Travel distance and patterns of health care utilization among children with medical complexity in Quebec: a population-based cohort study

Sara Long-Gagné MD FRCPC
Department of Epidemiology, Biostatistics and Occupational Health
McGill University

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Abstract

Title: Travel distance and patterns of health care utilization among children with medical complexity in Quebec: a population-based cohort study

Introduction: Children with medical complexity (CMC) represent a patient population with a wide range of medical conditions. CMC have high care needs and therefore are also high users of health care services, some that could potentially be reduced with optimal outpatient care such as readmissions to hospital. Although primary care providers are essential in the care of CMC, they cannot realistically provide the full range of care required by CMC without the support of a multidisciplinary team of specialized healthcare professionals. Currently, the majority of specialized services are provided within pediatric tertiary care centres.

Objectives and methodology: For children suffering from any chronic health problems, a travel distance of more than 80 kilometres from hospital has been shown to negatively affect family unit dynamics and increase family anxiety when caring for their child at home due to the disruption in routine associated with such travels for which families may have to dedicate a whole day or overnight stay in order to reach their destination. We expected that difficulties associated with prolonged travels would limit specialty follow-up in the early period following a hospital discharge for CMC living at a driving distance of 80 kilometres or more to a tertiary pediatric centre compared to those living closer. Considering a fair proportion of early issues that may arise in the early post-discharge period could be addressed with adequate outpatient expert care, CMC living farther would be at increased risk of readmission within 30 days. Our primary objective was to look at the association between driving distance to the closest pediatric tertiary care centre (less than 80 kilometres compared to 80 kilometres or more) and the pattern of health service utilization, including time to readmission within 30 days following an initial

hospital admission, in children aged 2 to 18 years with different levels of medical complexity from the province of Quebec. We used a population-based cohort design with multiple datasets from the *Régie de l'assurance maladie du Québec* and a Cox proportional hazard model to determine associations with our primary outcome.

Results: Overall, we found that CMC in Quebec represented 2.2% of the total population of children and that 24% of these children lived at a driving distance of 80 kilometres or more from a pediatric tertiary care centre. Compared to those living at a driving distance of less than 80 kilometres, CMC located at a driving distance of 80 kilometres or more had less outpatient visits to family physicians, pediatricians or specialists, but more emergency department visits and repeated hospital admissions—yet, no association was found for the risk of readmission within 30 days of an initial hospitalization.

Conclusion: Although driving distance was not associated with the risk of readmission within 30 days, we found that CMC living at a distance of 80 kilometres or more to a pediatric tertiary care centre utilized an increased number of unplanned/unscheduled services such as emergency department visits and repeated hospital admissions compared to those living at a driving distance less than 80 kilometres from a pediatric tertiary care centres. Moreover, a third of all CMC had no primary care provider. As these differences are unlikely to be solely explained by geographical barriers such as driving distance, the next steps would be to further understand the facilitators and barriers for families and for primary care caring for CMC, in order to develop programs and infrastructure that may reduce readmissions as well as improve the quality of care for CMC.

Résumé

Titre: L'influence de la distance à parcourir par rapport aux variations d'utilisation des services de soins de santé chez les enfants avec soins complexes au Québec, Canada: une étude de cohorte basée sur la population

Introduction: Les enfants avec soins complexes (ESC) font partie d'une population très hétérogène qui éprouve différents problèmes de santé, telles que des conditions complexes et chroniques(CCC). De plus, ces enfants ont recours à plusieurs services de santé pour combler leurs besoins, cependant certains services telles les réadmissions à l'hôpital pourraient potentiellement être diminuées avec de bons soins acheminés en clinique externe. En effet, les fournisseurs de soins primaires sont essentiels au bien-être des ESC mais doivent pouvoir compter sur l'apport d'une équipe multidisciplinaire constituée de professionnels de la santé spécialisés afin de répondre efficacement à la vaste étendue des besoins requis par la complexité médicale spécifique aux ESC. Il est également important de prendre en compte que la majorité des services de soins spécialisés se trouvent exclusivement au sein de centres pédiatriques tertiaires situés dans de grands centres urbains.

Objectifs et méthodologie: La situation d'enfants souffrant de maladies chroniques et devant parcourir une distance de plus de 80 kilomètres pour accéder à un centre hospitalier est notamment associée à un effet négatif tant sur la cellule familiale que sur sa dynamique. En effet, pour se rendre à destination ces familles doivent souvent consacrer une journée entière ou une nuitée, ce qui engendre un bris des activités routinières. Nous posons comme hypothèse que les difficultés, associées à un trajet prolongé, limitent les visites spécialisées en clinique externe à la suite d'un congé d'hôpital pour les ESC situés à une distance de 80 kilometres ou plus d'un centre pédiatrique tertiaire en comparaison à ceux plus près. De ce

fait, les ESC situés à de plus longues distances ont de plus grands risques de réadmissions dans un délai de 30 jours pour certaines conditions qui auraient pu être traitées à l'externe. Notre objectif primaire est d'examiner l'association de la distance à parcourir pour se rendre chez les fournisseurs de soins spécialisés (moins de 80 kilomètres comparé à 80 kilomètres ou plus) sur le degré d'utilisation des services de soins de santé, telles que les réadmissions à l'hôpital dans un délai de 30 jours suite à une hospitalisation initiale, pour les enfants âgés de 2 à 18 ans à des niveaux différents de complexité médicale dans la province de Québec. Les renseignements de cette étude de cohorte basée sur la population proviennent de plusieurs bases de données de la Régie de l'assurance maladie du Québec entre le 1 janvier 2010 et le 31 décembre 2013 et un modèle des risques proportionnels de Cox est utilisé pour tester les associations.

Résultats: De façon générale, les résultats démontrent que les ESC représentent 2.2% de la population entière d'enfants au Québec, dont 24% résident à une distance de 80 kilometres ou plus d'un centre pédiatrique tertiaire. Les ESC situés à une distance de 80 kilomètres ou plus d'un centre pédiatrique tertiaire en comparaison à ceux situés plus près, ont moins de visites médicales auprès de médecins de famille, pédiatres et spécialistes. Cependant, ces enfants ont plus de visites à l'urgence et d'hospitalisations à répétition. Toutefois, il n'y a pas d'association entre la distance à parcourir et le risque de réadmissions dans un délai de 30 jours lorsque les deux groupes de distance à parcourir sont comparés.

