)
51–75	291 (2.3)
≥76	318 (2.5)
Residence rurality[a,c]	
Metropolitan	7409 (58.7)
Urban	1358 (10.8)
Rural	2360 (18.7)
Remote rural	479 (3.8)
Missing	1015 (8.0)
Material deprivation quintile[a,d]	
1 (least deprived)	1699 (13.5)
2	1982 (15.7)
3	2072 (16.4)
4	2347 (18.6)
5 (most deprived)	3006 (23.8)
Missing	1515 (12.0)
Social deprivation quintile[a,d,]	
1 (least deprived)	1857 (14.7)
2	2002 (15.9)
3	2311 (18.3)
4	2493 (19.8)
5 (most deprived)	2443 (19.4)
Missing	1515 (12.0)
Mother use of MHS[e]	
Accessed MHS	2157 (17.1)
Did not access MHS	6243 (49.5)
Data unavailable	4221 (33.4)

Abbreviations: CMC, child with medical complexity; LOS, length of stay; MHS, mental health services.

[a] At initial admission.

[b] A CMC may fall into more than 1 clinical group, so percentages need not add to 100%.

[c] From the MyChild dataset, “moderate metropolitan influence” is merged into “metropolitan”, “moderate urban influence” is merged into “urban”, and “rural centre area” into “rural”.

[d] Percentages do not add to 100% due to rounding.

[e] In the 12 months prior to a CMC's initial admission.

3.4.2 Age at Initial Admission

Age was a significant predictor in the conditional submodels for hospital days and ED visits ($\chi_4^2 = 18.3$, $P < .004$ for hospital days; $\chi_4^2 = 84.0$, $P < .001$ for ED visits). Of particular note in the first year, newborns had significantly more conditional hospital days on average relative to the 1–4, 5–9, and 14–18 age groups (relative difference (RD) 1.375–1.913, standard error (SE) 0.129–0.163, $P \leq .04$) and significantly more conditional ED visits relative to the 5–9, 10–13, and 14–18 groups (RD 1.318–1.603, SE 0.090–0.125, $P < .001$). In the fifth year, no significant differences in cumulative number of hospital days were present between the age groups; patterns of differences in cumulative number of ED visits were comparable between the first and fifth years. For all comparisons, see Table 3.2.

Age was also a significant predictor in the binary submodels ($\chi_4^2 = 137.0$, $P < .001$ for hospitals days; $\chi_4^2 = 51.9$, $P < .001$ for ED visits), which suggested analogous reductions in the odds of having zero cumulative hospital days or zero cumulative ED visits over time.

Table 3.2. Comparisons of Conditional Number of Hospital Days and Emergency Department Visits Between Age Groups*

	Year 1			**Year 5**		
	Estimate (SE)	**Relative difference (SE)[a]**	***P* value**	**Estimate (SE)**	**Relative difference (SE)[a]**	***P* value**
Hospital days						
<1 vs						
1–4	0.516 (0.077)	1.676 (0.129)	<.001	0.138 (0.073)	1.148 (0.084)	.41
5–9	0.649 (0.101)	1.913 (0.194)	<.001	0.151 (0.093)	1.163 (0.108)	.52

	Year 1			Year 5		
	Estimate (SE)	**Relative difference (SE)[a]**	***P* value**	**Estimate (SE)**	**Relative difference (SE)[a]**	***P* value**
10–13	0.187 (0.128)	1.205 (0.155)	.44	−0.126 (0.122)	0.882 (0.108)	>.99
14–18	0.319 (0.118)	1.375 (0.163)	.04	−0.068 (0.112)	0.934 (0.104)	>.99
1–4 vs						
5–9	0.133 (0.103)	1.142 (0.118)	.44	0.013 (0.094)	1.014 (0.095)	>.99
10–13	−0.330 (0.127)	0.719 (0.080)	.05	−0.263 (0.120)	0.768 (0.092)	.24
14–18	−0.198 (0.117)	0.821 (0.096)	.37	−0.206 (0.109)	0.814 (0.089)	.41
5–9 vs						
10–13	−0.462 (0.122)	0.630 (0.077)	.001	−0.277 (0.110)	0.758 (0.083)	.12
14–18	−0.330 (0.112)	0.719 (0.091)	.02	−0.219 (0.099)	0.803 (0.079)	.24
10–13 vs 14–18	0.132 (0.106)	1.141 (0.121)	.44	0.058 (0.093)	1.060 (0.098)	>.99
ED visits						
<1 vs						
1–4	0.074 (0.046)	1.077 (0.049)	.21	0.015 (0.040)	1.015 (0.040)	>.99
5–9	0.471 (0.060)	1.602 (0.097)	<.001	0.318 (0.053)	1.374 (0.073)	<.001
10–13	0.472 (0.078)	1.603 (0.125)	<.001	0.239 (0.071)	1.270 (0.090)	.004
14–18	0.276 (0.068)	1.318 (0.090)	<.001	−0.008 (0.064)	0.992 (0.064)	>.99
1–4 vs						
5–9	0.397 (0.063)	1.488 (0.093)	<.001	0.302 (0.054)	1.353 (0.073)	<.001
10–13	0.398 (0.078)	1.489 (0.117)	<.001	0.224 (0.070)	1.251 (0.087)	.007
14–18	0.202 (0.069)	1.224 (0.084)	.01	−0.023 (0.064)	0.978 (0.062)	>.99
5–9 vs						
10–13	0.001 (0.075)	1.001 (0.075)	>.99	−0.078 (0.063)	0.925 (0.059)	.87

	Year 1			Year 5		
	Estimate (SE)	**Relative difference (SE)[a]**	***P* value**	**Estimate (SE)**	**Relative difference (SE)[a]**	***P* value**
14–18	−0.195 (0.065)	0.823 (0.054)	.01	−0.325 (0.057)	0.722 (0.041)	<.001
10–13 vs 14–18	−0.196 (0.063)	0.822 (0.052)	.01	−0.247 (0.052)	0.781 (0.041)	<.001

Abbreviations: ED, emergency department; SE, standard error.

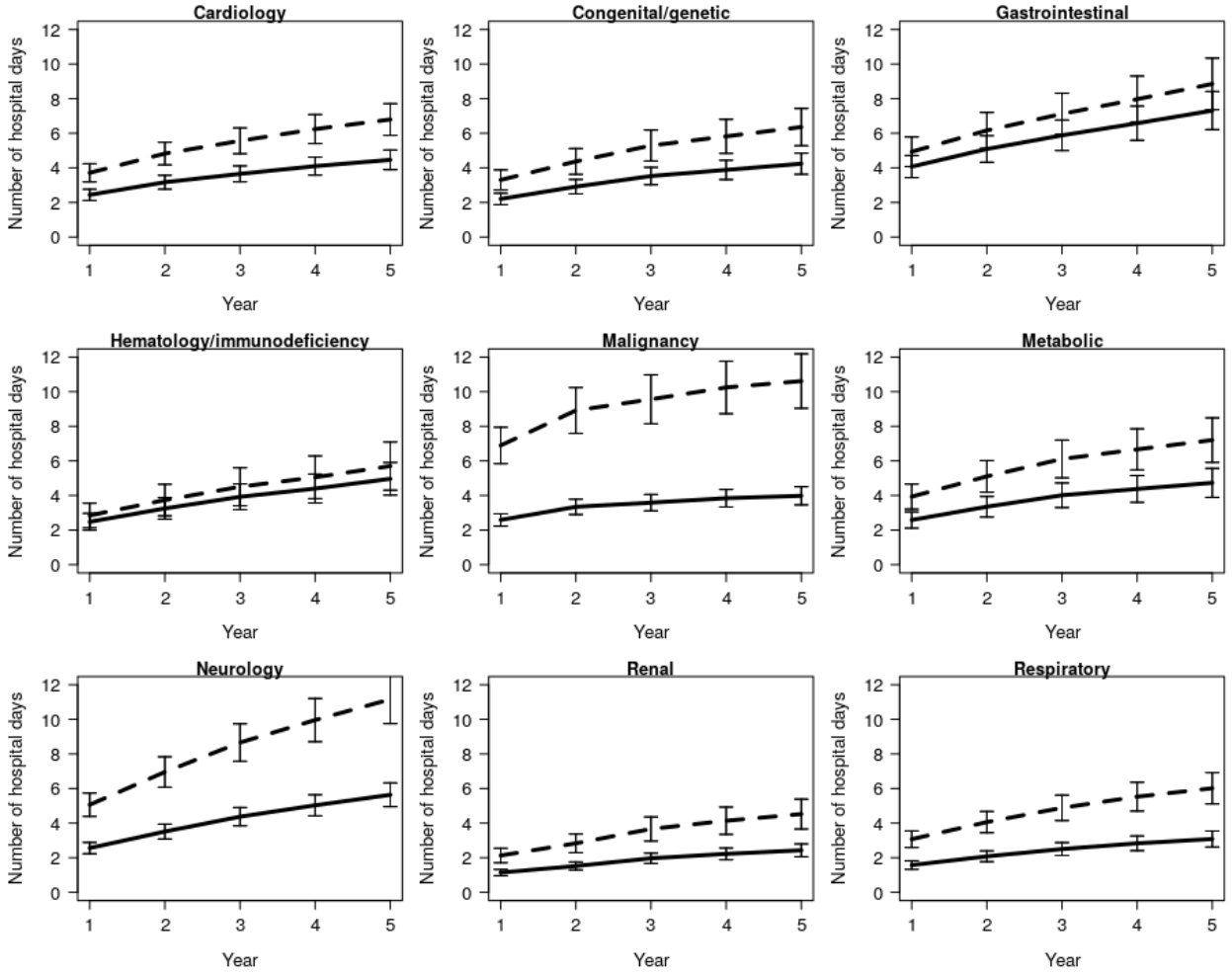
a Relative difference is the natural antilog of the estimate.

*Comparisons between Age groups are made while all other variables are held constant.

3.4.3 Clinical Conditions

Figure 3.1 illustrates differences in the conditional number of hospital days between patients in exactly 1 clinical group with an initial LOS of 3 and 12 days (approximately the median and 80th percentile of initial LOS in our sample). CMCs in the neurology category appeared to have the steepest increase in conditional hospital days over the 5 years. Differences between the groups in terms of cumulative number of hospital days were more apparent for larger values of initial LOS, as described in the following subsection. Overall, ED visit trajectories (Figure 3.3, Appendix B) showed less variation across the clinical groups and with initial LOS.

Figure 3.1. Conditional Hospital Day Trajectories by Clinical Group



Legend: Solid and dotted lines indicate estimates for an initial length of stay of 3 and 12 days, respectively. Error bars denote standard errors. Estimates correspond to a 14–19-year-old, male patient living in a metropolitan area with a 3-day initial hospitalization, no technology assistance, no chronic medications, and no readmissions within 30 days; deprivation factor scores are set to 0 and the patient's mother is assumed to have not used mental health services.

Of particular note, for a 3-day initial LOS, the neurology group had significantly more or a comparable number of hospital days (RD 0.734–1.549; SE 0.086–0.232; $P \geq .06$ relative to gastrointestinal, malignancy, metabolic, and year 1 congenital/genetic; RD 1.363–2.468, SE 0.130–0.395; $P \leq .002$ otherwise) and ED visits (RD 0.887–1.549, SE 0.072–0.435, $P \geq .07$ relative to gastrointestinal, hematology/immunodeficiency, malignancy, metabolic, year 1 renal,

and year 1 congenital/genetic; RD 1.272–2.468, SE 0.073–395, P ≤ .01 otherwise) in years 1 and 5 relative to other groups.

3.4.4 Initial Length of Stay

Initial LOS was a significant predictor in both conditional submodels ($\chi_1^2 = 464.2$, $P < .001$ for hospital days; $\chi_1^2 = 14.7$, $P = .001$ for ED visits). Table 3.3 presents estimates of average changes in the conditional response associated with a 10-fold increase in initial LOS, which was significant for all (RD 1.960–5.097, SE 0.161–0.610, $P < .001$) but the gastrointestinal and hematology/immunodeficiency groups in the hospital days model and significant only for the gastrointestinal, hematology/immunodeficiency, and malignancy groups in the ED visit model (RD 0.663–1.364, SE 0.086–0.138, $P \leq .05$).

Initial LOS was also significant in the binary submodels ($\chi 12 = 95.0$, $P < .001$ for hospital days; $\chi 12 = 159.0$, $P < .001$ for ED visits), where these associations were nonsignificant, negative (OR 0.520–0.897, SE 0.031–0.084, P ≤ .007 for cardiology, congenital/genetic, malignancy, neurology, renal, and respiratory) in all groups except the congenital/genetic group in the hospital days model (OR 1.308, SE 0.084, P < .001) (Table 3.5 Appendix B).

Table 3.3. Association Between Initial Length of Stay and Conditional Mean Number of Hospital Days and ED Visits*

Clinical category[a]	**Estimate (SE)**	**Relative difference (SE)**[b]	***P* value**
Hospital days			
Cardiology	0.698 (0.080)	2.009 (0.161)	<.001
Congenital/genetic	0.673 (0.138)	1.960 (0.271)	<.001

Gastrointestinal	0.318 (0.167)	1.374 (0.230)	.11
Hematology/immunodeficiency	0.229 (0.256)	1.258 (0.322)	.37
Malignancy	1.629 (0.120)	5.097 (0.610)	<.001
Metabolic	0.698 (0.169)	2.010 (0.339)	<.001
Neurology	1.134 (0.076)	3.109 (0.235)	<.001
Renal	1.034 (0.172)	2.811 (0.484)	<.001
Respiratory	1.111 (0.112)	3.039 (0.340)	<.001
ED visits			
Cardiology	0.048 (0.045)	1.049 (0.047)	>.99
Congenital/genetic	0.063 (0.081)	1.065 (0.087)	>.99
Gastrointestinal	0.311 (0.101)	1.364 (0.138)	.02
Hematology/immunodeficiency	−0.410 (0.153)	0.663 (0.102)	.05
Malignancy	0.241 (0.067)	1.273 (0.086)	.003
Metabolic	0.080 (0.096)	1.083 (0.104)	>.99
Neurology	0.104 (0.045)	1.110 (0.050)	.12
Renal	0.096 (0.091)	1.100 (0.100)	>.99
Respiratory	0.112 (0.064)	1.118 (0.072)	.41

Abbreviations: ED, emergency department; SE, standard error.

Change in initial length of stay: All estimates correspond to a 10-fold increase in initial length of stay.

*Comparisons between Age groups are made while all other variables are held constant

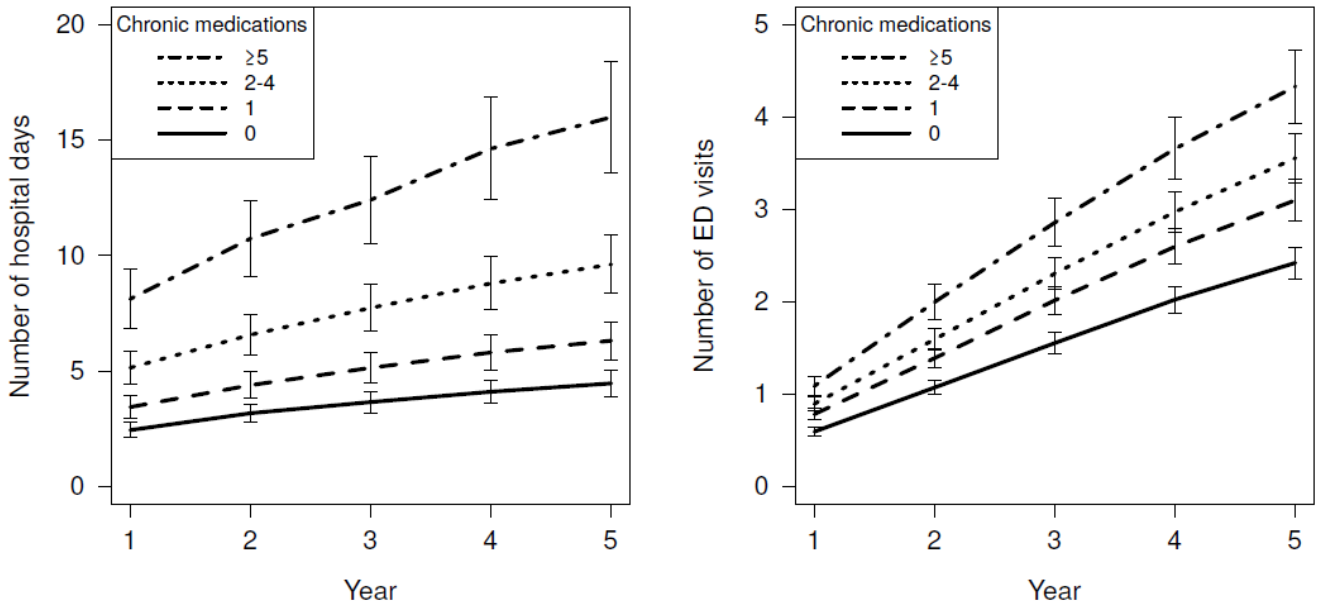
[a] Describes a CMC in exactly 1 clinical category.

[b] Relative difference is the natural antilog of the estimate.

3.4.5 Chronic Medications

Number of chronic medications was a significant predictor in both conditional submodels ($\chi_3^2 = 251.0$, $P < .001$ for hospital days; $\chi_3^2 = 193.1$, $P < .001$ for ED visits). Both conditional responses differed significantly between every pair of chronic medication groups. These differences were comparable between years 1 and 5. CMCs with more chronic medications had more conditional hospital days on average (between adjacent groups, RD 1.409–1.580, SE 0.089–0.170, $P < .001$ in year 1 and RD 1.415–1.663, SE 0.087–0.172, $P < .001$ in year 5); the results for ED visits were similar (between adjacent groups, RD 1.320–1.133, SE 0.053–0.091, $P \leq .009$ in year 1 and RD 1.147–1.280, SE 0.044–0.082, $P \leq .003$ in year 5). Figure 3.2 illustrates this positive association.

Figure 3.2. Conditional Hospital Day (left) and Conditional ED visits (Right) Trajectories by Number of Chronic Medications



Number of chronic medications was also a significant predictor in the binary submodels ($\chi_3^2 = 1690.8$, $P < .001$ for hospital days; $\chi_3^2 = 838.2$, $P < .001$ for ED visits), which yielded similar interpretations regarding the absence of hospital days (between adjacent groups,

OR 0.322–0.691, SE 0.032–0.069, $P < .001$) or ED visits (OR 0.563–0.811, SE 0.018–0.053, $P < .001$).

3.4.6 Residence Rurality

In the conditional submodels, residence rurality was significantly associated with cumulative ED visits ($\chi_3^2 = 583.5$, $P < .001$), but not with cumulative hospital days ($\chi_3^2 = 8.5$, $P = .09$). CMCs living in metropolitan areas had the fewest conditional number of ED visits, followed by those in urban (RD 1.087 vs metropolitan, SE 0.041, $P = .03$), rural (RD 1.727 vs urban, SE 0.075, $P < .001$), and remote rural (RD 1.114 vs rural, SE 0.122, $P < .001$).

Rurality was significant in both binary submodels ($\chi_3^2 = 316.2$, $P < .001$ for hospital days; $\chi_3^2 = 660.4$, $P < .001$ for ED visits), which suggested similar differences between ruralities in terms of the odds of having no hospital days (OR 1.342, SE 0.039, $P < .001$ for metropolitan vs urban; OR 1.047, SE 0.036, $P = .17$ for urban vs rural; OR 1.149, SE 0.056, $P = .009$ for rural vs remote rural) and ED visits (OR 1.135, SE 0.037, $P < .001$ for metropolitan vs urban; OR 1.758, SE 0.071, $P < .001$ for urban vs rural; OR 1.207, SE 0.079, $P = .004$ for rural vs remote rural).

3.4.7 Material and Social Deprivation

Material ($\chi_1^2 = 13.1$, $P = .001$ for hospital days; $\chi_1^2 = 12.3$, $P = .003$ for ED visits) and social ($\chi_1^2 = 8.6$, $P = .001$ for hospital days; $\chi_1^2 = 43.3$, $P < .001$ for ED visits) deprivation were both significant predictors in the conditional submodels. For CMCs with hospital days, material deprivation only among those with mothers who did not use mental health services had a significant association with hospital days (RD 1.099 for a 1-standard-deviation (SD) increase, SE 0.036, $P = .02$). On the other hand, both material (RD 1.057 for a 1-SD increase, SE 0.018, $P = .005$) and social (RD 1.073 for a 1-SD increase, SE 0.018, $P < .001$) deprivation scores had

significant, positive associations with conditional ED visits among CMCs whose mothers did not use mental health services. Among CMCs with hospital days and whose mothers did use mental health services, only social deprivation had a significant association with hospital days (RD 1.076 for a 1-SD increase, SE 0.030, $P = .019$).

In the binary submodels, material deprivation ($\chi_1^2 = 35.4$, $P < .001$) but not social deprivation ($\chi_1^2 = 1.4$, $P = .30$) were associated with hospital days, while both material ($\chi_1^2 = 26.8$, $P < .001$) and social ($\chi_1^2 = 68.3$, $P < .001$) deprivation were associated with ED visits. Associated with the absence of hospitals days were material deprivation among CMCs whose mothers used (OR 0.917 for a 1-SD increase, SE 0.018, $P < .001$) or did not use (OR 0.964 for a 1-SD increase; SE 0.013, $P = .02$) mental health services, and social deprivation only among mothers who did not use mental health services (OR 0.965, SE 0.013, $P = .02$). Associated with the absence of ED visits were material (OR 0.935, SE 0.016, $P = .02$) and social (OR 0.949, SE 0.014, $P < .001$) deprivation, but only among mothers who did not use mental health services.

3.5 Discussion

A key finding of this study is that clinical factors are more strongly associated with hospital utilization while socioeconomic factors are more strongly associated with ED visits.

3.5.1 Hospital Utilization

Although high health care needs are characteristic of CMCs (Berry et al., 2015), resource imbalances among our population suggest that certain subpopulations require more resources and support. This conclusion mirrors that of Gold et al., 2016 who identified a small group of CMCs with long initial hospitalizations that accounted for the majority of CMC hospital days and costs

across 44 U.S. children's hospitals. Targeting CMC subpopulations with high resource use may thus be more effective than targeting all CMCs in reducing hospital use.

In our cohort, CMCs with a longer initial hospitalization tended to have more hospital days relative to other clinical groups (consistent with Gold et al., 2016). We hypothesize that the varying associations with initial LOS and other differences between clinical groups are attributable to clinical severity and the availability of support programs for patients and caregivers. That home nursing care reduces hospital use in CMCs (Gay et al., 2016) supports this hypothesis.

The positive association between initial LOS and both the odds of having hospital days and the conditional number of hospital days is arguably intuitive: longer initial hospitalization indicates greater need for resources and clinical complexity not resolved by initial care (Gold et al., 2016, Berry et al., 2014). For the 1 exception to this trend in the congenital/genetic clinical group, a larger initial LOS may indicate that longer initial hospitalization has a positive long-term impact on the health of the child and that further hospitalization is not needed (e.g., congenital heart defects that receive early surgical treatment and remain well managed). Our findings agree with other studies in that certain CMC subpopulations require more health care resources and may benefit from increased care coordination and other types of caregiver support programs (Berry et al., 2014, Casey et al., 2011, Simon et al., 2017).

While the association between number of chronic medications and hospital days is plausibly related to clinical acuity, other factors may drive this relationship. Pordes et al., 2020 (pe478) found the CMC population to be at risk of overmedicalization ("the delivery of unnecessary and potentially harmful medical care"), which can propagate recurring hospitalizations (Kelly et al., 2000). Thus, chronic medications may be a sign of complex needs but may also indicate

overmedicalization. Further research is needed to examine specific medications prescribed for this population and identify whether overmedicalization is a potential cause of preventable hospitalizations in CMCs.

3.5.2 Emergency Department Visits

Residence rurality at initial admission is associated with number of ED visits: in our cohort, those living in remote rural areas had the highest number of ED visits, followed by those in rural, urban, and metropolitan areas. Possibly contributing to this pattern in Albertan contexts is the fact that many primary care physicians also work in local EDs. It is feasible that families and caregivers utilize EDs if appointments are not readily available or if access to urgent care is otherwise limited. More generally, the limited availability (Nageswaran et al., 2017) of quality home care, telephone access to knowledge providers, and next-day appointments (Pulcini et al., 2021) may contribute to higher ED utilization for rural CMCs.

Material and social deprivation have significant, positive associations with ED visits for CMCs whose mothers used mental health services prior to initial admission (as does social deprivation among CMCs whose mothers did not use these services). Since each measure rolls up multiple metrics, we cannot comment on the mechanism underlying this association. Nonetheless, social and material deprivation are important factors that must be accounted for when developing policy and planning health care programs. Our findings support the claim that improved social supports for families such as funding for care coordination and caregiver education programs may reduce ED visits and improve outcomes for CMCs.

The association between material/social deprivation, mental health services, and number of ED visits is an important finding. It is possible that the mothers in our study who accessed mental health support prior to their child's initial admission were better equipped to manage their

child's illness. However, we cannot determine the exact nature of this relationship since the type and duration of the supports accessed as well as access to mental health services outside the hospital or ED facilities are not available in our data. Further investigation of this relationship is needed to better understand the role of maternal mental health in resource use by CMCs.

3.5.3 Limitations

Only the outcome variables in our study were available over the 5-year observation window, so an analysis with time-varying predictors could yield more-interpretable results. Separately, studies with more-specific designs can further tease apart the associations and interactions observed in this work.

We suspect that missing socioeconomic measures stem largely from Statistics Canada withholding data for sparsely populated DAs; such DAs in our study are likely (remote) rural and may cause our analysis to underestimate (the significance of) comparisons with these ruralities and associations with socioeconomic status. While DAs are homogeneous by design, their use introduces the potential for underestimated effects and associations (Wood et al., 2014). Furthermore, our data does not capture CMCs receiving resources outside the scope of the provincial health care system (e.g., patients leaving the province, supplemental caregiver support), but we have no evidence that this occurs for nonignorable reasons. Another consideration for interpreting socioeconomic covariates such as rurality is that the availability of primary care services was not available in the dataset and thus may be a confounding factor.

Other limitations common to retrospective research with administrative data stem from the socioeconomic measures used and provided by the Pampalon Index. These measures are at the DA level and can thus lose important, individual-level variability beyond that lost by factor

analysis. Future studies may consider individual factors such as ethnicity or social supports, which are unmeasured in our data outside of social deprivation as a general proxy.

3.6 Conclusion

Our finding that clinical and socioeconomic factors play different roles in hospitalizations and ED visits suggests that policies and programs should account for socioeconomic factors such as rurality and socioeconomic status. These findings can feasibly be generalized to other publicly funded health care systems such as those within Canada and Europe and possibly to Medicaid-funded hospitals in the U.S. However, more research is needed to unravel the impacts of specific socioeconomic factors on families and caregivers so that health care systems can identify specific policies and interventions to improve CMC care.

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Chapter 4. Clinical and socioeconomic predictors of hospital use and ED visits among children with medical complexity in Alberta: A machine learning approach using administrative data.

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This paper was ready for submission at time of writing.

4.1 Abstract

Objectives: The primary objective of this study was to identify clinical and socioeconomic predictors of hospital and ED use among children with medical complexity within 1 and 5 years of an initial discharge. A secondary objective was to estimate marginal associations between important predictors and resource use.

Methods: This retrospective, population-cohort study of children with medical complexity in Alberta linked administrative health data with Canadian census data and used tree-based, gradient-boosted regression models to identify clinical and socioeconomic predictors of resource use. Each analysis models, at the patient level, the probability of (any) resource use and, when present, amount of resource use.

Results: The best short- and long-term predictors of having a hospital stay and number of hospital days were initial length of stay and clinical classification (single vs multiple complex chronic conditions). Initial length of stay, closely followed by residence rurality and other socioeconomic factors, were top predictors of short-term ED use. The top predictors of ED use in

the long term were almost exclusively socioeconomic in nature, with rurality a top predictor of number of ED visits (among children with ED visits). Estimates of marginal associations between initial length of stay and resource use showed that average number of hospital days increases as initial length of stay increases up to approximately 100 days. Children with medical complexity living in (remote) rural areas had more ED visits on average than those living in urban or metropolitan areas.

Conclusions: Clinical factors are generally better predictors of hospital use whereas socioeconomic factors are more predictive of ED use among children with medical complexity in Alberta. The results confirm existing literature on the importance of socioeconomic factors with respect to health care use by children with medical complexity.

4.2 Introduction

Children with medical complexity (CMCs) are characterized as having at least 1 complex chronic condition (CCC) that requires significant health care support or medical technology for daily living (Berry et al., 2015). Although relatively few in number (about 1% of children), studies from the United States, Australia, and Canada have reported that CMCs account for a substantial amount of pediatric hospitalizations (Srivastava et al., 2016; Berry et al., 2017; Canadian Institute for Health Information [CIHI], 2020). Specifically in Canada, CIHI (2020) reported that CMCs accounted for 57% of hospital care costs in 2015–2016 among children and youth. CMCs are also twice as likely to access the emergency department (ED) than non-CMC children, more likely to return to the ED after being discharged, and more likely to be admitted (CIHI, 2020).

A robust understanding of health care utilization trends, notably of hospital and ED use (Sidra et al., 2022), among CMCs over time supports the development of more-effective policies, the optimization of care delivery, and the enhancement of caregiver support through the development of models of care delivery (Pordes et al., 2018). In a study to establish research priorities for children and youth with special health care needs, Coller et al. (2020) identified several important factors, including social determinants of health, family support programs, and clinical care delivery. All these factors interact in complex ways over time, which can be challenging to model. Longitudinal studies are particularly important for the CMC population as health care needs and access to health care services change over time (Berry et al., 2013).

Much of the research on CMCs to date has focused on understanding associations between clinical factors and health care resources and consider only a limited number of socioeconomic factors. Socioeconomic factors play an important role in the health of children and are even more important for CMCs (Halfon et al., 2010). In general, the use of health care services such as hospitals and EDs depends on many factors that include the socioeconomic status of families such as location of residence and the availability of financial and community support (Andersen, 1995). However, there is a lack of administrative health data linked with socioeconomic data within Electronic Health Records (EHRs) to encourage and facilitate research in this area (Cadarette & Wong, 2015).

Both inferential and predictive models can be used to describe associations between clinical and socioeconomic factors and health care use over time. However, health care resource use for CMCs has historically only been described in terms of population-level trends, typically under traditional inferential regression frameworks (King & Strumpf, 2022) that are limited in terms of their complexity and predictive capacity. Predictive models, such as those common in

machine learning, are better able to handle complex interaction structures and report on the predictive importance of large numbers of variables (King et al., 2022). The lack of predictive modelling in CMC research represents a significant gap in the literature.

Despite its increasing accessibility, modern machine learning methods have seen slow adoption in CMC-related research. Applications of machine learning in the health care literature more generally have typically been limited to the diagnosis, screening, and prediction of outcomes for a small number of specific diseases such as cancers, nervous system diseases, or cardiovascular diseases (Jiang et al., 2017). As CMCs are a highly heterogeneous population with diverse clinical and socioeconomic characteristics, it is critical to accommodate the many interaction effects relevant to this group (Carrilero et al., 2020). Machine learning models in the study of CMCs thus have the potential to yield valuable insights and drive further research.

However, predictive modelling for CMCs has arguably been slowed by the availability and composition of administrative health care data. In this sense, the design of EHRs has the potential to accelerate or bottleneck research in this field (Casey et al., 2016). The marriage of EHRs and predictive modelling is an emerging area of innovation in health care that can improve patient outcomes and decision-making processes (Xu et al., 2018; Lee et al., 2020). Developments in this area are thus also broadly relevant to jurisdictions developing EHRs (e.g., Connect Care in Alberta) in addition to CMC-related research specifically.

The purpose of this study was to identify and compare important predictors of short- and long-term health care resource use (ie, hospital days and ED visits) at the individual level among CMCs in Alberta, Canada. A secondary objective was to estimate marginal associations for the most important predictors for further insight. Our results motivate, as another secondary

objective, further discussion of the utility of administrative data in CMC-related research and how EHRs can be designed to facilitate predictive analyses in the future for CMC populations.

4.3 Methods

This retrospective, population-cohort study links administrative health data to socioeconomic data from the Canadian census to identify clinical and socioeconomic predictors of hospital and ED visits for CMCs in Alberta. The University of Alberta Research Ethics Board (Pro00103550_REN2) granted research ethics approval for this study.

4.3.1 Study Design and Data

The Maternal Newborn Child Youth Strategic Clinical Network under Alberta Health Services (AHS) extracted data for this study from health administrative datasets housed in the AHS data repository (AHSDR). The AHSDR houses the Discharge Abstract Database (DAD), which contains hospital discharge data (CIHI, 2023a); the National Ambulatory Care Reporting System (NACRS) (CIHI, 2023b), which contains data on hospital ED visits; Alberta Vital Statistics (Government of Alberta, 2017), which lists all births and deaths in Alberta; and the AHS Pharmaceutical Information Network (PIN) (Government of Alberta, 2012), which contains data on pharmaceuticals dispensed in a community setting.

4.3.2 Study Population and Primary Outcome Variables

CMCs were identified using DAD via an ICD-10-based definition commonly used in the literature (Cohen et al., 2012). CMCs were included if they were Alberta residents, were at most 18 years old at admission, and had an initial discharge and diagnosis between 2010 and 2013.

The primary outcome variables were annual cumulative number of hospital days and ED visits (at the patient level) for the first and fifth years immediately following an initial hospital discharge (first admission and discharge with a Complex Chronic Condition diagnosis). These outcomes were obtained from DAD and NACRS.

4.3.3 Study Variables

Demographic data included patients' age and sex. Clinical information included initial hospitalization length of stay (LOS) in days, number of chronic medications 1 year prior to initial admission (from PIN data), clinical classification (ie, single vs multiple complex chronic conditions), 9 indicators for chronic disease involving different body systems, the presence of technology assistance (e.g., home oxygen), and 7-day and 30-day readmission following initial discharge. The presence of technology assistance utilized procedure codes established by Cohen et al., (2012). All variables were measured at initial admission unless otherwise specified.