Conclusion: Bien que la distance à parcourir ne semble pas influencer le risque de réadmission dans un délai de 30 jours, les résultats démontrent que les ESC demeurant à une distance de 80 kilomètres ou plus des centres spécialisés tertiaires en comparaison à ceux situés plus près, utilisent les services de soins de santé de manière plus sporadique et

imprévue, tel qu'illustré par la fréquence des visites à l'urgence et le grand nombre d'hospitalisations. De plus, un tiers de tous les ESC au Québec n'ont pas de fournisseurs en soins primaires. Ces différences ne peuvent s'expliquer que par des barrières géographiques, telle que la distance à parcourir. La prochaine étape consistera à comprendre les facteurs aidants mais également les entraves qui affectent les fournisseurs en soins de santé primaires impliqués dans les soins des ESC. Ces informations supplémentaires nous permettront de mieux comprendre le mécanisme influençant le taux de réadmission chez les ESC afin de développer des programmes et une infrastructure aptes à réduire les réadmissions et améliorer les soins chez les ESC.

List of tables and figures

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Statement of originality

In this study I performed the following aspects which included: 1) choice of the study question 2) design of the study protocol 3) data extraction, statistical analysis, and interpretation. I also wrote all chapters of this master's thesis.

Application to the *Régie de l'assurance maladie du Québec* (RAMQ) and the *Commission d'accès à l'information* (CAI) in order to obtain the multiple datasets used in this study was done by Drs Patricia Li and Bruno Riverin as part of a larger unpublished study funded by the Canadian Institutes of Health Research (CIHR)-*Association entre la réforme des services de santé de première ligne et la qualité des soins chez les enfants vulnérables au Québec, Canada.* Dr Patricia Li and Marc Dorais received the RAMQ raw data used in this thesis.

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Abbreviations

CA: Census agglomeration

CAI: *Commission d'accès à l'information*/Information access agency

CCC: Complex chronic condition

CLSC: *Centre local de services communautaires*/ Local community service center

CMA: Census metropolitan area

CMC: Children with medical complexity

CSHCN: Children with special health care needs

ICD: International classification of diseases

ICES: Institute for clinical evaluation sciences

IQR: Interquartile range

Km: kilometres

Med-Echo: *Maintenance et Exploitation des Données pour l'étude de la Clientèle Hospitalière*/Maintenance and Exploitation of data for the study of Hospital Users

MIZ: Metropolitan influenced zones

Multi-CCC: Multiple system complex chronic conditions

NI: Neurological impairment

RAMQ: *Régie de l'assurance maladie du Québec*/Quebec Health Insurance Board

SIG: Geography Information System

Single-CCC: Single-system complex chronic conditions

TA: Technology assistance

US : United States of America

Chapter 1: Introduction

As advances in therapeutics and medical technology have evolved exponentially there has been a distinct shift in the landscape of pediatric epidemiology, both in Canada and in similarly developed countries. Baseline improvements in pediatric health care, reflected in reduced child mortality rates, are in large part due to the prevention of fatalities related to perinatal conditions, congenital anomalies and external causes such as motor vehicle accidents and drownings. [1] Consequently, many survivors of childhood-related diseases and disorders have been left with chronic medical conditions and complex care needs. [2-4] Through the years, this emerging cohort of children with multiple chronic co-morbidities has been particularly evident in pediatric inpatient care facilities. Beyond their intrinsic medical fragility, these children have a legacy of prolonged hospital stays and frequent hospital readmissions, in part because of gaps in health care services and the fragmentation of their complex care needs. [5-7] Traditional hospital care has been built on the model of acute intermittent care which adequately suits the care needs of a previously healthy child. Unfortunately, this model and the culture which surrounds it is a poor fit for the child with multiple chronic care needs, and results in delivery of care by multiple providers in multiple settings. Not surprisingly, a study examining pediatric hospitalizations in the United States (US) reported that children with complex chronic conditions were the group of patients that contributed the most inpatient days, which consequently totaled 40% of all hospital pediatric-related costs. [8] Although clinicians and researchers noticed this trend many years ago, [7] [9] there was no official definition which would have facilitated cohort analyses for research initiatives and service planning. In 1998, “children who are at increased risk for a chronic physical, developmental, behavioral, or emotional condition and who also require health and related services of a type or amount beyond that required by children generally” were defined as

children with special health care needs (CSHCN). [2] Working from this broad definition, Cohen *et al*. proposed a definition framework focusing on a particular high need subset of CSHCN: *children with medical complexity* (CMC) .[4]

CMC are a particularly vulnerable group of children with a wide range of medical chronic conditions; they may be survivors of extreme prematurity, suffer from genetic neuro-developmental disorders or even be dependent on medical technology devices such as a tracheostomy tube. Although their medical diagnoses differ, these children share similar concerns, challenges and outcomes which are the basis of the definition framework for CMC: 1) Chronic conditions severe enough to require specialized pediatric care and often associated with multi-system impairments. The complexity and intensity of the care needs can lead to medical fragility which manifests as increased risk of rapid clinical deteriorations, morbidity and mortality; 2) Functional limitations affecting participation in activities of daily living due to health issues such as physical and mental handicaps or medical technology-dependence; 3) High health care use such as hospital admissions, multiple outpatient health professional visits, as well as community and home services; 4) Extensive requirements and burden of care put upon the family unit such as time and energy dedicated to daily routine care, prolonged meal schedules, difficult transportation, not to mention associated family stress and financial strains. [4 10]

The vast majority of CMC are cared for in the family home with family members providing the majority of the medical care. [11] Health care restructuring in the 1980's and early 1990's was influenced by the needs of a growing elderly population and by family preferences. This led to a shift of services, known in Quebec as the *virage ambulatoire*, [12] towards community and home

based support of patients with chronic medical issues, instead of previously structured institution-based services. [13] Nationwide, it is now common practice to have children with life supporting care needs discharged to their family home on intensive care plans or intricate medical technology such as mechanical ventilation devices or infusion pumps for the administration of total parenteral nutrition. The caveat of this well intentioned initiative is that the extent of home and community support not only varies significantly by provinces/territories in Canada but also by and within regions, thus leading to an unequal distribution of services. [13 14] According to the *Canada Health Act*, Canadians are entitled to universal hospital and physician care [15]; but there is no minimum standard for homecare services such as nursing support and equipment coverage. Therefore, many CMC and their family are left struggling with limited public funds and a burden of care that goes far beyond what would be expected for a child with a less fragile chronic condition. [16]

Moreover, in view of their intricate and specialized care needs, CMC are usually followed by a community generalist and multiple specialized health care professionals. Cohen *et al.* reported that CMC had a median of 13 distinct physicians providing outpatient care from a median of 6 distinct medical specialties in Ontario, Canada. [17] CMC and their families navigate a compartmentalized health care system not suitable for patients with multiple chronic conditions. [18] Therefore, when CMC seek medical help for their chronic complex issues, they often cannot get the care they need. They are faced with health services that are lacking continuity, coordination, as well as holistic depth. [19 20] Many studies have shown CMC to be at increased risk of frequent and prolonged hospital admissions [6 8 21-23], medical errors [24], as well as unmet

needs such as preventive care, specialist care, dental work and services meant to optimize physical and cognitive development. [25]

To improve care delivery for CMC, especially in terms of coordination/continuity of care, a variety of structured programs have been developed within pediatric health care infrastructures to serve CMC. Most are based on a philosophy of proactive, coordinated, child and family-centred care. Studies have shown that restructuring health care delivery for CMC is not only associated with decreases in health care costs, but also a decrease in hospital readmissions and improved family satisfaction with care, [26-28] Most programs for CMC are based on *The Medical Home* model which is described in a policy statement from the American Academy of Pediatrics that promotes the delivery of care that is within reach, ongoing, holistic, family-centered, harmonized, compassionate, and culturally sensitive for all infants, children and adolescents with special health care needs. [29] The medical home endorses strategies such as: multidisciplinary care collaborations, identification of a "case manager" for coordination of care, resource optimization, facilitating medical communication between health care providers, and elaborating clear goal-directed care plans. [30] Although programs reflecting these values appear to be beneficial [28], the vast majority are located within pediatric tertiary care hospitals of large urban centres, potentially distant from the actual place of residence of many CMC. In view of this concern, many programs are establishing community collaborations and forming community complex care clinics integrated within tertiary care centres to bridge the distance gap. [26] However, this is not yet the case in Quebec, Canada, the province with the largest surface area in Canada. [31]

Is health care in Quebec fundamentally different for those living in urban versus rural regions? Pampalon *et al*. studied health services in the general population of Quebec and found that residents of rural zones were more likely to have a family physician but less likely to consult specialized care. Rural areas, especially the zones with low levels of metropolitan influence, also had higher hospitalization rates and a higher number of avoidable hospitalizations compared to urban areas. These differences were attributed to outpatient health service accessibility and availability. [32] For example, in order to reach outpatient specialty clinics, most rural residents may be required to travel significant distances, which in turn may limit the frequency of visits to seek expert care and lead to unnecessary hospital admissions for conditions that could have been managed as an outpatient.

Is there perhaps an exaggeration of this effect in the sub-population of CMC? CMC have high care needs and therefore are also high users of health care services, some that could potentially be reduced with optimal outpatient care such as readmissions to hospital. Although local primary care providers are essential in the care of CMC, they cannot realistically provide the full range of care required by CMC without the support of a multidisciplinary team of specialized healthcare professionals. Currently, the majority of specialized services are provided within pediatric tertiary care centres situated in large urban areas. For children suffering from chronic health issues, a distance greater than 80 kilometres from the hospital has been shown to negatively affect family dynamics and increase family anxiety regarding their ability to care for their child at home. [33] Eighty kilometres was identified as the critical driving distance cut-off since families living outside this radius usually have to dedicate a whole day of travel, and even plan an overnight stay, in order to reach their destination. We expected difficulties associated

with prolonged travels would limit specialty follow-up in, especially in the early period following a hospital discharge for CMC living at a driving distance of 80 kilometres or more to a tertiary pediatric centre compared to those living closer. Considering a fair proportion of early issues that may arise in the early post-discharge period could be addressed with adequate outpatient expert care, CMC living farther would be at increased risk of readmission within 30 days.

Currently, there is no published literature examining the impact of distance on care for CMC in Canada and its influence on health care utilization. Moreover, there is no published literature examining the CMC population of Quebec, Canada. By describing the CMC population in Quebec and exploring the association between driving distance to outpatient specialized care situated within pediatric tertiary care centres and health service use such as hospital readmissions, we aim to further understand the needs of this population and focus future research on community collaborations with tertiary care hospitals to better serve CMC not only in Quebec but in developed settings worldwide.

Chapter 2: Literature review

2.1 Defining CMC for research purposes

CMC are a subset of CSHCN that also have additional medical complexity. The Cohen *et al.* CMC definition framework includes the following dimensions: complex chronic health conditions, high healthcare needs, significant functional limitation and extensive health resource utilization. [4] Identifying CMC on a one-on-one basis may be straightforward with a thorough medical history. However it is particularly difficult at a population-level to capture all characteristics attributed to CMC. [34] Due to the heterogeneity of the CMC population, it is very challenging to distinguish a specific medical diagnosis—alone or in combination with other diagnoses—associated with medical complexity. For example, asthma is usually not characterized as a complex diagnosis, however if asthma is poorly controlled and associated with multiple co-morbidities such as atopic dermatitis, obesity, hypertension and steroid-induced diabetes, then a more complex picture arises which is associated with additional health care needs, utilization of services, as well as a negative impact on daily activities. [35] Clearly, any single health care condition can vary on an individual basis; consequently, it is difficult on a larger scale to pinpoint the level of medical complexity of a subject solely based on a list of diagnoses. Beyond a list of specific diagnoses, in order to fully grasp the intensity of health care needs and functional limitations associated with the health status of CMC, additional survey data from families and caregivers would be required. Unfortunately, with a source population of 1, 245, 249 children in Quebec during our data collection period [36], gathering direct individual information on such a large population was not feasible nor retrievable in the form of detailed census data. In the absence of a comprehensive database linking diagnostic codes, intensity of health care needs and functional limitations, most

researchers of CMC have deferred to a diagnostic classification system that identifies health issues attributed to CMC based on International Classification of Diseases (ICD) codes. Many systems have been used in the literature to study CMC, the most common include: 1) The "Clinical Risk Groups" from 3M Health Information Systems which developed hierarchical pediatric diagnostic groups defined as minor, moderate, dominant, catastrophic, single and 2-to-3 chronic conditions. These groups can be combined to create a cohort of children with medical complexity. [34 37] 2) The "Patient Medical Complexity Algorithm" has been recently developed by the Seattle Children's Hospital Center of Excellence on Quality of Care Measures for Children with Complex Needs. This algorithm groups children in 3 categories which include a) complex, chronic disease b) non-complex, chronic disease c) non-chronic disease, with a sensitivity of 89% and specificity of 85% to identify children with complex chronic disease. [34 38] 3) The "Chronic Condition Indicator", developed by the Agency for Health care Research and Quality in the United States, dichotomizes diagnoses into chronic and non-chronic. This system can be divided into 18 clinical categories which can be used to count the number of chronic conditions as an indicator of medical complexity but is not specific to children. [34 39] 4) The "Complex Chronic Condition (CCC)" system, which is the system used in this study, was developed by Feudtner *et al* [40]. This classification system identifies childhood diagnoses of chronic health conditions that are associated with mortality, morbidity, significant resource utilization, functional impairment, as well as diagnoses that are targeted by dedicated complex care clinical programs. CCC are conditions that are expected to last at least 12 months and require specialized care offered in pediatric tertiary care centres. CCC can be further divided in organ-system categories and separated in clinically relevant groups with different levels of medical complexity such as neurological impairment (NI),

multiple system complex chronic conditions (multi-CCC), single-system complex chronic conditions (single-CCC) and technology assistance (TA). [34 40 41]