Socioeconomic variables were obtained through residential postal codes collected at initial discharge and included AHS zone, residence rurality, material and social deprivation scores, Canadian census data, and (from DAD and NACRS) mother use of mental health services in the 12 months prior to their child's initial admission. AHS has 5 zones (North, Edmonton, Central, Calgary, and South) and classifies regions of Alberta into 6 rurality categories ranging from metropolitan to remote rural. AHS zones are geographic areas with operational accountability to deliver health care services within an integrated provincial health system. Material and social deprivation were measured through the Pampalon index based on 6 variables from Canadian census data measured at the dissemination area (DA) level (Predy et al., 2008). These 6 component variables, which include average household income as well as proportions of single-

parent families; individuals who are divorced, widowed, or separated; individuals living alone; unemployed individuals; and individuals without a high school diploma were also included.

4.3.4 Analysis

We used tree-based, gradient-boosted regression models to predict each of the 2 primary outcomes (cumulative hospital days and cumulative ED visits) after 1 and 5 years following initial discharge using clinical and socioeconomic factors. Gradient boosting with regression trees is well established in the statistical literature (Friedman, 2001) and is conceptually similar to random forests: both frameworks obtain a sequence of decision trees that are used in tandem to predict a response. Unlike random forests, which grow trees independently, gradient boosting models grow trees that successively improve upon the previous trees. For a gentle introduction to tree-based methods, see King & Strumpf (2022).

Each of the 4 models contained 2 submodels: the first predicted the presence of hospital days or ED visits (in a binary model), while the second predicted number of hospital days among CMCs with nonzero resource use (in a conditional model). Together, these 2 submodels fit into a more-general hurdle model framework. Linear hurdle regression models have previously been used in studies of health care resource use and expenditure (Deb & Norton, 2018), also specifically for CMCs (Sidra et al., 2023). We identified important predictors of hospital days and ED visits in the first 1 and 5 years following an initial admission through the relative importance of each variable. Intuitively, this summary measure describes the proportion of variance explained by each predictor included in the model (out the total amount of variance explained by the model) (Friedman, 2001).

We included as predictors time, sex, age group, clinical conditions (using age category and ICD-codes based on Cohen et al. [2012] and Feudtner et al. [2000]), technology assistance, number of chronic medications, readmission indicators, residence rurality, mother mental health service use (with missing data treated as a separate categorical level, "no data available"), initial LOS, material and social deprivation scores derived from the Pampalon index, and the 6 DA-level measures described in the previous section. The clinical factors included are proxies for clinical complexity, while socioeconomic factors are generally known to be associated with health resource use and are important to consider together with clinical factors (García-Altés et al., 2018).

All analyses were performed with R (version 3.6.3) (R Core Team, 2020) using gbm (package version 2.1.8.1) (Greenwell et al., 2022). We separately tuned the hyperparameters (namely, number of trees, tree height, shrinkage, and terminal node size) of each model via caret (version 6.0-93) (Kuhn, 2022) by iteratively refining a coarse hyperparameter grid to maximize prediction accuracy (of the binary submodels) or to minimize root mean-squared error (of the conditional submodels). We applied a log transformation to both responses in the conditional submodels to reduce the effect of outliers.

We used a training set of 90% of the cohort's CMCs to train the models and estimated generalization error using a hold-out testing set of the remaining 10%. We summarized binary and conditional model performance on both sets with AUC (ie, area under the ROC curve) and R^2 (ie, proportion of response variability explained), respectively. AUC is interpretable as the probability that a model will be able to correctly differentiate between a randomly selected (case/non-case) pair of subjects. In a secondary analysis, we presented estimates of marginal associations for select important variables. These estimates were calculated by integrating out all

other variables, as described in Friedman (2001). All confidence intervals were obtained using a simple nonparametric bootstrap (with 100 bootstrap samples). To assess model fit, we used calibration curves for the binary model (to assess predicted probabilities of a non-zero response) and for the conditional model we examine plots of true vs. predicted values.

4.4 Results

4.4.1 Cohort Characteristics

In total, 12 621 CMCs were included in the sample. Over half of this CMC population were newborns (<1 year old) at initial admission. Of the cohort, 82.3% had 1 CCC, 13.8% had 2, and 3.9% had 3 or more. Number of chronic medications was most commonly 0 (79.5%), 1 (9.4%), or 2–4 (8.9%). Most CMCs lived in metropolitan areas (58.7%) and a minority lived in rural (18.7%) or remote rural (3.8%) areas. See Table 4.1 for detailed cohort characteristics.

We excluded 1516 CMCs (12.0%) who were missing socioeconomic data (1515) or had a sex of "other" (1). The excluded CMCs were comparable to the remaining 11 105 CMCs in the analytic cohort except on the basis of 5 of the DA-level measures (Table 4.1).

Table 4.1. CMC Cohort Characteristics (n = 12 621)

Characteristic	**Analytic sample, no. (%) (n = 11 105)**	**Excluded patients, no. (%) (n = 1516)**	***P* value[a]**
Female[b]	5105 (48.6)	716 (47.2)	.36
Age[c], years			
Median (Q_1, Q_3)	0.0 (0.0, 9.0)	0.0 (0.0, 8.0)	.71
Newborns (<1)	5699 (51.3)	766 (50.5)	.58
Clinical group[c,d]			
Cardiology	3438 (31.0)	488 (32.2)	.35
Neurology	3165 (28.5)	441 (29.1)	.66

Malignancy	1531 (13.8)	184 (12.1)	.09
Congenital/genetic	1347 (12.1)	177 (11.7)	.64
Respiratory	1125 (10.1)	152 (10.0)	.94
Renal	1016 (9.1)	126 (8.3)	.31
Gastrointestinal	630 (5.7)	93 (8.3)	.51
Metabolic	589 (5.3)	92 (6.1)	.24
Hematology/immunodeficiency	291 (2.6)	44 (2.9)	.58
Technology assistance	461 (4.2)	69 (4.6)	.51
Number of chronic medications[e]			.14
0	8844 (79.6)	1187 (78.3)	
1	1039 (9.4)	143 (9.4)	
2–4	980 (8.8)	139 (9.2)	
≥5	242 (2.2)	47 (3.1)	
Initial LOS, days, Median (Q_1, Q_3)	3.0 (1.0, 8.0)	3.0 (1.0, 8.0)	.42
Residence rurality[c,f]			<.001
Metropolitan	7172 (64.6)	237 (15.6)	
Urban	1292 (11.6)	66 (4.4)	
Rural	2164 (19.5)	196 (12.9)	
Remote rural	477 (4.3)	2 (0.1)	
Missing[g]	0 (0.0)	1015 (67.0)	
Mother use of MHS[e]			.82
Accessed MHS	1904 (17.1)	253 (16.7)	
Did not access MHS	5482 (49.4)	761 (50.2)	
Data unavailable	3719 (33.5)	502 (33.1)	
DA-level summaries,[h,i] median (Q_1, Q_3)			
Material deprivation	0.01 (-0.02, 0.04)	-	-
Social deprivation	0.00 (-0.02, 0.03)	-	-
Single-parent families	0.14 (0.10, 0.21)	0.15 (0.09, 0.21)	.52
Separated, divorced or widowed[j]	0.17 (0.13, 0.22)	0.21 (0.15, 0.27)	<.001
Living alone[j]	0.07 (0.04, 0.12)	0.11 (0.07, 0.19)	<.001
Income,[i] CAD 1000	44.6 (36.0, 48.2)	40.8 (34.7, 53.6)	<.001
Employed[j]	0.70 (0.62, 0.76)	0.67 (0.55, 0.74)	<.001
No high-school diploma[j]	0.18 (0.12, 0.26)	0.22 (0.13, 0.33)	<.001

Abbreviations: CAD, Canadian dollars; CMC, child with medical complexity; DA, dissemination area; LOS, length of stay; MHS, mental health services.
[a] Comparisons were conducted via Wilcoxon signed-rank tests (for age, initial LOS, and DA-level summaries), 2-sample tests for proportions (for sex, proportion of newborns, and clinical groups), Pearson's chi-squared tests (for number of chronic medications, rurality, and MHS use). *P* values are reported without correction.
[b] All other CMCs were male, with the exception of 1 with a noted sex of "Other".
[c] At initial admission.
[d] A CMC may fall into more than 1 clinical group, so percentages need not add to 100%.
[e] In the 12 months prior to a CMC's initial admission.
[f] From the original dataset, "moderate metropolitan influence", "moderate urban influence", and "rural centre area" were merged into "metropolitan", "urban", and "rural", respectively.
[g] Excluded from the comparison
[h] Summary of DA-level proportions (or averages, for income) across the sample.
[i] Calculated for the excluded sample where data was available. Deprivation scores were available for only 1 CMC in the excluded sample, so summaries and comparisons are omitted here.
[j] Among individuals in a DA at least 15 years of age.

4.4.2 Performance

On the training set and across both periods and primary outcomes, AUC for the binary models was moderate–high between 0.70 and 0.78. On the testing set, AUC was moderate (Hosmer et al., 2000) at about 0.61 for the ED visit models and 0.71 for the hospital day models. Performance was poorer for the conditional submodels, where R^2 was low, falling between 0.10 and 0.29, and was generally lower in year 5. See Appendix C Table C4.1 for exact performance measures.

4.4.3 Predictor Importance

Refer to Figures 4.1 and 4.2 for individual variable importances in the binary and conditional submodels, respectively.

Short-term Predictors

The best predictors of having a hospital stay within the first year were initial LOS and clinical classification, which were at least 2–3 times as important as age (the third most important predictor) and the other socioeconomic variables. Among CMCs with hospital days in the first year, initial LOS was by far the most important predictor of number of hospital days,

with an importance at least 9 times higher than age and clinical classification (the second and third most important predictors) and the socioeconomic variables.

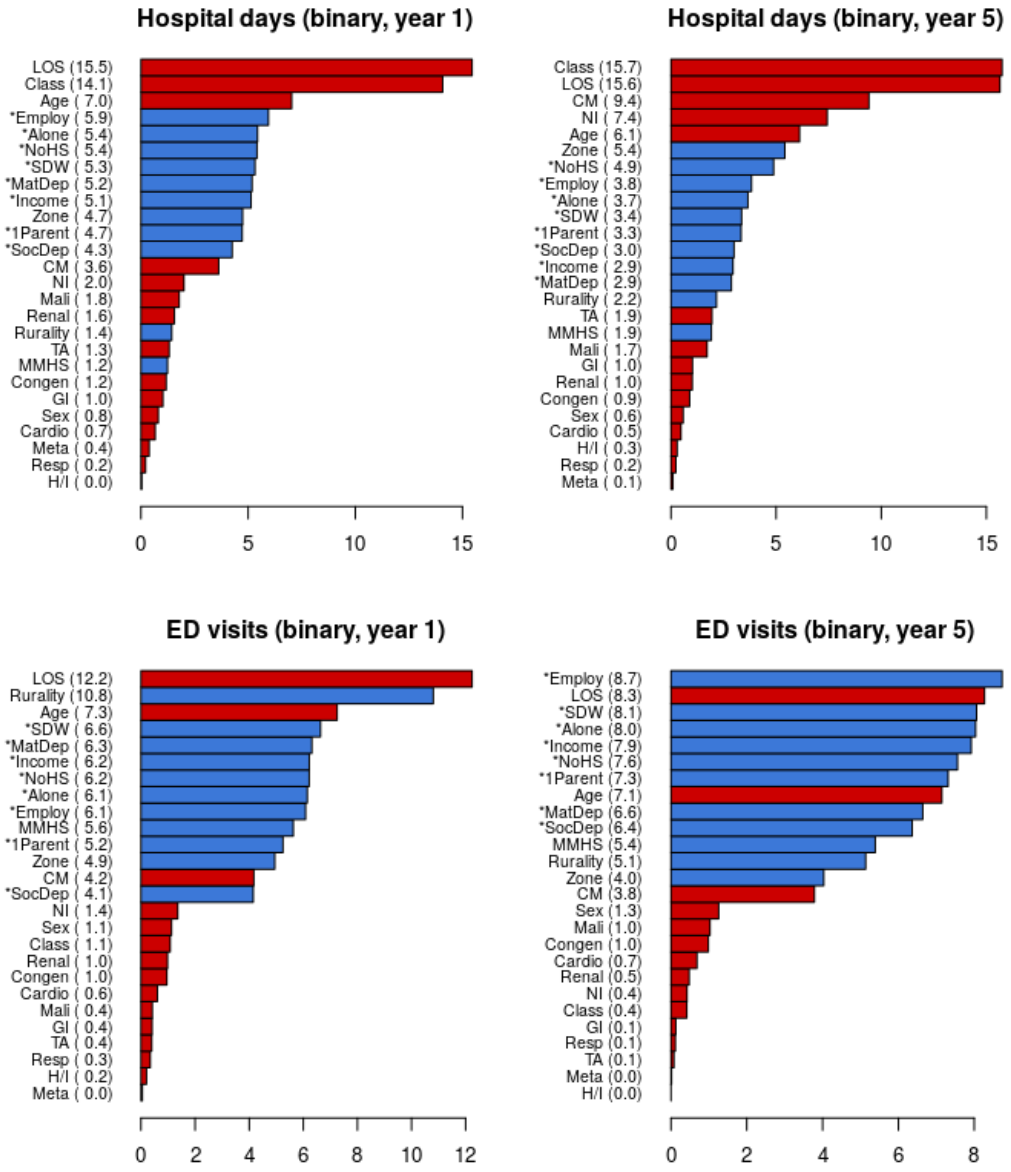
Initial LOS was also the most important predictor of ED visits within the first year, but was close in importance to residence rurality and about 1.5–2.5 times as important as age (the third most important predictor) and the other socioeconomic factors. Among CMCs with ED visits in the first year, however, socioeconomic variables (namely rurality, AHS zone, and mental health service use) were the most important predictors of number of ED visits. In particular, rurality was at least 3 times as important as initial LOS and age, the most important clinical/demographic variables.

Long-term Predictors

Clinical classification and initial LOS had similar importances and were top predictors of the presence of a hospital stay within 5 years. Both were at least 2 times as important as any socioeconomic factor. Number of chronic medications, the presence of neurological impairment, and age were the third through fifth most important predictors. Similar to the short-term results, initial LOS was by far the most important predictor of number of hospital days (among CMCs with a hospital stay within the 5 years), and was at least 5.5 times as important as clinical classification and number of chronic medications (the next most important predictors) and any other socioeconomic factor.

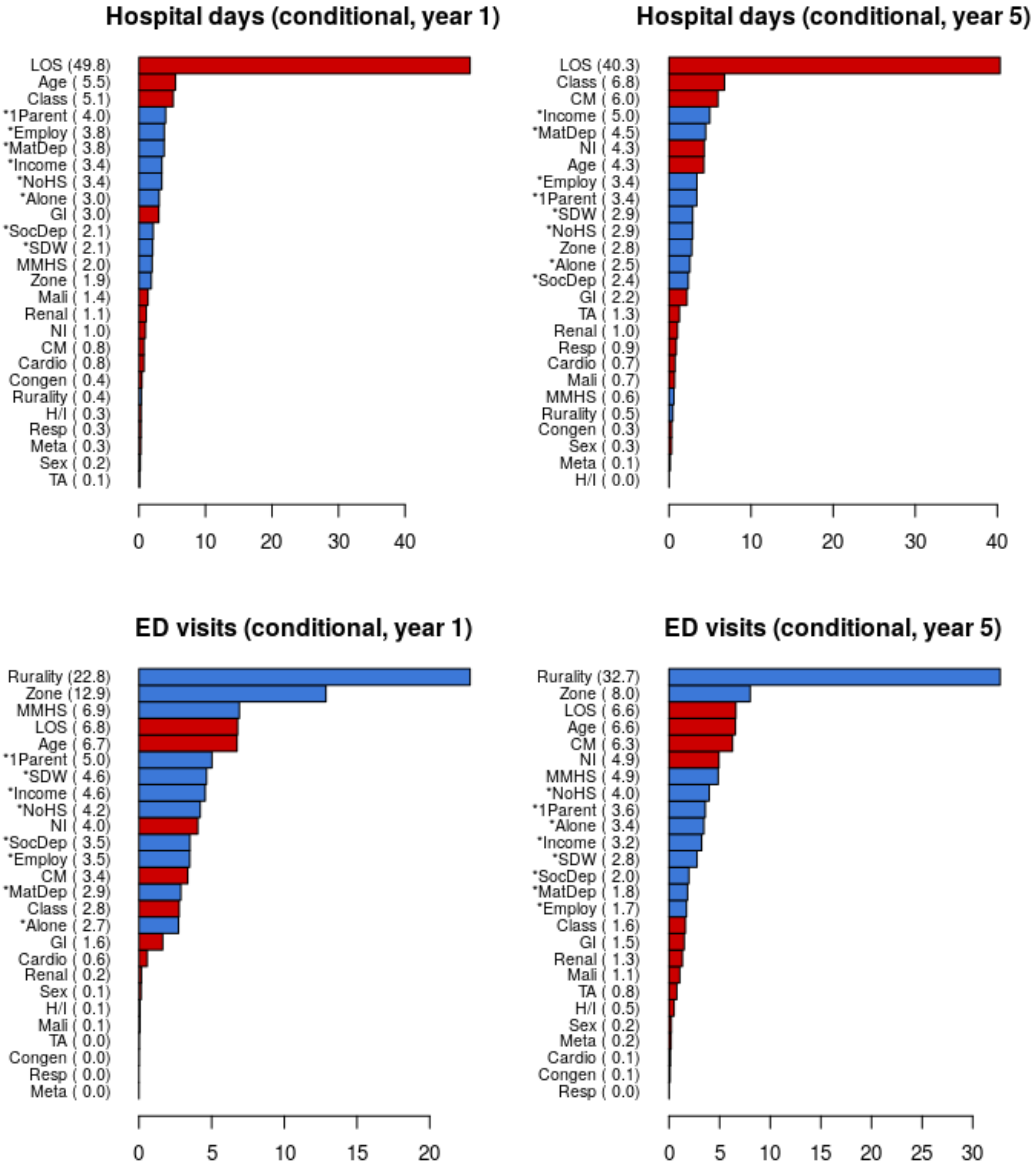
The top 10 predictors of the presence of ED visits were comparable in terms of relative importance. All but 2 (initial LOS and age) of these predictors were socioeconomic factors. Among CMCs with ED visits within 5 years, residence rurality was at least 4 times as important as every other predictor.