In this study, we selected the CCC system to identify CMC in our health administrative database for the following reasons: 1) Although it is not designed to capture all chronic diagnoses such as asthma or bipolar disorder, the CCC-system was preferred because it is open-access, specific to the pediatric population and targets diagnoses that are often followed in designated complex care programs due to their association with high health care service use and functional limitations; 2) This system can also be divided into different levels of medical complexity and organ-systems which can be useful from a public health standpoint for identifying the most vulnerable groups or the ones more likely to benefit from an intervention; 3) Finally, the CCC system has been widely used in health service research including studies exploring hospital readmissions. [6 17 21 42-45]

Throughout the current thesis, we use the term CMC when referring to children meeting Cohen *et al.*'s framework (complex chronic health conditions, high health care needs, significant functional limitation, extensive health resource utilization).[4] We also use the term CCC where studies identified and described conditions related to CMC within health administrative data using Feudtner's CCC system. [40]

2.2 Access to optimal care for CMC

Finding the optimal structure of care to address the care needs of CMC has been challenging. Sections 2.21, 2.2.2 and 2.2.3 are meant to illustrate the context in which families and care

providers are trying to adapt and overcome the struggles associated with care delivery for this ever-growing population of medically complex children.

2.2.1 Access to optimal care for CMC: family perspectives

Most CMC, following their initial hospitalization—after birth or subsequent to an acute deterioration—are discharged home with active chronic medical issues requiring ongoing therapies that would often have formerly been administered in a hospital setting. [46] As caregivers discover how to deal with concerns related to their child's long-term well-being and uncertain prognosis, social stigma and new family dynamic [33], they are also expected to learn how to provide vital therapies to fulfill the care needs of their sick child at home. Typically, CMC are cared for in the family home with close family members assuming the majority of the care needs. In a national profile exploring CMC caregiver challenges in the United States, Kuo *et al*. reported that 54.1% of CMC had a family member quit their job due their child's medical condition, 56.8% had financial difficulties and 48.8% had at least one unmet care need, which was defined from a list of 14 needs such as preventive care, specialty care, dental care, homecare services, and physical therapy. Moreover, caregivers of CMC reported a median of 11 to 20 hours per week of direct home care, a median of 2 hours per week of care coordination and a median of 11 to 15 physician visits per year. [16] When asked about the ideal care provider, families valued continuity of care, effective and open-communication with clear explanations and ongoing education regarding their child's diagnosis and treatment, as well as consideration and respect of family values and opinions when building a relationship with their healthcare providers. On the other hand, limited access to a trusted health care provider left parents with high emotional stress, lack of trajectory and perceived loss of control which

would push them to seek medical treatment via clinics or the emergency department more frequently. [19 45 47]

2.2.2 Access to optimal care for CMC: the medical home

In 1992, the American Academy of Pediatrics released a statement encouraging all primary care providers to put into practice elements of the medical home model. This model aims at delivering care that is easily accessible and that partners with the school, community and specialty care in order to be proactive, preventive, holistic, harmonized and centered on the child and family unit continuously throughout the years. [48] The importance and necessity of the medical home model for children with special health care needs, especially CMC, has been reinforced in subsequent publications. [29 49 50] Palfrey *et al*. assessed the implementation of the medical home for CMC in 6 community pediatric practices, which included the following main interventions: assigning a case manager for coordination of care, creating individualized healthcare plans, modifying clinic routines to optimize accessibility, promoting ongoing medical education and creating well-established links with a specialized multidisciplinary pediatric team. They found that such interventions were particularly beneficial for CMC. According to caregivers surveyed in the study, health care delivery was characterized as being "much easier" after implementation of the medical home, including getting resources, appointments or referrals to specialists and early medical care during illnesses. Although emergency visits were not reduced, there was a significant decrease in hospitalizations from 58% to 43% from baseline to after the implementation of the medical home, respectively. [27]

2.2.3 Access to optimal care for CMC: identifying a medical home provider

Although the medical home seems like the ideal care delivery concept, especially for children with chronic conditions, many challenges remain when it comes to CMC. For example, it is still unclear whether the medical home for CMC should be provided by primary or specialty care providers, or both. Although it is implied that primary care practices should provide a medical home for all children including CMC, in reality many community primary care providers lack the support and infrastructure to maintain all elements of the medical home model in their practice. [29 30] Caring for CMC and their highly intricate medical needs requires the guidance and expertise of a specialized multidisciplinary health care team. By providing a medical home for CMC, primary care providers are not only expected to deliver primary care, but also offer follow-up on multi-system conditions and their acute-on-chronic issues. Primary care providers are also given a case manager role to ensure communication and coordination between services offered in the school, community and specialized centres. All these responsibilities are time consuming and hindered by the demands and constraints of a busy community outpatient clinic or office. [27 30] In a cross-sectional survey of 132 primary care providers (family physicians and pediatricians) in the United States, 77% reported caring for CMC as "difficult". The difficulties identified included: 1) lack of communication between health professionals; 2) difficulty setting up referrals with specialists; 3) difficulty providing coordinated care between providers; 4) lack of community support; 5) lack of CMC specific medical knowledge; and 6) poor awareness of available community resources. [51] In another survey including only pediatricians, 65% agreed that primary care providers should be at the center of the medical home for CSHCN. However, for the subset of these children with medical complexity, 43% agreed that specialty care would be more appropriate; cost and time

were viewed as barriers. [52]

As a result, multiple complex care programs and affiliated clinics have been created within tertiary care hospitals to centralize and improve care of CMC. In a caregiver survey assessing fulfillment of CMC health care needs following enrollment in a structured-care program within a tertiary care centre, Kuo *et al.* reported that 56% still considered their primary care provider as the initial point of contact for medical issues, but acknowledged that the structured-care program addressed some of their previously unmet care needs such as primary check-ups, various therapies (speech therapist, physiotherapist, occupational therapist, etc.), respite care, referrals to specialist and mental health support. [50] While these specialized programs appear promising, they are generally located in major pediatric tertiary referral centres. This raises the issue of distance to such a centre as a potentially significant barrier to optimal care.