Figure 4.1. Relative variable importances (in parentheses) for the binary submodels. Clinical and socioeconomic variables are indicated with red and blue, respectively. An asterisk (*) denotes a DA-level measure.



Abbreviations: 1Parent, proportion of single-parent families; Alone, proportion of individuals living alone; Cardio, cardiology condition indicator; Class, single or multiple complex chronic conditions; CM, number of chronic medications; Congen, congenital/genetic condition indicator; Employ, employment rate; GI, gastrointestinal condition indicator; H/I, hematology/immunodeficiency condition indicator; MatDep, material deprivation score; Mali, malignancy condition indicator; Meta, metabolic condition indicator; MMHS, mother mental health service use; NI, neurological impairment indicator; NoHS, proportion of individuals without a high school diploma; Renal, renal condition indicator; Resp, respiratory condition indicator; SDW, proportion of individuals who are separated, divorced, or widowed; SocDep, social deprivation score; TA, technology assistance indicator.

Figure 4.2. Relative variable importances (in parentheses) for the conditional submodels. Clinical and socioeconomic variables are indicated with red and blue, respectively. An asterisk (*) denotes a DA-level measure.



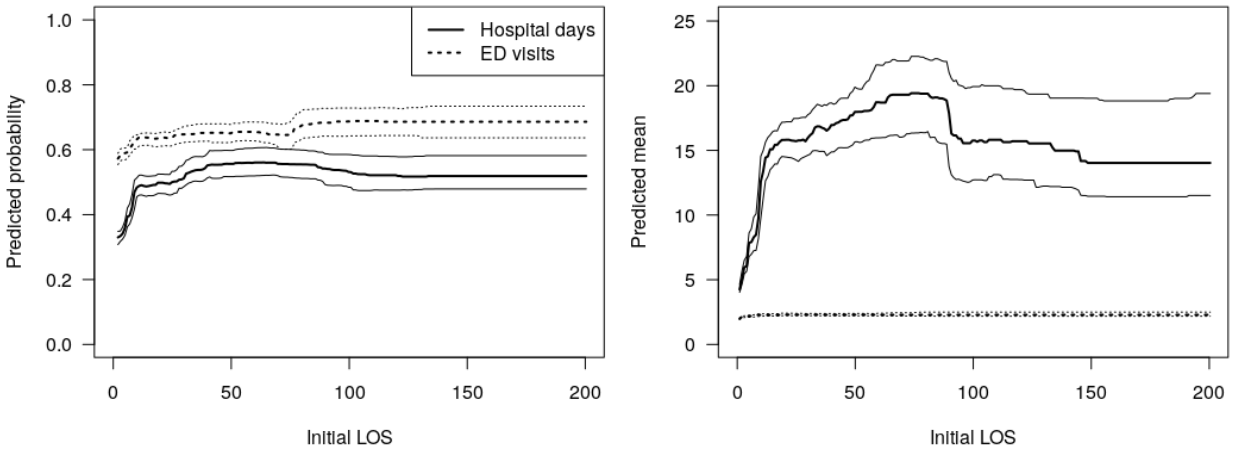
Abbreviations: 1Parent, proportion of single-parent families; Alone, proportion of individuals living alone; Cardio, cardiology condition indicator; Class, single or multiple complex chronic conditions; CM, number of chronic medications; Congen, congenital/genetic condition indicator; Employ, employment rate; GI, gastrointestinal condition indicator; H/I, hematology/immunodeficiency condition indicator; MatDep, material deprivation score; Mali, malignancy condition indicator; Meta, metabolic condition indicator; MMHS, mother mental health service use; NI, neurological impairment indicator; NoHS, proportion of individuals without a high school diploma; Renal, renal condition indicator; Resp, respiratory condition indicator; SDW, proportion of individuals who are separated, divorced, or widowed; SocDep, social deprivation score; TA, technology assistance indicator.

4.4.4 Marginal Associations

Short-term Predictors

Figure 4.3 shows marginal association estimates for initial LOS, a consistently important predictor of hospital days. Among CMCs with a hospital stay, the mean number of hospital days in year 1 increased from about 5 to 20 days as initial LOS increased to 100 days, and afterwards decreased and plateaus. The marginal association between initial LOS and the probability of a hospital stay was similar and increased (from about 0.30 to 0.55) with initial LOS up to 100 days.

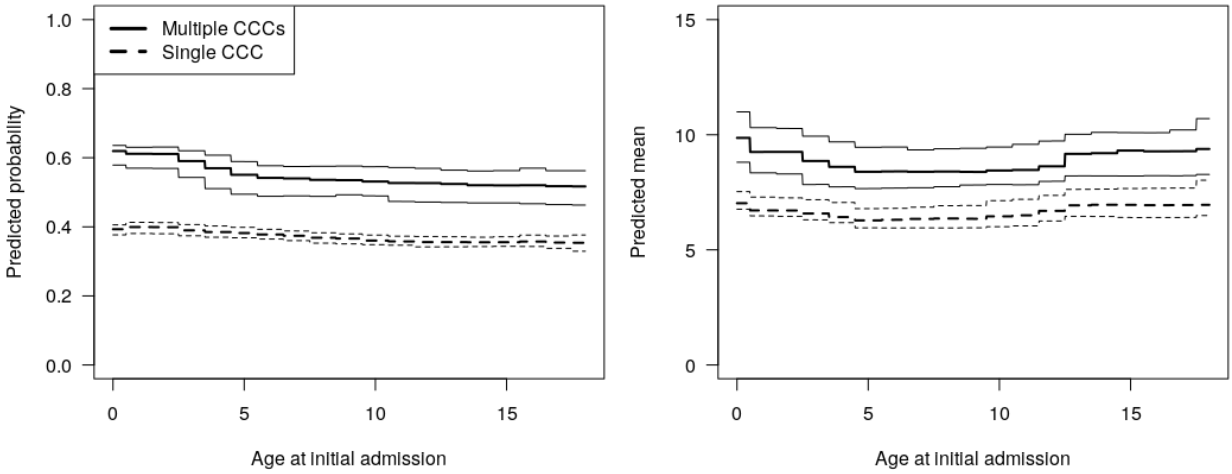
Figure 4.3. Marginal associations (and 95% confidence intervals) for initial LOS in the binary (left) and conditional (right) submodels for hospital days (solid lines) and ED visits (dotted lines) in the year following initial discharge.



Abbreviations: ED, emergency department; LOS, length of stay.

As shown in Figure 4.4, younger patients were slightly more likely to be hospitalized than older patients in the first year. This was particularly true for CMCs with multiple CCCs. The marginal association between age and the probability of having an ED visit was similar, but was nearly zero in the conditional submodel for number of ED visits (Appendix B Figure B4.1).

Figure 4.4 Marginal associations (and 95% confidence intervals) for age at initial admission in the binary (left) and conditional (right) submodels for hospital days among CMCs with multiple CCCs (solid lines) and a single CCC (dotted lines) in the year following initial discharge.



Abbreviations: CCC, complex chronic condition; CMC, child with medical complexity.

Relative to CMCs with a single CCC, those with multiple CCCs were more likely to have hospital days (0.59, 95% CI 0.55–0.60 vs 0.38, 95% CI 0.36–0.39) in the first year. Similarly, among CMCs with hospital days in the first year, those with multiple CCCs had more hospital days on average (9.37, 95% CI 8.49–6.61 vs 6.85, 95% CI 6.61–7.28) than those with a single CCC.

CMCs in remote rural (0.67, 95% CI 0.64–0.70) areas were the most likely to have an ED visit in the first year, followed by those in rural (0.67, 95% CI 0.64–0.68), urban (0.55, 95% CI 0.54–0.57), and metropolitan (0.55, 95% CI 0.54–0.56) areas. Similarly, among CMCs who had ED visits, those in remote rural (2.62, 95% CI 2.45–2.80) areas had the most visits on average, followed by those in rural (2.56, 95% CI 2.45–2.80), metropolitan (2.00, 95% CI 1.94–2.04), and urban (1.99, 95% CI 1.93–2.05) areas. See Appendix C Table C4.2 for estimates by AHS zone and rurality, which are consistent with the above marginal estimates.

Marginally, among CMCs with ED visits in the first year, those whose mothers had used mental health services had a higher number of ED visits relative to those whose mothers did not use these services (2.42, 95% CI 2.31–2.52 vs 2.09, 95% CI 2.04–2.13). Estimates where data on service use was unavailable were comparable to those for the latter group (2.09, 95% CI 2.04–2.14).

Long-term Predictors

The marginal effects of initial LOS on 5-year outcomes were similar to those for the first year (Appendix C Figure C4.2).

As in the first year, CMCs with multiple CCCs were more likely to have hospital days than CMCs with a single CCC (0.68, 95% CI 0.65–0.70 vs 0.51, 95% CI 0.50–0.52). Among CMCs with hospital days, those with multiple CCCs also had more hospital days on average than those with a single CCC (12.7, 95% CI 11.38–13.83 vs 8.54, 95% CI 8.15–8.89).

Marginally, CMCs with more chronic medications were more likely to have a hospital stay within the 5 years (0.70, 95% CI 0.63–0.73 for ≥5 medications; 0.62, 95% CI 0.58–0.65 for 2-4 medications; 0.58, 95% CI 0.55–0.61 for 1 medication; 0.52, 95% CI 0.51–0.53 for 0 medications). Similarly, among CMCs with hospital days, those with more chronic medications had more on average (15.15, 95% CI 12.37–17.71 for ≥5 medications; 10.93, 95% CI 9.70–11.93 for 2-4 medications; 9.92, 95% CI 9.14–10.77 for 1 medication; 8.76, 95% CI 8.44–9.20 for 0 medications).

While employment rate (at the DA level) was the most important predictor of the presence of ED visits by the fifth year, its marginal effect was close to zero (Appendix C Figure C4.3). Similarly, while marital status (namely, the proportion of individuals in a DA who are

single, divorced, or widowed) was the third most-important predictor of the presence of hospital days, its marginal effect was also plausibly zero (Appendix C Figure C4.4).

Similar to short-term outcomes, CMCs in remote rural (0.88, 95% CI 0.86–0.88) and rural (0.88, 95% CI 0.86–0.89) areas were the most likely to have an ED visit within the 5 years, followed by those in urban (0.83, 95% CI 0.82–0.85) and metropolitan (0.83, 95% CI 0.82–0.84) areas. Similarly, among CMCs who had ED visits, those in remote rural (6.16, 95% CI 5.70–6.61) areas had the most on average, followed by those in rural (6.04, 95% CI 5.74–6.28), metropolitan (3.96, 95% CI 3.86–4.07), and urban (3.96, 95% CI 3.84–4.12) areas.

4.5 Discussion

To our knowledge, this is the first study to develop a machine learning model to identify predictors of hospital days and ED visits among CMCs. We used a typical curated administrative dataset containing proxies for both clinical complexity and socioeconomic status. Generally, and for both short- and long-term outcomes, clinical variables were the most important predictors of (the presence and number of) hospital days while socioeconomic variables were the most important predictors of ED visits. Our results are consistent with the Andersen (1995) behavioural model of access to health care resources, specifically regarding the importance of socioeconomic and demographic characteristics beyond health care availability.

4.5.1 Clinical Variables

As might be expected from proxies for clinical complexity, initial LOS and clinical classification (ie, single vs multiple CCCs) were top predictors of hospital use. The generally consistent importance of initial LOS as a predictor of future resource use should prompt further

investigation of factors related to initial LOS, including the collection of more-nuanced data from the initial hospitalization event (e.g., procedures performed, medications administered, physician assessment notes, etc). Understanding specific risk factors that are related to initial LOS and are predictive of subsequent hospital and ED utilization would support service planning and the development of care pathways.

Similarly, the importance of clinical classification and number of chronic medications as predictors of hospital use could suggest that CMCs with multiple CCCs or a large number of chronic medications (at initial admission) be provided additional supports beyond the first year in response to elevated projections for long-term resource use.

Differences in the patterns of importance between the short- and long-term models for hospital stays and ED visits suggested that individual clinical indicators were typically less useful as predictors than the other variables considered. However, the neurology clinical indicator was at least 4–5 times as important as the other clinical indicators in predicting the number of hospital days and ED visits over the 5 years among CMCs who used these resources. This result suggests that the subpopulation of CMCs with neurological conditions are more vulnerable and may require increased health care resources relative to other CMCs which is consistent with other studies in the literature (Berry et al. [2009], Hudson et al. [2013], Agrawal et al. [2016]).

4.5.2 Socioeconomic Variables

CMCs in rural and remote rural locations as well as those in the “mainly rural” AHS zones had the highest probability of an ED visit. This could be due to several reasons, including unmet care or the availability of after-hours care or a medical home. This finding is consistent

with a U.S. study by Barnert et al. (2018), who reported that socioeconomic factors such as geographic location and economic position are associated with access to care and parental stress for CMCs and their families.

The finding that CMCs whose mothers accessed mental health services (prior to their child's initial admission) were more likely to have ED visits in the first year prompts further investigation. It is possible that mothers with a history of accessing mental health services are less able to manage their child's illness and thus require more support (Dewan et al., 2023). Conversely, it is also possible that mothers who access the health system for themselves are more likely to be able to navigate and use health care resources for their child. Since we do not have the data to understand what services these mothers are utilizing, we must leave delineating the exact nature of this relationship to future work.

Speaking to ED visits specifically, the most important predictors of long-term resource use (except initial LOS) were socioeconomic in nature. From the relatively high importance of employment status and marital status as predictors of the presence of ED visits, we hypothesize that family structure, residence rurality, and financial resources are important to CMC outcomes. The near-zero marginal associations of these and other socioeconomic variables (not shown in this work) such as living alone, income, education, single parenting, material and social deprivation, and maternal mental health access, however, suggests that their importance is "hidden" in higher-order interactions with other variables. The complexity of these interactions makes it difficult to directly identify policies improving CMC and family outcomes without further specific research.

These results confirm the importance of population characteristics as described in the Anderson (1995) model: socioeconomic factors such as financial resources and residence location together with health care policies influence how CMCs and families use health care resources. For example, Alberta has 2 large pediatric hospitals located in large metropolitan cities. It is possible that CMCs who live in (remote) rural locations have higher ED use due to unequal access to health care resources and supports such as medical homes or care-coordination services, and that this inequity makes EDs the most feasible option for CMC families. It is possible that different policies and processes in rural care delivery contribute to this inequity. For example, in rural Alberta, primary care physicians provide service in the local emergency department, which potentially reduces available clinic hours. According to Hudson (2013), there are few studies examining hospital readmission and ED use for children with special health care needs in rural and community settings relative to regional centres.

Lastly, the marginal effect of age showed that younger CMCs with multiple CCCs were more likely to be hospitalized than patients diagnosed at an older age. This finding is consistent with Agrawal et al. (2016), who showed that the top 5% of child health care users on Medicaid did not remain in the top 5% in subsequent years, indicating reduced health care resource needs over time. This is an important consideration since about half of our cohort was less than 1 year old at initial admission. Anticipating changes in health care resource needs for younger CMCs over time can aid in effective policy and care delivery programs.

4.5.3 Machine Learning and Health Administrative Data

Beyond existing applications in health care to improve diagnostics and identify risk factors for various diseases (Jiang, et al., 2017), there is substantial potential for machine

learning in health care. This includes supporting clinical decision-making (Arbet et al., 2020), identifying risk factors within patient populations (Xu et al., 2018; Su et al., 2020), supporting research in epidemiology (Casey et al., 2016), and identifying factors affecting health care resource use (Maynard et al., 2019). Machine learning models, relative to traditional statistical methods such as regression, may be superior for predictive modelling in heterogeneous, clinically complex populations such as CMCs because they are better equipped to handle high-dimensional data and complex interactions (King & Strumpf, 2022). However, the content of administrative health data, including those collected in existing EHRs, may be limiting this potential. EHRs should be designed to facilitate and encourage predictive modelling by collecting diverse, detailed, patient-level characteristics. Patient-level measures such as income level, family structure or parental access to respite care are all examples of socioeconomic data that can be included in EHRs. Clinical notes such as home-care nursing visits, structured assessment notes (beyond ICD-10 codes) can also be included in support of this goal but are substantially more complicated to analyze. Early evidence suggests that the use of predictive modelling with well-designed EHR systems can improve patient outcomes and clinical decision making (Lee et al., 2020).