2.3 Barriers to care for CMC

Social determinants of health are conditions of the environment of individuals and their everyday life that affects wellbeing and health outcomes. Multiple frameworks exist in reference to the elements that comprise those determinants. [53] However *access to health services* is regularly presented as a key constituent. [54] Barriers in accessing health services are often described in the context of health behaviors and the cost of a health action. These barriers are created from systemic inequities influenced by social and economic factors. [25 55]

Many references presented in sections 2.3.1 and 2.3.2 expose barriers related to CSHCN.

Although CMC are the small subset of CSHCN that also have additional medical complexity, we consider the findings presented to be representative of our population or even a conservative estimate of the effect of those barriers, as medical complexity itself has been shown to be related to health care inequities. [25]

2.3.1 Traditional barriers

When it comes to CSHCN, certain traditional barriers have been highlighted in the literature such as race, language and outcome. Strickland *et al*., in their analysis of the 2002 *National Survey of Children with Special Health Care Nee*ds in the United States, examined access to components of the medical home including: usual source of care, personal doctor or nurse, referrals for specialty care, coordinated care and family-centered care. They found that the odds of not having access to a medical home was significantly increased for families of non-Caucasian race, poor financial status and for a child with a condition that impacts daily activities. Additionally, children without a medical home were twice as likely to have unmet care needs compared to children with a medical home (adjusted odd ratio of 2.1, 95% confidence interval (CI) 1.8- 2.4). [49] These social disparities were in line with other publications assessing medical home access and unmet needs for children with chronic disease [56-58].

2.3.2 Geographical barriers

Geographical barriers have been described for CSHCN in the United States; the highest percentage of unmet needs is experienced in the western and southern states [59 60], as well as in urban communities. [51 61] These inequities have been partly attributed to difficulty accessing

certain health care services due to differing plans of individual medical insurance coverage and state-level initiatives and funding campaigns to support such services. [60]

Despite having a universal healthcare system, Canada is not immune to such geographic disparities in health services. Certainly, inter-province disparities exist but with a country as large as Canada, regional differences especially between urban and rural areas are an issue. [13] [53] In fact, given that specialized resources are often concentrated in metropolitan areas, many individuals have to seek such services far outside their community For example, as mentioned above, most Canadian structured complex care programs and other specialized medical services for CMC are situated within tertiary care hospitals of large cities. Thus, factors such as transportation, travel time, and driving distance can potentially affect timely access of required health care services.

2.3.2.1 Transportation

Transportation barriers are often cited in literature exploring health care access; 25 separate studies have reported that 10-51% of patients perceive transportation as a hurdle to health care access. Difficulties in transportation have also been linked to missed or forgone appointments; in fact two pediatric studies stated that 18-21% of families blamed transportation as the reason for not seeking a required health care service. Additionally, patients of lower compared to higher socio-economic status were shown to have higher odds of being affected by transportation barriers when having to reach care. [62] [63]

When it comes to CSHCN, Skinner *et al.* looked at differences between rural and non-rural families. They reported that 91% of patients in rural areas had a usual source of care provided by a generalist doctor or nurse practitioner but were significantly less likely to be treated by a pediatrician compared to their urban counterparts (adjusted odds ratio of 0.5, P <0.01). Despite limited access to a pediatrician, these rural families had a similar proportion of reported unmet needs compared to the urban ones at 16.4% versus 15.4% respectively (P >0.05). However, there were significant differences in the reported reasons behind those unmet needs. Rural families were more likely to report that the type of care needed was not available in their area or that they had transportation problems, compared to urban families (adjusted odds ratio 1.81 and 1.58 respectively, P <0.05). [61]

These results are of great importance considering that CMC have additional barriers to overcome when it comes to travelling to access care. In fact, CMC often experience laborious and difficult travels due in part to safety issues related to transportation and prolonged immobilization, their need for a wheelchair-accessible vehicle, as well as concurrent transport of medical equipment such as feeding pumps, oxygen compressors and emergency kits. CMC require frequent scheduled and unplanned health care visits for adjustment of their management plans, escalation of care during acute deteriorations and various interventions; delays in these visits can result in medical complications, increased emergency room visits and hospitalizations. [64]

2.3.2.2 Travel time and driving distance

Travel time and driving distance are related, but represent slightly different entities. Driving distance refers to the kilometers to travel via standard road maps to reach medical care, while

travel time takes into account the time required to reach such care considering the terrain between point A and point B such as bodies of water (i.e., requiring a ferry) or traffic. [65]

Limited studies are available on travel time, but Bosanac *et al*. reviewed various health service publications and reported a one-way travel time of 30 minutes as a standard in health care planning. A travel time of more than 30 minutes is hypothesized to negatively influence one's inclination to travel to a certain health care facility but also the subsequent pattern of health care utilization, therefore reducing the frequency of health care visits. [65]

Meanwhile, Syed *et al.* examined the impact of distance in a review that evaluated transportation barriers in the general population. They reported mixed evidence when looking specifically at distance and its effect of healthcare access. Most studies examined heterogeneous populations and different outcomes such as non-compliance with medical appointments or treatments, specific patient outcomes, as well as patient reports on perceived barriers. Six out of 9 studies found an association between longer distance to care and difficulties with access, 2 found equivocal results, while 1 assessing survival of cancer patients on phase II clinical trials found a decreased hazard ratio for death with increasing distance. [62]

When looking more specifically at children with chronic health issues, Yantzi *et al*. [33] studied the impact of distance on these families using questionnaires evaluating family stress and functioning. They found that distance to a hospital was associated with a negative effect on the family dynamics and increased family anxiety regarding their ability to care for their child at home. They found that families living at a distance of more than 80 kilometres were 224%

more likely to have difficulties keeping the family unit in harmony. The critical distance of 80 kilometres, especially during a child's hospitalization would separate:

> "…families who live close enough to the hospital that only part of a day is required to visit with the child, and those families whose visiting the child requires a whole day or even an overnight stay. Theoretically those families who live 80 km or less from the hospital would also be able to return home, rest, and maintain parts of the family's routine. Those families who live more than 80 km from the hospital would face a more significant disruption in their family life." [33]

Although most families will not hesitate to dedicate the time and energy to reach the most comprehensive level of care for their children, Yantzi *et al*. suggested that in some cases the added burden of distance on an already stressed and overwhelmed family could potentially act as a tipping point. [33] Interestingly, despite these associated strains, Cohen *et al*. found no difference in the residential moving pattern of families in Ontario, Canada, caring for a child with chronic conditions compared to healthy children in order to be situated closer to specialized care. [66]

2.4 Geography and health in Quebec, Canada

Pampalon *et al*. [32] studied the demographics, health status and health service use of Quebec residents living in rural versus urban areas using Statistics Canada categorization of location of residence by census metropolitan area (CMA), census agglomeration (CA) and metropolitan influenced zones (MIZ). CMA and CA are based on population density and represent the largest

population centres according to Statistics Canada's 2011 *Census*. MIZ are zones less populated, geographically outside CMA or CA, and have different levels of metropolitan influences as distance to the CMA or CA increases ("Strong MIZ" to "No MIZ"). MIZ do not exactly correspond to a specific distance cutoff from a CMA or CA but most "Weak MIZ" and "No MIZ" are situated outside that 80 kilometres radius. [67] In Quebec all 4 pediatric tertiary care centres are situated within the 3 largest CMA: Montreal, Quebec City and Sherbrooke as shown in Figure 2.1

Pampalon *et al.* [32] found a decline in employment, education level and financial status with increasing distance from urban centres. Health status of rural citizens did not vary significantly from urban ones. When examining health services, rural residents—no matter the level of MIZ—were more likely to have a family physician but less likely to consult physicians such as a specialists compared to individuals in urban centres. Hospitalizations were higher in rural areas; up to 40% more likely in areas with "weak MIZ" or "no MIZ". On that same theme, avoidable admissions would increase progressively from areas of "strong MIZ" to "no MIZ". These differences were attributed to suboptimal health service availability and poor accessibility in rural regions; such that outpatient visits for specialty services may be limited by significant travel distances and consequently lead to the increase of unnecessary hospitalizations for conditions that could potentially have been managed as an outpatient. [32]

2.5 Health care utilization of CMC

CMC are high users of healthcare services with many homecare needs, outpatient visits and readmissions. A previous Canadian publication from Cohen *et al*., exploring health care

utilization and cost for CMC (identified in health administrative data using the CCC system) in Ontario, Canada, attributed one third of total pediatric healthcare spending to this population. [17]

2.5.1 Homecare

Thirty-six percent of Cohen's cohort of CMC in Ontario, Canada, used homecare services in the form of nursing support, coordinator visits and various therapies. The highest users of homecare services were children suffering from neurologic impairment (NI) with technology assistance at 81%, followed by multi-system CCC with technology assistance at 73%. [17]

2.5.2 Outpatient visits

Several studies have reported on the utilization of outpatient services by CMC when it comes to primary care, specialty and emergency department visits. In fact, over a 2-year period, CMC were reported as having a median of 13 distinct physicians (IQR 8 to 20) including a median of 12 primary care visits (IQR 6 to 20) and a median of 6 distinct specialist visits (IQR 4 to 8). Multi-CCC with technology assistance led the subgroups of medical complexity with a median of 24 distinct physicians (IQR 17 to 38). All groups with technology assistance, including single-CCC, multiple-CCC and NI, had the largest medians for distinct physicians ranging from 18-24, compared to 11-16 for those without technology assistance. Meanwhile, the median number of emergency department visits for all groups ranged from 2 to 3, while all groups had a median of 1 for same-day surgery. [17] Kuo *et al.* reported CMC to have a mean of 19 outpatient visits and mean of 0.5 emergency department visits per year; the two main reasons for consultation included pneumonia and convulsions. [20] Lastly, O'Mahony *et al.*

reported that over a 1-year retrospective study, 20% of all emergency department visits within their pediatric tertiary care centre were from children with chronic disease, of which 12% were classified as medically complex. [64]

2.5.3 Inpatient care

Many studies have also focused on inpatient services for CMC, especially in regards to the descriptions of hospitalizations and readmissions. For example, Cohen *et al.* found that overall, 44.1% of CMC experienced at least one hospital admission over a 2-year observation period. Multi-CCC with technology assistance had a 2-year admission rate of 78.3% while NI with technology assistance were in second place with 67.1% [17]. Kuo *et al.* reported a mean of 0.26 inpatient visits per year for CMC. [20] Berry *et al*. explored hospitalization of children across 37 US pediatric hospitals in a 5-year retrospective study and found that 18.8% of admissions and 23.2% of total costs were attributed to a subset of patients with recurrent hospital admissions that represented 2.9% of the total population. Recurrent hospital admissions were more likely to be seen in children with CCC. [6]

2.6 Pediatric readmissions

Readmission following index hospitalization is commonly used in the literature, as well as in health care policies, to represent the quality of care received in the period surrounding hospital discharge. [6 21 68] Readmissions are also a common topic when it comes to CMC health service research as this group of vulnerable children is known to have some of the highest rates of pediatric readmissions. For instance, Berry *et al.* in a study of readmissions in the general pediatric population reported odds of increased readmission frequency of 5.61(95% CI 5.45-

5.78) for children with CCC compared with no CCC, and 2.85(95% CI 2.74-2.96) for children with technology assistance compared to no technology assistance. [6]

Overall rates of readmissions for CMC vary in the literature. A 2-year retrospective study found a 30-day readmission rate (i.e., readmission within 30 days of a previous hospital admission) of 6.3% for patients with CCC, while more in-depth analysis showed patients with 3 or more CCC had the highest odds of readmission compared to those with 1 CCC (odds ratio 2.3, 95% CI 1.5 -3.5).[42] Jurgens *et al*. reported in an 18-month retrospective study a 30-day readmission rate of 19% for patients with at least 1 CCC and increased odds of readmission with increasing number of discharge medications (odds ratio 1.11, 95% CI 1.03-1.20). [21] Berry *et al.* in a study exploring hospitalization characteristics of CMC, reported a rate as high as 25.4% of 30-day readmission over 2-years in a sample of CMC involved in a dedicated structured clinical program for CMC (inpatient and/or outpatient services); however, when CMC were stratified based on their level of medical complexity, it was a sample of more severely affected CMC (conditions affecting a minimum of 2-3 organ systems or severe neuro-developmental disability, in addition to technology assistance) that drove this high rate of readmission within 30 days. [44] Briefly, rate of readmissions for CMC seem variable, yet we can appreciate a trend suggesting that rates of readmissions amplify with increasing medical complexity.