This study examined both clinical and socioeconomic variables and showed that the latter were particularly important predictors of (binary) ED utilization in the short and long terms. The performance of our conditional models suggested a high amount of unexplained variability in both the number of hospital days and ED visits. We originally observed this unexplained variability in a set of random forest models (not presented here) trained on the same data. Despite the relatively greater capacity and expressiveness of the boosted models presented in this work, we saw little improvement in the performance of the conditional models. While this

reflects the fact that forecasting resource use for CMCs is an inherently difficult task (Wang & Alexander, 2020), it may also suggest that there is insufficient signal in the administrative data used in this analysis. These findings motivate our previous calls for more-robust clinical and socioeconomic data in EHRs in order to better support predictive modelling for CMC populations in the future. Socioeconomic factors are particularly important to consider for the CMC population because the families of CMCs struggle with economic and psychosocial effects that impact the child, family health, and subsequent use of health care resources (Barnert et al., 2017; García-Altés et al., 2018).

4.6 Limitations

This study has limitations common to retrospective research with curated administrative data. First, the socioeconomic data linked to our clinical data represent DA-level characteristics, which do not accurately reflect individual- or household-specific variability. Second, the independent variables in this study were only available around initial admission or discharge, while time-varying predictors (ie, varying across the 5 years) may yield more-interpretable and more-practical models. Third, while ICD codes are well defined and readily used in administrative health data to identify CMCs, they lack detail regarding the medical complexity of the patient, which can further contribute to a reduced ability to represent individual CMCs with available data. Another consideration for interpreting socioeconomic predictors such as rurality and AHS Zone is that the availability of primary care services was not available in the dataset and uneven distribution of pediatric specialty services (e.g., large pediatric centers are available in Edmonton and Calgary Zones but not the rest of the province) may be confounding factors.

As discussed above, finer patient-level data such as details of the initial LOS encounter, patient-level socioeconomic factors, and names of chronic medications prescribed may be necessary to explain the large amount of variability in resource use observed in this study. This is in line with our call for EHRs to collect more-detailed patient-level data, particularly that related to socioeconomic conditions. However, future work is necessary to examine practical aspects of EHR design and burdens associated with data collection.

4.7 Conclusion

This study used a machine learning model and readily available administrative health care data to identify important predictors of hospital days and ED visits, in both the short and long terms, among CMCs in Alberta, Canada. Initial LOS and clinical classification, factors indicative of medical complexity, were strong predictors of hospital days. Residence rurality, socioeconomic metrics, and initial LOS were predictive of ED use. Our results are in line with existing literature and suggest the necessity of further examinations of the impact of residence rurality and how an initial hospitalization event influences outcomes for CMCs and their families. We encourage health care systems to actively design data collection systems and EHR processes that support robust analyses and the development of high-accuracy predictive models for the highly heterogeneous CMC population.

4.8 Acknowledgment

The authors thank Taranpreet Kaur, lead maternal and child data analyst at Alberta Children's Hospital (Calgary, Canada), who pulled and provided information on the data set used. She was not compensated beyond her established salary for her contributions.

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Chapter 5. Discussion and Conclusion

5.1 Overview and Significance

The work in this dissertation filled several research gaps and added to existing knowledge about the CMC population, both in Alberta and generally. The systematic review identified that the Complex Chronic Condition classification system was the most common approach in the literature for identifying CMCs. It also highlighted variation in how health care costs are reported across and within jurisdictions. The quantitative analyses found associations between proxy measures of clinical complexity (e.g., number of chronic medications, initial LOS) and hospital days and between socioeconomic factors (e.g., residence rurality, income) and ED visits.

Studies have demonstrated significant variation in how CMCs use acute care resources within and across jurisdictions as well as the potential to optimize and improve care for this population (Coller et al., 2017, 2019). In MyChild, 1.5% of CMCs accounted for 28% of all hospital days and 1.7% of CMCs accounted for 15.7% of ED visits among this cohort. Uneven resource use within the Alberta CMC population suggests that targeted interventions for CMCs be considered, specifically for CMC subpopulations' higher acute care use. Existing literature supports this suggestion with reported reductions in CMC hospitalization following enhanced care coordination and other child- and family-focused interventions (Palfrey et al., 2004) and numerous promising strategies for reducing ED visits (Pulcini et al., 2021). From a health system perspective, optimizing health care delivery is a growing priority for governments as health care spending accounts for an increasingly large percentage of government spending. Alberta's 2023–2024 budget, for example, allocates $24.5 billion (almost 35% of Alberta's budget) to health care, which is an increase of 4% from the previous budget (Government of Alberta, 2023). From

patient and family perspectives, hospitalizations and ED visits present additional stresses such as out-of-pocket expenses, the stress of having a child treated in an ED by physicians that might not be familiar with the child's medical history, and the need to arrange childcare for the siblings of CMCs.

Understanding how CMCs are identified in the literature and how costs are reported is important for knowledge translation and policy development. Chapter 2 presented a systematic review of how CMCs are identified in the administrative health literature and how health care resource use by CMCs is measured and reported. The review found considerable variation in reported costs, including health care resources and costs incurred by families. Most (89%) of the studies reported hospital costs, while some (U.S.-based, for-profit hospitals) reported hospital charges, others reported number of hospital days, and others reported macro-level hospital costs that included diagnostic fees and overhead allocations. This variation makes it difficult to compare jurisdictions and highlights the need for local evidence on CMC health care utilization. Information on the Alberta CMC population was a gap identified in the systematic review.

Chapters 3–4 built on the systematic review and the existing literature by quantitatively examining how clinical and socioeconomic factors are associated with hospital and ED use. The relatively simple summaries of CMC resource use available in previous literature did not provide sufficient information on these associations or health care costs. This limitation is particularly problematic in light of established theoretical frameworks of health service use such as that by Andersen et al. (1995), which highlights the importance of both patient and delivery system characteristics as factors associated with resource use. The analyses in Chapters 3–4, consider both clinical and socioeconomic factors simultaneously, and are therefore significant additions to the literature. The trends and relationships described in these chapters, such as between clinical

complexity (e.g., initial LOS) and hospital days and between socioeconomic factors (e.g., residence rurality) and ED visits, are relevant to both health care providers and administrators. From a health system perspective, these findings are important as they enhance understanding and enable more-informed decision-making for the CMC population, which is known to use substantial amounts of acute care resources. As hospitalizations and ED visits place significant financial and psychosocial burdens on the families of CMCs (Connor et al., 2010), identifying factors associated with health care utilization may lead to more-effective spending of public funding, reduced stress for families, and improved access to health systems for CMCs (Ungar, 2009). The results of this dissertation further the understanding of these associations and enable more-focused discussion on how to improve care, support families, and optimize resource use for CMCs in Alberta.

Chapter 3 presented a longitudinal, inferential regression analysis of associations between clinical/socioeconomic factors and hospital and ED use over the five-year period following initial hospitalization. The analysis highlighted the importance of initial LOS, particularly within certain clinical groups (discussed in Sections 5.2.1–5.2.2), and number of chronic medications (see Section 5.2.3) since both were positively associated with hospital days (both in the sense of having hospital days and, when present, number of hospital days). The positive associations between number of chronic medications and resource use may be attributable to increasing medical complexity or overmedicalization (as discussed in Section 5.2.4). Socioeconomic factors were significantly associated with ED visits, which confirmed the importance of socioeconomic factors posited in theoretical frameworks for health care resource use (see Section 5.2.5). The results of Chapter 3 point to how clinical, demographic, and socioeconomic factors play different but important roles in how CMCs utilize health care resources. Clinical factors such as initial

LOS and number of chronic medications are more associated with hospital days while socioeconomic factors such as material/social deprivation and residence rurality are more associated with ED visits.

Chapter 4 complemented the inferential analysis of Chapter 3 with a predictive analysis conducted via a boosted regression tree model. This chapter accomplished two objectives: it identified clinical and socioeconomic predictors of hospital days and ED visits and evaluated the utility of curated administrative health care data in predictive modelling for CMCs. The analysis in this dissertation quantified the importance of clinical and socioeconomic factors as predictors of each outcome in the short and long terms (i.e., one and five years following an initial admission) with a model that is novel in CMC research. The decision to use tree-based ensemble method (boosted regression tree in this case), is in line with recommendations from existing health service research literature (King & Strumpf, 2022). While single regression trees were considered, ultimately, a gradient boosted model was used because of its' superior statistical properties (Hastie et al., 2009).

Initial LOS was among the top predictors of hospital and ED use in both time frames. Other top predictors of hospital days were predominantly clinical in nature (e.g., the presence of single or multiple chronic conditions, number of chronic medications). In contrast, other top predictors of ED use were primarily socioeconomic in nature (e.g., rurality, mother use of mental health services, DA-level marital status). Chapter 4 was able to identify important predictors (rather than just correlates) and accommodate complex interaction structures in a way that complements the traditional regression analysis of Chapter 3. Chapter 4 sets a baseline for future research using administrative health data to study the heterogeneous CMC population. While this analysis was able to identify important predictors at a level suitable for policy-making, existing,

routinely collected administrative health data might not be sufficient for high-accuracy predictive modelling in CMC research. The study makes suggestions regarding the type of data that, if added to administrative health databases, might support more-robust predictive modelling in the future.

5.2 Specific Implications

The results presented in this dissertation show that both clinical (e.g., initial length of stay, number of chronic medications, type of chronic condition) and socioeconomic factors (e.g., residence location, income, marital status) were associated with hospital and ED use, respectively. The following section discusses specific implications of these results.

Information on factors impacting hospital and ED use among Alberta CMCs is necessary to effectively evaluate and optimize care delivery for CMCs. This dissertation addressed a gap in the extant literature by identifying specific clinical and socioeconomic factors associated with and predictive of hospital use and ED visits longitudinally. The analyses presented used a rigorous statistical framework that properly accounted for characteristics of data on CMC resource use (the disproportionate amount of zeros) that have the potential to invalidate statistical analyses with standard methods but have not previously been considered in other literature. Several findings from this dissertation carry important implications pertaining to hospital and ED use by CMCs and should be considered when optimizing care for this population.

5.2.1 Initial Length of Stay: A Case for Robust Electronic Health Records

Initial LOS consistently had significant associations with hospital days and ED visits in the inferential analysis and was consistently among the top five predictors of both responses in

the predictive analysis. In the former, a 10-fold increase in initial LOS had a significant, positive association with number of hospital days (among CMCs with hospital days) within every clinical category except for gastrointestinal and hematology/immunodeficiency. Log transformation of the responses in the conditional model enabled this level of interpretation. The analysis was able to estimate the effect of initial LOS within each clinical group and further motivates research on initial LOS (with specific clinical groups), since initial LOS is itself a summary of multiple factors related to the initial hospitalization that should be explored in more detail in the future.

The marginal effect of initial LOS as a predictor in the boosted regression model was similarly positive, here increasing until about 100 days before levelling off. Initial LOS was also among the top two predictors, in both the first and fifth years, of the presence of hospital days and (among CMCs using resources) number of hospital days. The importance of LOS was also highlighted by Gold et al. (2016), who found associations between a high initial LOS (>10 days) and receiving care in the ICU, respiratory complex chronic conditions, and transfers to another acute hospital during initial admission. These findings support the development of policies that consider CMCs with a high initial LOS for more support in order to better manage care outside the hospital. While not all CMC hospitalizations are avoidable, reducing preventable hospitalizations benefits patients and the health system. Maynard et al. (2019) found that 5%–7% percent of pediatric discharges needed home care support due to clinical complexity or technology assistance, and that unavailability of home care caused over half of delayed discharges and resulted in longer LOS.

As described above, the literature has considered LOS from all hospital stays, but the results of this dissertation suggest more-detailed research related to initial LOS within specific clinical groups. Future work can investigate factors specific to individual clinical categories that

contribute to increased hospital and ED use though long initial LOS. This could be accomplished, for example, by considering information on specific medications prescribed, physician assessment notes, or procedures performed during initial hospitalization. However, not all this data is easily accessible in existing administrative health datasets.

Initial LOS provides an apt example of how designing more-robust data collection procedures, such as through EHRs, can aid in expanding evidence and understanding of CMCs. Chapter 4 discussed the importance of detailed administrative health data from the perspective of predictive modelling. Longitudinal, patient-level data such as clinical assessment notes from initial LOS and other variables known to be associated with initial LOS are examples of data that would be useful in further investigations and, more generally, help improve the performance of predictive models.

5.2.2 Clinical Groups: Identifying High-risk Subpopulations

The analysis in Chapter 3 found that, regardless of clinical group, mean cumulative hospital days and ED visits increased steadily over the five years. Notably, the neurology subgroup appeared to have the steepest trajectories among all other subgroups. The effect of initial LOS was furthermore second largest within the neurology group. Similarly, the analysis in Chapter 4 found the neurology indicator to be the fourth most-important predictor of the presence of hospital days in the fifth year and the sixth most-important predictor of number of hospital days and ED visits (when present). No other clinical indicator was more important than that for neurology.

These results suggest that patients with neurological conditions have a particularly high risk of requiring hospital and ED services over time relative to other clinical groups. This is an

important finding because parents and caregivers of these CMCs are likely to experience more stress, which may lead to worse outcomes for the child and family. In Canada, CIHI (2020) found that 32% of CMCs had at least one hospital stay, and that 61% of children with neurological impairment together with other conditions had at least one hospital stay (in contrast to 36% of all CMCs in Canada between 2015 and 2016). Furthermore, primary caregivers of children and youth with neurological impairment reported more feelings of anger, distress, or depression than the caregivers of CMCs with a single or multiple chronic conditions.

CMCs with neurological impairment are also at a higher risk of receiving uncoordinated care, having unmet needs, and overusing inpatient hospital services, especially among youth transferring from pediatric to adult care (Berry et al., 2011). More-detailed investigations of resource utilization patterns within this group in the larger health system, including outpatient services, primary care, and home care, would be beneficial towards optimizing care and providing adequate support for families.

5.2.3 Age: Importance of Longitudinal Analysis

The longitudinal focus of Chapters 3–4 is a salient feature of this work considering the previous finding that CMCs, as they grow older, are more likely to develop comorbidities due to their primary illness (Crow et al., 2017). Longitudinal assessments can provide further insight through time-aware analyses of associations between resource utilization and patient-level clinical characteristics. The hurdle model in Chapter 3 accomplished this by including repeated measures and various interaction effects with time. Chapter 4 analysis took a different approach by modelling outcomes for the first and fifth years separately in order to identify differences in variable importance in the short and long terms.

In terms of age, the analysis in Chapter 3 found that newborns had significantly more hospital days and ED visits. In the year following an index admission (first admission with a Complex Chronic Condition) and among CMCs with hospital days, newborns had significantly more hospital days on average than CMCs aged one–nine years. Similarly, newborn CMCs had more ED visits on average in the first year relative to CMCs between the ages of five and eighteen. In Chapter 4, age was the third-most-important predictor of hospital and ED use in the first year. Interestingly, there were no significant differences in mean cumulative hospital days (among CMCs with hospital days) between the age groups in the fifth year, which indicated that resource use in and differences between CMCs groups (on the basis of age at initial admission) change over time. These results are consistent with a U.S.-based study by Agrawal et al. (2016) that found that children accounting for the top 5% of all Medicaid spending did not remain in the top 5% and had lower health care costs in subsequent years. Knowing that hospital and ED use decreases as CMCs age has implications for resource allocation. For example, health administrators may anticipate that more care coordination and other family support programs are needed for CMC cohorts with more newborns.

5.2.4 Chronic Medications: CMCs and Overmedicalization

Number of chronic medications had significant associations with and was generally an important predictor of hospital days and ED visits throughout Chapters 3–4. Despite being unconstrained, parameters in the hurdle regression model suggested a strictly increasing effect of chronic medications, i.e., more chronic medications in the year after an initial hospitalization was associated with a higher average cumulative number of hospital days and ED visits (when these resources were used) throughout the five-year period. Similarly, the boosting model consistently

identified number of chronic medications (here instead for the year prior to an index admission) as a strong predictor of hospital days in the fifth year.

While it is intuitive that CMCs would require multiple chronic medications, it is important for clinicians to also consider the possibility of overmedicalization—the overuse of diagnostic testing and unnecessary medical treatments including drug prescriptions (Marty et al., 2019; Pordes et al., 2020). According to Pordes et al. (2020), many risk factors, including the child's medical condition, caregiver circumstances, and health care system issues, may be indicative of overmedicalization. Despite the relatively small number of CMCs with one or more chronic medications (35.1%), the results of this dissertation showed that the number of chronic medications is positively associated with and predictive of increased hospitalization and ED use. Although this finding could feasibly be explained as a confounding association with medical complexity, further evaluation of the potential for overmedicalization among CMCs with multiple chronic medications appears to be warranted. Pordes et al. (2020) presents a framework for how this evaluation could be completed. Decision makers should consider the number of chronic medications in criteria for allocating resources (e.g., care coordination) to CMCs and their families.