Studying pediatric readmissions is relevant if we think a considerable percentage of those readmissions are potentially preventable. Yet, the proportion of those potentially preventable readmissions also varies widely in the pediatric literature. For example, Toomey *et al.*

recently conducted an analysis of medical records and 1,459 interviews with primary care providers, inpatient physicians and parents of patients readmitted within 30 days to freestanding children's hospitals to measure readmissions preventability; they reported 29.5% of general pediatric readmissions as potentially preventable. [69] Hain *et al.* conducted a single-centre retrospective chart review and estimated rates of avoidable 15-day pediatric readmission at 20.0%(95% CI 14.8 to 26.4).[70] Finally, Gay *et al*. looked at the top 30 diagnostic groups most commonly readmitted to 58 US pediatric hospitals using the *3M-Potentially Preventable Readmissions software* and found that for 16 of the 30 diagnostic groups, 50% or more of the 30-day readmissions were considered potentially preventable. This study further delineated that the 5 diagnostic groups with the largest proportion of potentially preventable readmissions included appendectomy, connective tissue disorder, ventricular shunt procedure, bronchiolitis and asthma; and concluded that the leading cause for potentially preventable readmissions in pediatric hospitals that had a mean average of at least 50 admissions per year was identified as post-operative complications and device infections. [71] While there is no clear consensus on the percentage of preventable readmissions, the literature universally illustrates that improvements can be made.

Certainly, many studies support that a fair portion of readmissions, even for CMC, could potentially be prevented with good quality outpatient care and improved hospital-to-community continuity of care which includes proper discharge planning, timely outpatient follow-up and adequate inter-provider communication. A systematic review examining prevention of admission for CMC highlighted the immediate period following hospital discharge as a high-risk time for readmission due to its relation to poor compliance with

therapeutic plans, issues with medical technology use/maintenance, family-related stress and inadequate outpatient follow-up. [23] Indeed, multiple elements come in play when factoring the variability of readmissions: some are unknown or inherent to the context, while others can be changed, such as outpatient follow-up. One illustration of this comes in a study from Brittan *et al*., in which early follow-up was associated with lower odds of readmission for CMC. [23 42 68] A time-to-event analysis showed a 50% decrease in the hazard of 30-day readmission for children with CCC followed as an outpatient between day 4 and 29(hazard ratio of 0.5, 95% CI 0.4-0.7) compared to those with no post-discharge follow-up. [42]

In sum, although there is variability in the reported rates of potentially preventable readmission in children, a considerable proportion of readmissions are considered preventable and there is an opportunity for further research and quality improvement initiatives, especially ones targeting the early post-discharge period for CMC. Again, the solution for preventing unnecessary readmissions is more complex and cannot be simplified to a sole factor, but gaining information on that early high-risk period following hospital discharge is an important starting point**.**

2.7 Why focus on readmissions within 30 days and geographical barriers?

CMC have one of the highest rates of pediatric readmission. [6] Although it is clear not all CMC readmissions are preventable, current literature suggests room for improvement especially when it comes to facilitating and optimizing the transition of care from the hospital to the community. Readmissions in CMC warrants further research in order to identify various obstacles, such as geographical barriers, that may influence the variability in readmissions. Understanding the

underlying mechanisms of readmission is primordial as the ultimate goal is to guide targeted interventions in order to reduce unnecessary readmissions in CMC.

The study of readmissions can be challenging simply due to the wide range of time intervals that "readmissions" may represent. For example, how does a readmission within 15 days of hospital discharge differ from a readmission within 30 or 365 days of hospital discharge? In recent years, readmissions within 30 days have been the main focus of health care reforms in the US due to ubiquitous use as a quality of care measure. [72] The focus on readmission within 30 days is largely due to its close relationship with the care received around hospital discharge and the foreseen opportunity to prevent avoidable readmissions with improved discharge care—discharge care not only received during the inpatient stay but also extended to and maintained in the community once the patient is at home. For example, 1 out of 10 early readmissions for CMC were due to complications related to technology assistance [44]; such admissions could possibly be avoided with ongoing parent education and appropriate post discharge care.

Furthermore, the body of literature that focuses on readmissions within 30 days and the early post-discharge environment is interesting since it addresses elements that we can improve upon or adapt to. For instance, assuming an early follow-up appointment is the key to a successful discharge plan, what are the real life barriers that can interfere with that plan? Even though post-discharge outpatient follow-up may seem straightforward, this routine practice can pose some difficulty for CMC residing far from specialized centres or those with barriers to transportation. CMC, especially following a hospital admission, are likely to have a new or

modified care plan that requires fine-tuning or reinforcement as an outpatient. Considering specialized professionals are often significantly involved in the management of CMC, easy access to such expertise, in addition to assessment by their local primary care provider, may influence their post-discharge outcomes. Therefore, since most pediatric specialists are situated within pediatric tertiary care centres, travel distance may affect the timing and frequency of early outpatient follow-up and leave remote CMC at increased risk of a readmission within 30 days, especially for conditions that could have been managed in outpatient settings.

In summary, while more and more structured clinical programs for CMC are being created in pediatric hospitals to promote continuous, coordinated and family-centred care with the ultimate goal of improving care delivery for these children, these programs are concentrated in large urban centres. These initiatives seem beneficial overall, yet CMC still experience an excessive rate of readmission. [44] Nevertheless, a proportion of these readmissions is likely preventable and can potentially be reduced by facilitating the continuum of care from the hospital to the community. As follow-up in the early post discharge has been associated with reduced odds of readmissions within 30 days, could proximity to specialized outpatient expertise contribute to the equation and reduce readmissions for CMC? As previously described, specialized care is often exclusively offered within pediatric tertiary care centres; access to such outpatient services post discharge may be more difficult for families living more remotely. Consequently, these CMC may be at more risk for hospital readmission related to conditions that could have been managed in the outpatient setting guided by the proper medical expertise. In order to shed some light on the matter, this study explores

whether geographical barriers affect care transitions of CMC to the community after hospital discharge by estimating the risk of readmission within 30 days according to driving distance to the closest pediatric tertiary care centre. We will also describe the health care utilization of CMC in Quebec, Canada and other factors that may potentially affect readmission within 30 days such as having a primary care provider, the range of needed specialty care, and dependence of medical technology. [6 73 74]

Figures and tables (Chapter 2)