5.2.5 Socioeconomic Factors

Theoretical frameworks such as the Andersen Model and the Institute of Medicine models highlight the importance of socioeconomic factors for understanding health care resources use. Factors such as residency, clinical characteristics, and socioeconomic factors are all considered important predictors of health care resource use under these frameworks (Karikari-Martin, 2010). To the author's knowledge, this is the first quantitative analysis of the CMC

population in Alberta and the first Canadian study to link socioeconomic and administrative health care data in an analysis of CMCs.

The models in this dissertation found statistically and clinically significant associations between socioeconomic factors and resource use (both in terms of hospital days and ED visits) and consistently point to the importance of rurality in ED use by CMCs. For example, the conditional submodels in Chapter 3 suggested that CMCs in urban, rural, and remote rural areas were more likely to have ED visits and (among CMCs with ED visits) had significantly more cumulative visits than those living elsewhere. The models in Chapter 4 agree with this conclusion and additionally found that rurality was the second-best predictor of the presence of ED visits in the first year (among CMCs with ED visits) and the best predictor of the number of ED visits in the first and fifth years after initial hospitalization.

While more research is needed to explore causal pathways between socioeconomic factors and resource use among CMCs, a feasible hypothesis supported by the results in this dissertation is that CMCs in rural areas utilize the ED instead of other care services such as primary care clinics, after-hours care, home care, or specialized care either because these services are unavailable or are difficult to access. This hypothesis is consistent with existing literature in this field. For example, only 8% of physicians in Canada are located in Urban areas whereas 92% are in larger centers (CIHI, 2021). Primary care physicians in rural Alberta deliver service at the local ED in addition to their clinic, which reduces access to primary care clinic hours. Access issues in rural locations are not limited to Alberta. Young et al. (2016) illustrates worse health outcomes in Canada's Northern communities compared to the rest of Canada. Major et al. (2018) describes an intervention in Ontario designed to fill service gaps for CMCs in the northern rural and remote rural areas of the province. Most rural ED practitioners are likely

not knowledgeable regarding the unique needs of CMCs: in a survey by Murphy et al. (2012), 77% of primary care providers (including physicians and nurse practitioners) indicated having difficulty treating patients with complex chronic conditions. For the families of CMCs, moving closer to metropolitan areas and to specialty care might not be an option due to significant financial hardships or a loss of support from extended family and the community (CIHI, 2020).

Andersen et al. (1995) posited that socioeconomic factors together with equity of access to health care play an important role in determining health service use. The results of this dissertation, specifically the importance of rurality, are consistent with and affirm the Andersen Model. Furthermore, the results of this dissertation, along with other current literature on this topic, motivate further investigations of the availability of care services to CMCs outside of EDs in nonmetropolitan areas of Alberta. Programs such as care coordination, caregiver training, and respite care have been shown to reduce the burden on families of CMCs and reduce health care resource use (Cohen et al., 2012b). The hub-and-spoke delivery model piloted in Ontario (Major et al., 2018) showed promise in supporting rural and northern CMC communities and may also be an effective model in Alberta. Other types of interventions, such as real-time access to providers familiar with the child, 24/7 access to ambulatory appointments with knowledgeable providers (Pulcini et al., 2021), and family training and support (Van Orne et al., 2018), may be useful in optimizing ED use and reducing stress on families and caregivers.

Chapter 4 further identified several specific socioeconomic measures as important predictors of hospital days and ED visits: nine of the top 12 predictors of the presence of hospital days in the year following a CMC's initial admission were components of the Pampalon deprivation scores or other socioeconomic indicators, as were 10 of the top 12 predictors of the presence of ED visits (e.g., employment status, education level, income).

Interestingly, mothers' access to mental health services the year before initial admission was the third-most-important predictor of number of ED visits (among CMCs with ED visits) in the year following initial admission. While this dissertation alone cannot conclusively delineate this relationship, previous evidence supports the hypothesis that the parents and caregivers of CMCs are more likely to experience suboptimal mental health (Dewan et al., 2023) that can have negative implications on the whole family (Barnert et al., 2017). The results of this work further motivate the need for additional investigations of the relationship between CMC outcomes and caregiver mental health.

5.3 Improving Care for CMCs in Alberta

Socioeconomic metrics such as education, income, and employment reinforce the importance of social determinants of health (Braveman et al., 2011). These factors are even more critical for CMCs (Barnert et al., 2017). Families struggle with the economic and psychosocial effects of caring for CMCs, which may have other adverse effects on their medically complex child (García-Altés et al., 2018) that in turn cause more stress for the family (Barnert et al., 2017). The analyses and commentary within this dissertation therefore make considerations beyond health care alone. The importance of socioeconomic factors supports the need for a holistic policy perspective that goes beyond health care resources to include education, employment, and income support (Woodgate et al., 2015; Thomson et al., 2016).

Barnert et al. (2018) proposed a framework for conceptualizing a healthy life for a CMC. The framework consists of ten domains: basic needs, inclusive education, child social integration, child-health-related quality of life, long-term child and family self-sufficiency, family social integration, community system support, health care system support, patient-centred

medical homes, and family-centred care. The findings in this dissertation provide empirical evidence to support the importance of several of these domains, namely health care system support, long-term child and family self-sufficiency, patient-centred medical homes, and family-centred care. For example, the associations observed between residence rurality and ED use suggests that health care systems might not be accessible to CMCs and families living in rural areas. Further associations between ED visits in the long-term may indicate a lack of self-sufficiency for families with material and social deprivation. Alberta could consider various effective support programs for CMCs (Berry et al., 2015), including care planning, care coordination, risk assessments, financial support, and the treatment and management of specific health care problems.

5.4 Knowledge Translation

The findings in this dissertation will be disseminated through a number of means. The first is through the publication of each chapter in peer-reviewed journals read by academics and practitioners working with CMCs. Chapter 2 was published in the Journal of Child Health Care (Sidra et al., 2022). At the time of writing, Chapters 3–4 have been submitted to other journals.

These results will also be presented at conferences where academics and practitioners meet to share knowledge about children's health care, such as at regular webinars (hosted by the nonprofit organization Children’s Healthcare Canada) designed to share emerging information on children's health. Specific to Alberta, the author plans to raise awareness of these results within AHS through the Maternal Newborn Child and Youth Strategic Clinical Network, which is composed of clinicians, academics, and practitioners who care for or research children’s health and are an ideal audience for this work.

5.5 Strengths and Limitations

As with any research of this type, this dissertation has several strengths and limitations that should be considered together with its results and commentary.

5.5.1 Strengths

As a key strength, the analyses of Chapters 3–4 concern a population cohort of Alberta CMCs, which reduces complications related to the generalization of the results therein. MyChild identified CMCs from Alberta administrative health care data via a well-established definition of CMCs. Furthermore, the longitudinal nature of the response data (i.e., hospital days and ED visits) available in MyChild enabled longitudinal modelling. Our observation window of five years is the longest among any Canadian study in the existing literature.

The systematic review in Chapter 2 presented a thorough examination of the literature and identified important limitations and research gaps in cost-reporting for CMCs. Chapters 3–4 presented rigorous and complementary quantitative analyses that added substantially to knowledge of the Alberta CMC population specifically and integrated these findings into existing frameworks for enhancing future predictive modeling for this population. Methodologically, analyses considering both socioeconomic and clinical factors simultaneously are not readily available in the existing CMC literature. The submodel structure (i.e., the use of binary and conditional submodels) in both chapters accounted for the large prevalence of zero responses in the MyChild dataset. This problem, which likely arises in other research datasets and has the potential to invalidate statistical inference, does not appear to be considered or accounted for in other studies of CMCs. Chapters 3–4 are complementary in the sense that they identified correlates and predictors of resource use under inferential and predictive modelling

frameworks, respectively. The predictive model was better able to handle a large number of variables and could automatically account for complex interaction effects, while the inferential model provided broad results at the population level that were easier to interpret. Taken together, these models enabled more-thorough analyses and interpretations of relationships between clinical and socioeconomic predictors and the outcome variables.

5.5.2 Limitations

Outcomes for CMC families other than those included in census data and summarized at the DA level are not routinely collected in administrative health care data and require the design and implementation of a data collection strategy to incorporate into research. This limits the extent to which the present work can quantify, rule out, or comment on the effects of socioeconomic factors. The presented results do not account for other specific socioeconomic factors such as race or immigration status (beyond general proxies for socioeconomic status) that might have otherwise modified the effects observed here. Other factors important to health care resource use were also not considered because of limitations inherent to administrative data. For example, the functional needs of individual patients, which play a role in how and when they access health care resources, were not readily available and were not included in this study.

Another limitation is that only data on hospital days and ED visits were available for the five-year period. The inclusion of clinical and socioeconomic as time-varying over the same period would improve the rigour of the analyses. Some socioeconomic data at the DA level were also missing, likely due to Statistics Canada withholding information for sparsely populated areas. This limitation may have caused the study to underestimate the significance of the effect of living in rural or remote rural locations. While DAs are intended to be homogenous by design,

the use of DA-level variables may introduce bias that causes the effects and importances of these variables to be underestimated. As an important consideration, the availability of primary care services was not available in the dataset and thus may be a confounding factor especially with respect to socioeconomic covariates such as rurality (e.g., there are two large pediatric hospitals in the metropolitan cities of Edmonton and Calgary but not in rural areas). Lastly, interpretability of the results regarding maternal access to mental health services was limited due to the amount and presentation of available data (e.g., no information was available on the type of mental health services accessed and mental health services outside the hospital and ED were not included in the data).

From a clinical perspective, while hospitals and EDs are essential services for CMCs, other factors such as primary care, diagnostic testing, and home care access were outside the scope of this work. As Alberta has a publicly funded health system, the results here may generalize to other publicly funded systems (with consideration of limitations mentioned above) but possibly not to for-profit health systems such as those in the United States (e.g., due to access to privately funded health insurance, which was not considered in this dissertation).

5.6 Future Research

Future research could include more-detailed analyses of specific components comprising the factors studied in this dissertation. Specifically, research on CMC index admissions may provide insight into what makes initial LOS such an important predictor of hospital days and ED use. As the subgroup of CMCs with neurological conditions was more vulnerable than other CMCs, future research could explore how specific health and socioeconomic policies impact patients in this vulnerable subpopulation and their families. Exploring these relationships further

was not possible for this dissertation due to data availability but could feasibly be explored in future studies with a concerted effort to preemptively gather this data.

Further family-oriented research would augment the results presented in this work. For example, this dissertation identifies CMC families living in urban, rural or remote rural locations as important subpopulations that require more resources than their metropolitan counterparts. Analysis of how the roles of communities, extended families, and health care professionals differ between metropolitan and nonmetropolitan areas could inform the design of programs and policies aiming to optimize use of the EDs for this population. Similarly, to improve patient and provider experiences, future studies could examine (1) the extent to which health care providers working in urban, rural, and remote rural locations are equipped and supported to deal with medically complex children and (2) how the observed more-frequent ED visits by CMCs affect providers, CMCs, and their families.

This dissertation also identified that chronic medications are an important predictor of hospital days in the long term and posited that CMCs may be at higher risk of overmedicalization, consistent with Pordes et al. (2020) and Marty et al. (2019). Overmedicalization can result in unnecessary and potentially harmful care for the child. Diagnosing overmedicalization requires a consideration of not only medication use, but the complex interplay between caregiver, health care provider, and the child (Portes et al., 2020). Overmedicalization is a complex issue that cannot be further explored in this work, but it is crucial that future research considers this important issue.

From a methodological perspective, as discussed in Chapter 4, exploring mechanisms to include patient-level socioeconomic data in EHRs would bolster the potential of future work, especially that using machine learning models for high-dimensional data, and holds important

implications for the development of clinical tools and policy. Since variability in hospital admission and readmission affects the clinical quality of care for CMCs (Ralston et al., 2015), improvements in the accuracy of predictive models in this field can support the development of standardized practices towards optimizing the use of health resources and improving outcomes for CMCs and their families. For example, Ralston et al. (2015) found considerable variation in the administration of diagnostic testing (e.g., magnetic resonance imaging and electroencephalograms) and neurological examination rates for CMCs and hypothesized that the variation may be due in-part to uncertainty in medical evidence. The use of predictive modelling has the potential to provide evidence and support medical consensus, for example, for appropriate administration of diagnostic testing.

5.7 Conclusion

This dissertation and the results therein reinforce the importance of holistic approaches that consider clinical and socioeconomic factors in the pursuit of improved care for CMCs. The results identify characteristics of CMC subpopulations (e.g., the presence of neurological conditions, a long initial LOS) that require more resources and may have unmet care needs. These subpopulations may be an ideal target for care coordination programs and other social support. Ultimately, access to high-quality health care and social support programs are necessary to improve the health and wellbeing of CMCs and their families.

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Appendix A. Search Strategy for Reported Costs of Children with Medical Complexity

The following databases were searched on April 23, 2019:

- Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily <1946 to April 22, 2019>
- PubMed, 1946-Current
- Ovid EMBASE, 1974-April 22, 2019
- EBSCO CINAHL Plus with Full-text,1937-Current
- Wiley Cochrane Library, Inception to Current

Search results were limited to English language publications. No date or format limits were applied.

4395 items were retrieved in total. 3205 items remained after duplicates were removed.

Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily <1946 to April 23, 2019>

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1 children with medical complexity.ti,ab,kf. (178)

2 multiple chronic conditions/ or ((Multiple or concurrent or co-occur* or cooccur* or simultaneous or complex*) adj3 (condition* or disease* or illness*)).ti,ab,kf. or (medical* adj2 complex*).ti,ab,kf. (75645)

3 exp comorbidity/ or (co-morbidit* or comorbidit* or multimorbidit* or multi-morbidit* or multiple morbidit*).ti,ab,kf. (195241)

4 Tertiary Health care/ or life support care/ or exp respiration, artificial/ or biomedical technology/ or exp Biomedical Enhancement/ or exp parenteral nutrition/ or enteral nutrition/ or long-term care/ or (ventilator* or parenteral* or enteral* or dialysis or he?modialysis or incubator* or long term care or ((continuity or transition*) adj2 care) or (technolog* adj2 depend*) or (special adj3 need*) or ((lifelong or multidisciplin*) adj3 (manage* or care)) or complex* or life limiting or life threatening or severe or severity or poorly controlled or debilitat* or disab*).ti,ab,kf. (3135256)

5 3 and 4 (59632)

6 2 or 5 (132949)

7 exp Economics/ (576387)

8 (economic* or cost* or expenditure* or spending or financ*).m_titl. (182268)

9 (((illness* or disease* or condition* or medical or health care or health care or hospital* or system* or treat* or therap* or manag* or care or impact*) adj3 (cost* or economic* or spending or financ*)) or ((economic* or financial*) adj3 burden*) or direct cost* or indirect cost* or cost driv*).ab. or (economic* or cost* or expenditure* or spending or financ*).ab. /freq=2 or (economic* and (cost* or expenditure* or spending or financ*)).ab. or (cost* and (economic* or expenditure* or financ*)).ab. or (expenditure* and (economic* or cost* or financ*)).ab. or (financ* and (economic* or cost* or expenditure*)).ab. (328947)

10 ec.fs. (405072)

11 7 or 8 or 9 or 10 (923377)

12 6 and 11 (8341)

13 exp Intensive Care Units, Pediatric/ or exp Pediatrics/ or exp Child Care/ or exp Child, Institutionalized/ or exp Child Mortality/ or exp Child Health/ or exp Child Health Services/ or exp Child, Hospitalized/ or exp Adolescent, Hospitalized/ or exp Adolescent Health Services/ or exp Adolescent Health/ or exp Adolescent, Institutionalized/ or exp Adolescent Medicine/ or exp Infant, Premature, Diseases/ or exp Intensive Care, Neonatal/ or exp Intensive Care Units, Neonatal/ or exp Infant, Premature/ or exp Infant, Newborn, Diseases/ or exp Infant Care/ or exp Infant, Low Birth Weight/ or exp Infant, Small for Gestational Age/ (341482)

14 exp child/ or exp infant/ or exp adolescent/ or ((child or children or childhood or infan* or neonat* or newborn* or baby or babies or toddler* or preschool* or pre-school* or school age or school child* or boy or boys or girl* or kindergarten* or adolescen* or teen* or youth* or juvenile or pediatric* or paediatric* or nicu or picu) not baby boom*).ti,ab,kf. (4044654)

15 13 or 14 (4086890)

16 12 and 15 (1710)

17 1 and 11 (49)

18 16 or 17 (1710)

19 limit 18 to english language (1624)

20 remove duplicates from 19 (1619)

Embase <1974 to 2019 April 23>

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1 children with medical complexity.ti,ab,kw. (203)

2 multiple chronic conditions/ or (complex chronic condition* or multiple chronic condition*).ti,ab,kw. (3111)

3 chronic disease/ or ((Multiple or concurrent or co-occur* or cooccur*or simultaneous or complex*) adj3 (condition* or disease* or illness*)).ti,ab,kw. or exp comorbidity/ or (co-morbidit* or comorbidit* or multimorbidit* or multi-morbidit* or multiple morbidit*).ti,ab,kw. (587477)

4 exp tertiary health care/ or long term care/ or exp artificial ventilation/ or medical technology/ or exp parenteral nutrition/ or enteric feeding/ or transition to adult care/ or transitional care/ or (ventilator* or parenteral* or enteral* or dialysis or he?modialysis or incubator* or long term care or ((continuity or transition*) adj2 care) or (technolog* adj2 depend*) or (special adj3 need*) or ((lifelong or multidisciplin*) adj3 (manage* or care)) or complex* or life limiting or life threatening or severe or severity or poorly controlled or debilitat* or disab*).ti,ab,kw. (4237910)

5 3 and 4 (200991)

6 2 or 5 (202725)

7 *economic aspect/ or exp *health economics/ or exp *"health care cost"/ or exp *economic evaluation/ (250393)

8 (economic* or cost* or expenditure* or spending or financ*).ti. (228133)

9 (((illness* or disease* or condition* or medical or health care or health care or hospital* or system* or treat* or therap* or manag* or care or impact*) adj3 (cost* or economic* or spending or financ*)) or ((economic or financial) adj3 burden*) or direct cost* or indirect cost* or cost driv*).ab. or (economic* or cost* or expenditure* or spending or financ*).ab. /freq=2 or

(economic* and (cost* or expenditure* or spending or financ*)).ab. or (cost* and (economic* or expenditure* or financ*)).ab. or (expenditure* and (economic* or cost* or financ*)).ab. or (fianc* and (economic* or cost* or expenditure*)).ab. (456469)

10 7 or 8 or 9 (679898)

11 6 and 10 (11871)

12 juvenile/ or exp adolescent/ or exp child/ or exp infant/ or exp childhood disease/ or exp infant disease/ or exp adolescent disease/ or child care/ or infant care/ or child health/ or exp child health care/ or child health insurance/ or exp newborn intensive care/ or exp neonatal intensive care unit/ or exp pediatric intensive care unit/ or prematurity/ or exp low birth weight/ (4093237)

13 ((child or children or childhood or infan* or neonat* or newborn* or baby or babies or toddler* or preschool* or pre-school* or school age or school child* or boy or boys or girl* or kindergarten* or adolescen* or teen* or youth* or juvenile or pediatric* or paediatric* or nicu or picu) not baby boom*).ti,ab,kw. (2703720)

14 12 or 13 (4691597)

15 11 and 14 (2008)

16 1 and 10 (49)

17 15 or 16 (2028)

18 limit 17 to english language (1945)

19 remove duplicates from 18 (1920)

CINAHL Plus with Full-text (433 items retrieved)

S1 "children with medical complexity"

S2 (Multiple or concurrent or simultaneous or "co-occur*" or cooccur*) w2 (condition* or disease* or illness*) or medical* n2 complex*

S3 ((MH "Tertiary Health Care") OR (MH "Life Support Care") OR (MH "Respiration, Artificial+") OR (MH "Biomedical Enhancement") OR (MH "Parenteral Nutrition+") OR (MH "Enteral Nutrition") OR (MH "Continuity of Patient Care") OR (MH "Transitional Care") OR (MH "Long Term Care")) OR (ventilator* or incubator* or parenteral* or enteral* or dialysis or hemodialysis or haemodialysis or "long term care" OR (continuity or transition*) w2 care or technolog* n2 depend*) OR special w2 need* or (lifelong or multidisciplin*) w2 (manage* or care) or complex* or "life limiting" or "life threatening" or severe or severity or "poorly controlled" or debilitat* or disab*)

S4 (MH "Comorbidity") OR "co-morbidit*" or comorbidit* or multimorbidit* or "multi-morbidit*" or "multiple morbidit*"

S5 S3 AND S4

S6 S2 OR S5

S7 (MH "Economics") OR (MH "Costs and Cost Analysis") OR (MH "Health Care Costs+") OR (MH "Economic Aspects of Illness") OR (MH "Health Resource Utilization/EC") OR TI(economic* or cost* or expenditure* or spending or financ*)

S8 AB ((illness* or disease* or condition* or medical or health care or health care or hospital* or system* or treat* or therap* or manag* or care or impact*) n3 (cost* or economic* or spending or financ*) OR (economic* or financial*) n3 burden* or "direct cost*" or "indirect

cost*" or "cost driv*" OR (economic* and (cost* or expenditure* or spending or financ*)) OR (cost* and (economic* or expenditure* or financ*)) OR (expenditure* and (economic* or cost* or financ*)) OR (financ* and (economic* or cost* or expenditure*)))

S9 S7 OR S8

S10 S6 AND S9

S11 (child or children or childhood or infan* or neonat* or newborn* or baby or babies or toddler* or preschool* or "pre-school*" or "school age" or boy or boys or girl* or kindergarten* or adolescen* or teen* or youth* or pediatric* or paediatric* or nicu or picu or juvenile*) not "baby boom*"

S12 S10 AND S11

S13 S1 AND S7

S14 S12 OR S13

Limit: English language

Cochrane Library (Advanced Search/Search Manager; 109 items retrieved)

#1 ("children with medical complexity"):ti,ab,kw

#2 (((Multiple or concurrent or simultaneous or "co-occur*" or cooccur*) near/2 (condition* or disease* or illness*)) or (medical* near/2 complex*)):ti,ab,kw

#3 (ventilator* or incubator* or parenteral* or enteral* or dialysis or hemodialysis or haemodialysis or "long term care" OR ((continuity or transition*) near/2 care) or (technolog* near/2 depend*) OR (special near/2 need*) or (lifelong or multidisciplin*) near/2 (manage* or

care) or complex* or "life limiting" or "life threatening" or severe or severity or "poorly controlled" or debilitat* or disab*):ti,ab,kw

#4 ("co-morbidit*" or comorbidit* or multimorbidit* or "multi-morbidit*" or "multiple morbidit*"):ti,ab,kw

#5 #3 AND #4

#6 #2 OR #5

#7 (economic* or cost* or expenditure* or spending or financ*):ti

#8 ((illness* or disease* or condition* or medical or health care or health care or hospital* or system* or treat* or therap* or manag* or care or impact*) near/3 (cost* or economic* or spending or financ*) OR (economic* or financial*) near/3 burden* or "direct cost*" or "indirect cost*" or "cost driv*" OR (economic* and (cost* or expenditure* or spending or financ*)) OR (cost* and (economic* or expenditure* or financ*)) OR (expenditure* and (economic* or cost* or financ*)) OR (financ* and (economic* or cost* or expenditure*))):ab

#9 #7 OR #8

#10 #6 AND #9

#11 ((child or children or childhood or infan* or neonat* or newborn* or baby or babies or toddler* or preschool* or "pre-school*" or "school age" or boy or boys or girl* or kindergarten* or adolescen* or teen* or youth* or pediatric* or paediatric* or nicu or picu or juvenile*) not "baby boom*"):ti,ab

#12 #10 AND #11

#13 #1 AND #9

#14 #12 OR #13

PubMed (314 items retrieved)

#1 "children with medical complexity" [Text Word]

#2 ("Multiple chronic conditions" [MeSH Terms] OR "multiple conditions" [Text Word] OR "concurrent conditions" [Text Word] OR "simultaneous conditions" [Text Word] OR "complex conditions" [Text Word] OR "multiple illnesses" [Text Word] OR "concurrent illnesses" [Text Word] OR "simultaneous illnesses" [Text Word] OR "complex illnesses" [Text Word] OR "multiple diseases" [Text Word] OR "concurrent diseases" [Text Word] OR "simultaneous diseases" [Text Word] OR "complex diseases" [Text Word] OR "complex chronic conditions" [Text Word] OR "multiple chronic illnesses" [Text Word] OR "concurrent chronic illnesses" [Text Word] OR "simultaneous chronic illnesses" [Text Word] OR "simultaneous chronic conditions" [Text Word]) OR co-occuring [Text Word] OR cooccuring [Text Word] OR "co-occurence" [Text Word] OR cooccurence [Text Word] OR "co-occur" [Text Word] OR coocur [Text Word])

#3 "comorbidity"[MeSH Terms] OR ("co-morbidity"[Text Word] OR comorbidity[Text Word] OR multimorbidity[Text Word] OR "multi-morbidity"[Text Word] OR "multiple morbidity"[Text Word] OR "co-morbidities"[Text Word] OR comorbidities[Text Word] OR multimorbidities[Text Word] OR "multi-morbidities"[Text Word] OR "multiple morbidities"[Text Word]) AND ("tertiary health care"[MeSH Terms] OR "life support care"[MeSH Terms] OR "respiration, artificial"[MeSH Terms] OR "biomedical technology"[mesh:noexp] OR "biomedical enhancement"[MeSH Terms] OR "parenteral nutrition"[MeSH Terms] OR "enteral nutrition"[MeSH Terms] OR "long-term care"[MeSH Terms] OR ventilator[Text Word] OR ventilators[Text Word] OR parenteral[Text Word] OR

enteral[Text Word] OR dialysis[Text Word] OR hemodialysis[Text Word] OR haemodialysis[Text Word] OR "long-term care"[Text Word] OR "continuity of care"[Text Word] OR "transitional care"[Text Word] OR "technology dependent"[Text Word] OR "special needs"[Text Word] OR "lifelong management"[Text Word] OR "lifelong care"[Text Word] OR "multidisciplinary management"[Text Word] OR "multidisciplinary care"[Text Word] OR "life limiting"[Text Word] OR "life threatening"[Text Word] OR severe[Text Word] OR severity[Text Word] OR "poorly controlled"[Text Word] OR debilitating[Text Word] OR disabling[Text Word])

#4 #2 OR #3

#5 Economics [MeSH Terms] OR economic [ti] OR economics [ti] OR cost [ti] OR costs [ti] or expenditure [ti] OR expenditures [ti] OR spending [ti] OR financial [ti] OR finance [ti] OR financing [ti] OR financed [ti]

#6 ((economic [Title/Abstract] OR economics [Title/Abstract] OR cost [Title/Abstract] OR costs [Title/Abstract] or expenditure [Title/Abstract] OR expenditures [Title/Abstract] OR spending [Title/Abstract] OR financial [Title/Abstract] OR finance [Title/Abstract] OR financing [Title/Abstract] OR financed [Title/Abstract]) AND (illness[Title/Abstract] OR disease[Title/Abstract] OR condition[Title/Abstract] OR "health care"[Title/Abstract] OR health care[Title/Abstract] OR hospital[Title/Abstract] OR hospitalization[Title/Abstract] OR hospitalisation[Title/Abstract] OR hospitalized[Title/Abstract] OR hospitalised[Title/Abstract] OR treat[Title/Abstract] OR treatment[Title/Abstract] OR therapy[Title/Abstract] OR therapies[Title/Abstract] OR management[Title/Abstract] OR care[Title/Abstract] OR impact[Title/Abstract] OR impacts[Title/Abstract] OR impacted[Title/Abstract] OR impacting[Title/Abstract]))

#7 #5 OR #6

#8 #4 AND #7

#9 ("intensive care units, pediatric"[MeSH Terms] OR "pediatrics"[MeSH Terms] OR "child care"[MeSH Terms] OR "child, institutionalized"[MeSH Terms] OR "child mortality"[MeSH Terms] OR "child health"[MeSH Terms] OR "child health services"[MeSH Terms] OR "child, hospitalized"[MeSH Terms] OR "adolescent, hospitalized"[MeSH Terms] OR "adolescent health services"[MeSH Terms] OR "adolescent health"[MeSH Terms] OR "adolescent, institutionalized"[MeSH Terms] OR "adolescent medicine"[MeSH Terms] OR "infant, premature, diseases"[MeSH Terms] AND "intensive care, neonatal"[MeSH Terms] OR "intensive care units, neonatal"[MeSH Terms] OR "infant, premature"[MeSH Terms] OR "infant, newborn, diseases"[MeSH Terms] OR "infant care"[MeSH Terms] OR "infant, low birth weight"[MeSH Terms] OR "infant, small for gestational age"[MeSH Terms] OR "child"[MeSH Terms] OR "infant"[MeSH Terms] OR "adolescent"[MeSH Terms] OR child[TITLE/ABSTRACT] OR children[TITLE/ABSTRACT] OR childhood[TITLE/ABSTRACT] OR infant[TITLE/ABSTRACT] OR neonate[TITLE/ABSTRACT] OR neonates[TITLE/ABSTRACT] OR neonatal[TITLE/ABSTRACT] OR newb[TITLE/ABSTRACT] AND ORn[TITLE/ABSTRACT] OR newb[TITLE/ABSTRACT] AND ORns[TITLE/ABSTRACT] OR baby[TITLE/ABSTRACT] OR babies[TITLE/ABSTRACT] OR toddler[TITLE/ABSTRACT] OR toddlers[TITLE/ABSTRACT] OR preschool[TITLE/ABSTRACT] OR "pre-school"[TITLE/ABSTRACT] OR preschooler[TITLE/ABSTRACT] OR preschoolers[TITLE/ABSTRACT] OR "pre-schooler"[TITLE/ABSTRACT] OR "pre-schoolers"[TITLE/ABSTRACT] OR "school age"[TITLE/ABSTRACT] OR "school

child"[TITLE/ABSTRACT] OR "school children"[TITLE/ABSTRACT] OR boy[TITLE/ABSTRACT] OR boys[TITLE/ABSTRACT] OR girl[TITLE/ABSTRACT] OR girl[TITLE/ABSTRACT] OR kindergarten[TITLE/ABSTRACT] OR kindergartener[TITLE/ABSTRACT] OR kindergarteners[TITLE/ABSTRACT] OR adolescent[TITLE/ABSTRACT] OR adolescents[TITLE/ABSTRACT] OR adolescence[TITLE/ABSTRACT] OR teen[TITLE/ABSTRACT] OR teens[TITLE/ABSTRACT] OR teenager[TITLE/ABSTRACT] OR teenagers[TITLE/ABSTRACT] OR youth[TITLE/ABSTRACT] OR youths[TITLE/ABSTRACT] OR pediatric[TITLE/ABSTRACT] OR pediatrics[TITLE/ABSTRACT] OR paediatric[TITLE/ABSTRACT] OR paediatrics[TITLE/ABSTRACT] OR nicu[TITLE/ABSTRACT] OR picu[TITLE/ABSTRACT] OR juvenile[TITLE/ABSTRACT] OR juveniles[TITLE/ABSTRACT])

#10 #8 AND #9

#11 #1 AND #7

#12 #10 OR #11

#13 "baby boomer" [Text Word] OR "baby boomers" [Text Word] OR "baby boom" [Text Word]

#14 #12 NOT #13

Limit to English language

Appendix B – Supplementary Tables and Figures for Chapter 3

Table B3.5 Association Between Initial Length of Stay and the Odds of Having Zero Hospital Days and ED Visits by Clinical Group

Clinical category[a]	**Estimate (SE)**	**Relative difference (SE)**[b]	***P* value**
Hospital days			
Cardiology	−0.109 (0.034)	0.897 (0.031)	.007
Congenital/genetic	0.269 (0.064)	1.308 (0.084)	<.001
Gastrointestinal	−0.011 (0.082)	0.989 (0.081)	.90
Hematology/immunodeficiency	0.101 (0.126)	1.106 (0.140)	.85
Malignancy	−0.501 (0.053)	0.606 (0.032)	<.001
Metabolic	−0.305 (0.079)	0.737 (0.058)	<.001
Neurology	−0.113 (0.035)	0.894 (0.032)	.007
Renal	−0.236 (0.076)	0.790 (0.060)	.007
Respiratory	−0.329 (0.051)	0.719 (0.037)	<.001
ED visits			
Cardiology	−0.197 (0.040)	0.821 (0.033)	<.001
Congenital/genetic	−0.431 (0.072)	0.650 (0.047)	<.001
Gastrointestinal	0.075 (0.097)	1.078 (0.104)	.87
Hematology/immunodeficiency	−0.108 (0.157)	0.898 (0.141)	.87
Malignancy	−0.365 (0.061)	0.694 (0.042)	<.001
Metabolic	−0.169 (0.093)	0.844 (0.078)	.21

Neurology	−0.188 (0.042)	0.829 (0.035)	<.001
Renal	−0.655 (0.095)	0.520 (0.049)	<.001
Respiratory	−0.346 (0.061)	0.708 (0.043)	<.001

Abbreviations: ED, emergency department; SE, standard error.

Change in initial length of stay: All estimates correspond to a 10-fold increase in initial length of stay.

[a] Considers a CMC in exactly one clinical category.

[a] Relative difference is the natural antilog of the estimate.

Table B3.6 Comparisons of the Odds of Having Zero Hospital Days and ED Visits Between Age Groups

	Year 1			**Year 5**		
	Estimate (SE)	**Relative difference (SE)[a]**	***P* value**	**Estimate (SE)**	**Relative difference (SE)[a]**	***P* value**
Hospital days						
<1 vs						
1–4	0.516 (0.077)	1.676 (0.129)	<.001	0.138 (0.073)	1.148 (0.084)	.41
5–9	0.649 (0.101)	1.913 (0.194)	<.001	0.151 (0.093)	1.163 (0.108)	.52
10–13	0.187 (0.128)	1.205 (0.155)	.44	−0.126 (0.122)	0.882 (0.108)	>.99
14–18	0.319 (0.118)	1.375 (0.163)	.04	−0.068 (0.112)	0.934 (0.104)	>.99
1–4 vs						
5–9	0.133 (0.103)	1.142 (0.118)	.44	0.013 (0.094)	1.014 (0.095)	>.99
10–13	−0.330 (0.127)	0.719 (0.080)	.05	−0.263 (0.120)	0.768 (0.092)	.24
14–18	−0.198 (0.117)	0.821 (0.096)	.37	−0.206 (0.109)	0.814 (0.089)	.41
5–9 vs						
10–13	−0.462 (0.122)	0.630 (0.077)	.001	−0.277 (0.110)	0.758 (0.083)	.12
14–18	−0.330 (0.112)	0.719 (0.091)	.02	−0.219 (0.099)	0.803 (0.079)	.24
10–13 vs 14–18	0.132 (0.106)	1.141 (0.121)	.44	0.058 (0.093)	1.060 (0.098)	>.99
ED visits						
<1 vs						
1–4	0.074 (0.046)	1.077 (0.049)	.21	0.015 (0.040)	1.015 (0.040)	>.99
5–9	0.471 (0.060)	1.602 (0.097)	<.001	0.318 (0.053)	1.374 (0.073)	<.001
10–13	0.472 (0.078)	1.603 (0.125)	<.001	0.239 (0.071)	1.270 (0.090)	.004
14–18	0.276 (0.068)	1.318 (0.090)	<.001	−0.008 (0.064)	0.992 (0.064)	>.99
1–4 vs						
5–9	0.397 (0.063)	1.488 (0.093)	<.001	0.302 (0.054)	1.353 (0.073)	<.001
10–13	0.398 (0.078)	1.489 (0.117)	<.001	0.224 (0.070)	1.251 (0.087)	.007
14–18	0.202 (0.069)	1.224 (0.084)	.01	−0.023 (0.064)	0.978 (0.062)	>.99
5–9 vs						

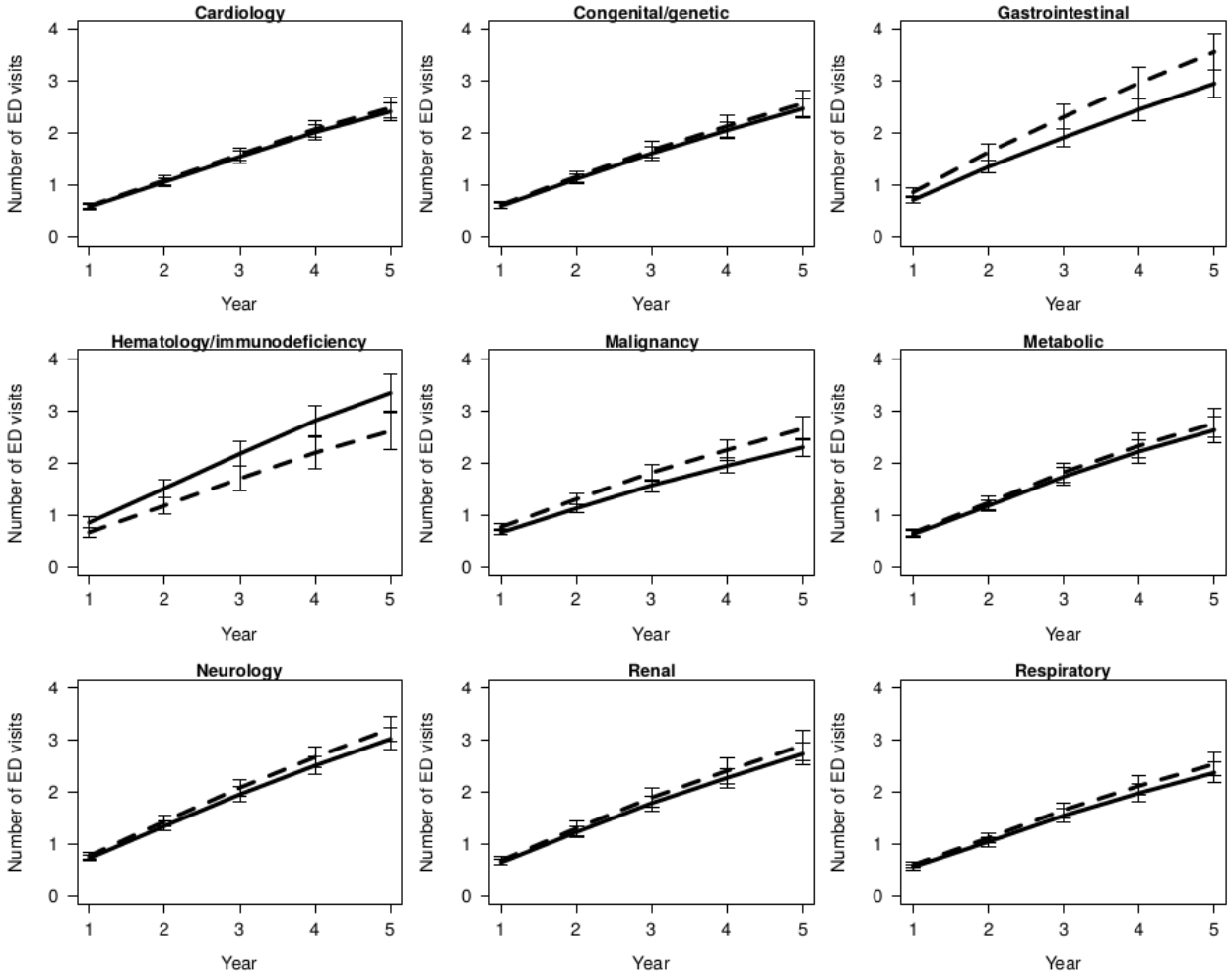
10–13	0.001 (0.075)	1.001 (0.075)	>.99	−0.078 (0.063)	0.925 (0.059)	.87
14–18	−0.195 (0.065)	0.823 (0.054)	.01	−0.325 (0.057)	0.722 (0.041)	<.001
10–13 vs 14–18	−0.196 (0.063)	0.822 (0.052)	.01	−0.247 (0.052)	0.781 (0.041)	<.001

Abbreviations: ED, emergency department; SE, standard error.

[a] Relative difference is the antilog of the presented estimate.

[b] Comparisons for year 5 are the same as those for year 1 and are omitted.

Figure B3.3. Conditional ED Visit Trajectories by Clinical Group and Initial LOS (solid and dotted lines for 3 and 12 days, respectively). Estimates correspond to a 14–19 year-old, male patient in a metropolitan area with a 3-day initial LOS; no TA, chronic medications, or readmissions; and deprivation factor scores of 0.



Appendix C – Supplementary Tables and Figures for Chapter 4

Table C4.1 Model Performance on the Training and Testing sets: AUC and R2 for the Binary and Conditional Submodels, Respectively

Year	Outcome	Binary model performance, AUC		Conditional model performance, R^2	
		Training	**Testing**	**Training**	**Testing**
Year 1	Hospital days	0.75	0.71	0.29	0.18
	ED visits	0.70	0.61	0.24	0.19
Year 5	Hospital days	0.73	0.72	0.13	0.10
	ED visits	0.78	0.62	0.14	0.10

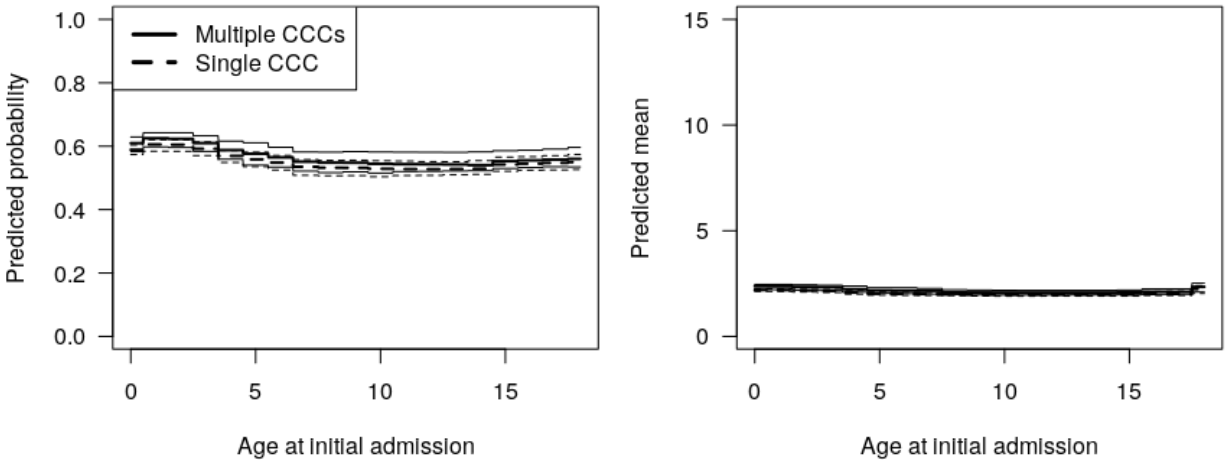
Abbreviations: AUC, area under the ROC curve; ED, emergency department; ROC, receiver–operator characteristic.

Table C4.2 Estimated Mean Conditional Number of ED Visits (95% Confidence Interval) by AHS Zone and Residence Rurality

AHS zone	Year	Residence rurality			
		Metropolitan	Urban	Rural	Rural remote
Calgary	1	1.87 (1.83, 1.93)	-	2.37 (2.22, 2.52)	-
	5	3.80 (3.71, 3.91)	-	5.74 (5.42, 5.99)	-
Central	1	2.03 (1.90, 2.13)	-	2.57 (2.40, 2.75)	-
	5	3.85 (3.74, 4.08)	-	5.83 (5.50, 6.17)	-
Edmonton	1	1.96 (1.88, 2.03)	-	-	-
	5	3.86 (3.75, 4.01)	-	-	-
North	1	-	2.33 (2.20, 2.49)	3.05 (2.84, 3.20)	3.11 (2.85, 3.31)
	5	-	4.70 (4.40, 5.01)	7.27 (6.77, 7.73)	7.40 (6.77, 8.00)
South	1	-	1.96 (1.87, 2.09)	2.53 (2.34, 2.67)	-
	5	-	3.95 (3.77, 4.25)	6.00 (5.63, 6.53)	-

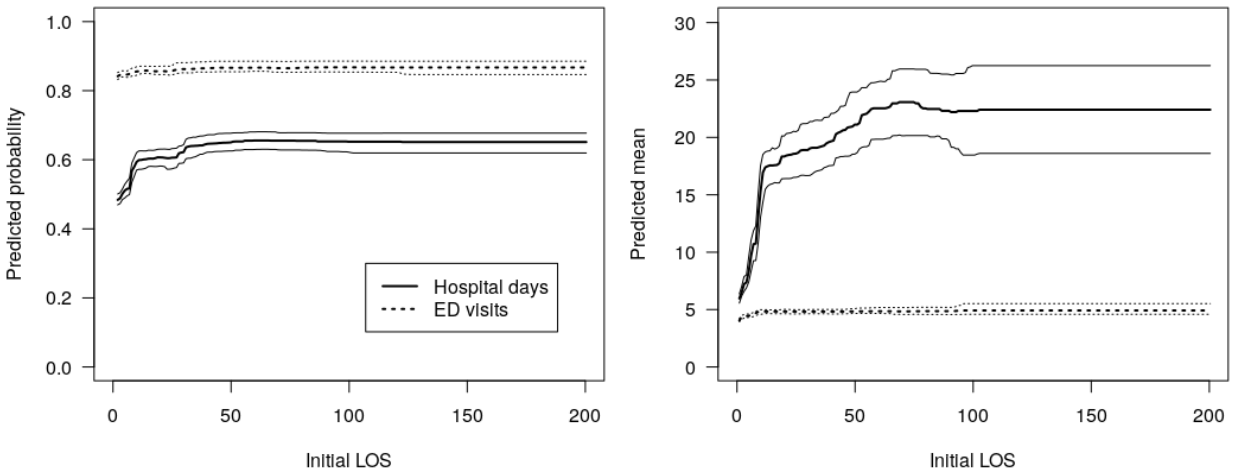
Abbreviation: AHS, Alberta Health Services; ED, emergency department.
Note: A - indicates estimates omitted due to the composition of a zone (e.g., no remote rural areas exist in the Calgary Zone) or the small number of CMCs in the cohort (i.e., <50 in urban Central, rural Edmonton, and metropolitan North).

Figure C4.1 Marginal associations (and 95% confidence intervals) for age at initial admission in the binary (left) and conditional (right) submodels for ED visits among CMCs with a single CCC (dotted lines) or multiple CCCs (solid lines) within the year following initial discharge.



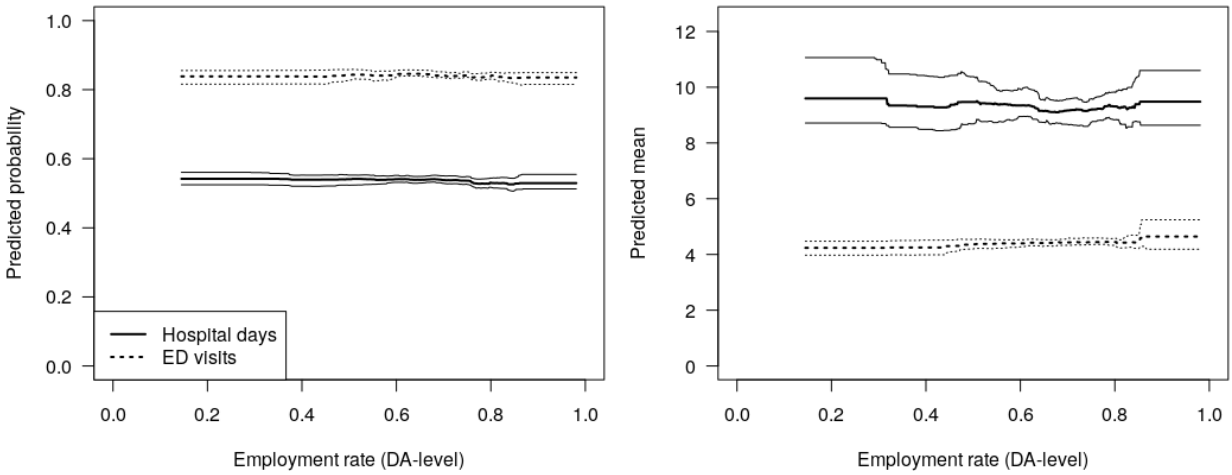
Abbreviations: CCC, complex chronic condition; CMC, child with medical complexity; ED emergency department.

Figure C4.2 Marginal associations (and 95% confidence intervals) for initial LOS in the binary (left) and conditional (right) submodels for hospital days (solid lines) and ED visits (dotted lines) within the 5 years following initial discharge.



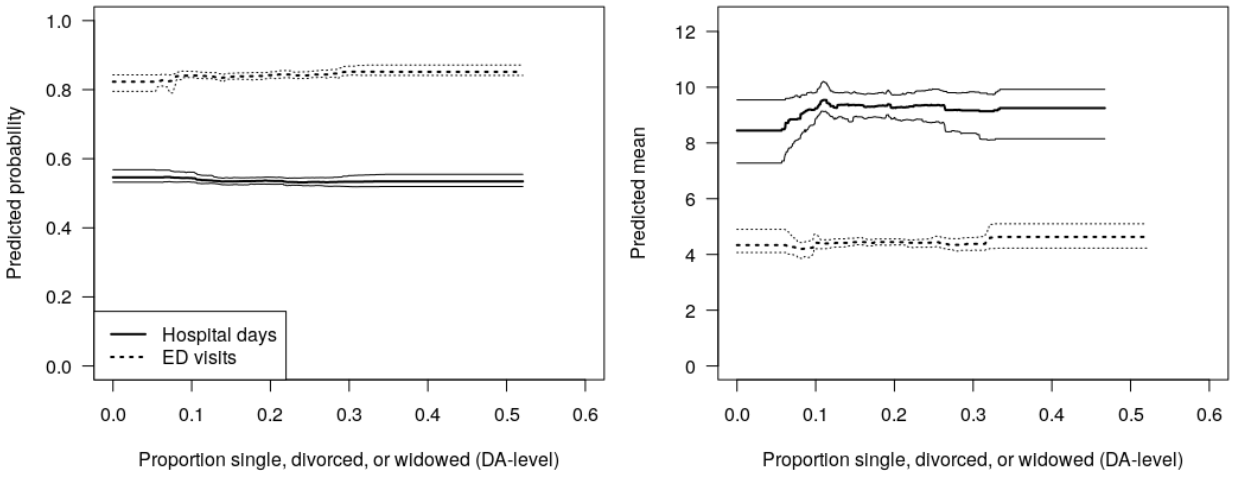
Abbreviations: ED, emergency department; LOS, length of stay.

Figure C4.3 Marginal associations (and 95% confidence intervals) for employment rate (measured at the DA level) in the binary (left) and conditional (right) submodels for hospital days (solid lines) and ED visits (dotted lines) within the 5 years following initial discharge.



Abbreviations: DA, dissemination area; ED, emergency department.

Figure C4.4 Marginal associations (and 95% confidence intervals) for the proportion of single, divorced, or widowed individuals (measured at the DA level) in the binary (left) and conditional (right) submodels for hospital days (solid lines) and ED visits (dotted lines) within the 5 years following initial discharge.



Abbreviations: DA, dissemination area; ED, emergency department.