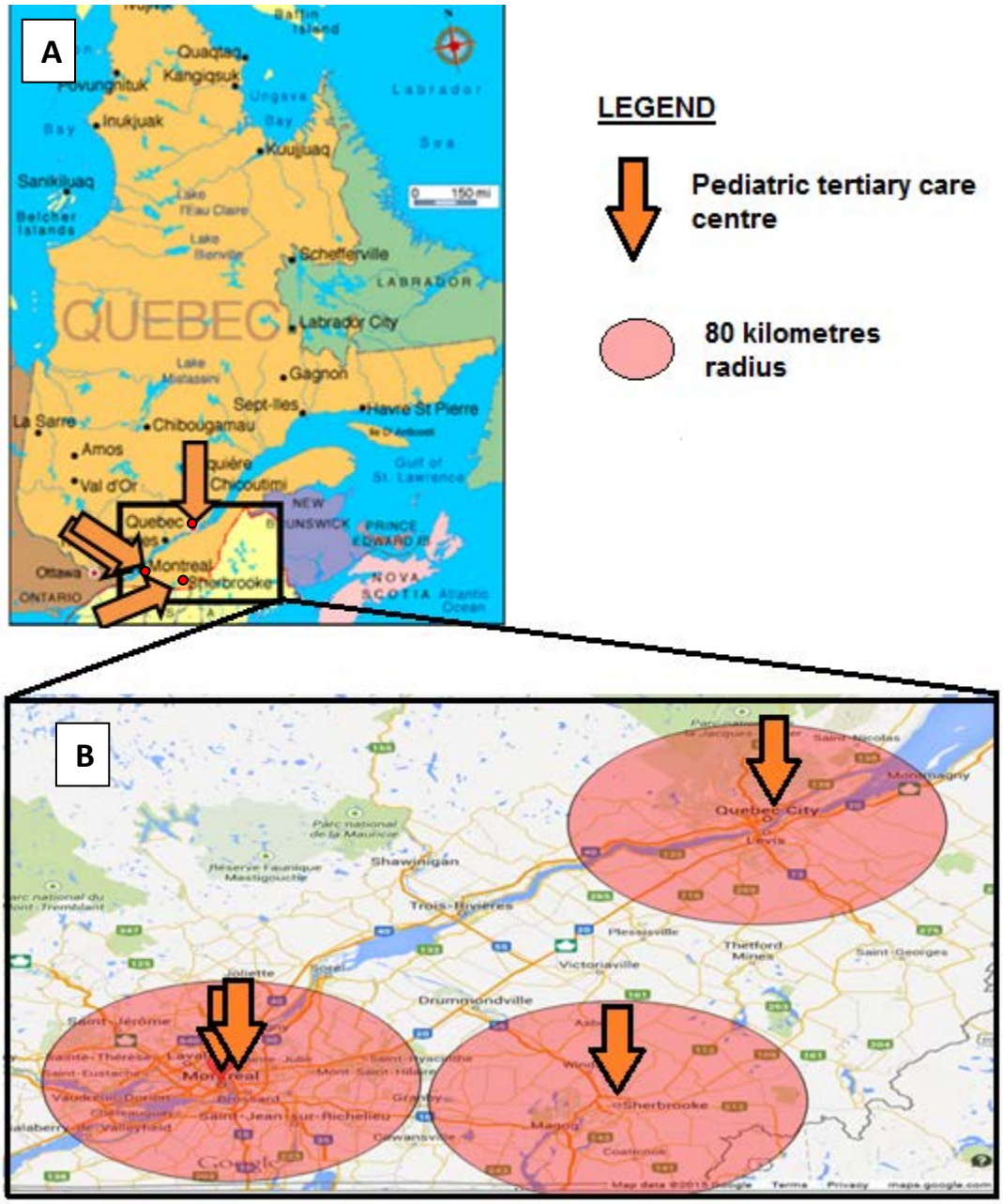

Figure 2.1 (A) Map of Quebec, Canada; (B) Focus on the location of the 4 pediatric tertiary care centres (in Quebec City, Sherbrooke, and Montreal) and their surrounding 80 kilometres radius

Images modified from: (A) Magallan Geographix(1997).*Map of Quebec.* www.maps.com (B): Google Maps. (2016). *Map of Quebec*. www.googlemaps.com

Chapter 3: Hypothesis and objectives

3.1 Hypothesis:

The general hypotheses guiding this study are:

1) *The distribution of CMC according to distance to the closest pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more) will be similar to the proportions reported by Cohen et al* [66] *and will not be significantly different by level of medical complexity(i.e., single CCC vs. multiple CCC vs. neurologic impairment, with or without technology assistance).*

In a study examining residential movement patterns of children with chronic conditions in Ontario, Canada, Cohen *et al.* described the proportions of single-CCC and multiple-CCC living at least 80 kilometres from a specialized pediatric care centre at a proportion of 22.8% and 21.1% respectively. As Ontario is the neighbouring province to Quebec, Canada, and shares similar demographic and geographic characteristics, as well as a universal health care structure, we hypothesize that the residential distribution of CMC according to distance to specialized pediatric care will be similar to the proportions reported by Cohen *et al.* [66]

2) *CMC living beyond 80 kilometres driving distance to a pediatric tertiary care centre will have more visits to primary care providers (family physicians or pediatricians) but fewer specialist visits compared to children living within a driving distance of 80 kilometres due to geographical barriers such as driving distance.*

By definition, CMC are characterized by having one or more chronic medical conditions that are severe enough to require expert specialized pediatric care most often offered in pediatric tertiary care centres. [4] CMC are also defined by their medical fragility leading to frequent medication or care plan adjustments, especially following an acute deterioration, to avoid certain health complications or illnesses that if present would inevitably require an emergency room visit or hospital admission for proper management. [4] While CMC may have easy access to their local primary care provider (PCP) and although a PCP is a key player in the care of CMC, it is unrealistic for them to provide the full spectrum of care required by CMC without appropriate support. [30] Because of this, many families may be obliged to travel on a regular basis to pediatric tertiary care centres in order to receive the care they need. The costs and time required to reach specialized care for CMC living more remotely may influence the frequency of those visits.

3) *CMC residing at a driving distance of 80 kilometres or more from the closest pediatric tertiary care centre will put CMC at more risk of readmission within 30 days of hospital discharge from the index admission as driving distance may limit access to outpatient specialty care services following hospital discharge.*

As most outpatient pediatric specialty services are situated in pediatric tertiary care centres within large urban centres, we presume that driving distance may act as a geographical barrier for CMC to obtain appropriate follow-up or preventive scheduled visits to seek expert opinion following a hospital discharge. CMC living at a driving distance of 80 kilometres or more from specialized care may be more at risk of a subsequent deteriorations or complications following

hospital discharge due to limited or difficult outpatient specialty care access and subsequent readmissions for conditions that could have been managed in the outpatient setting.

4) *Exposure to a travel distance of 80 kilometresor more in order to reach the closest pediatric tertiary care centre in combination with the highest levels of medical complexity—children with neurological impairment, multiple organ-system involvement and technology assistance—will have an even greater effect on risk of readmission than the individual effect of each factor.*

Many studies have shown that children with increasing level of medical complexity are more likely to experience a readmission within 30 days following an index hospitalization. [23 64] We expect the level of medical complexity to act as an effect measure modifier with travel distance on the risk of readmission within 30 days.

3.2 Overall objectives

Our overall objectives were to describe and analyze the association of driving distance to reach the closest pediatric tertiary care centre with health care utilization patterns of children with varying level of medical complexity in a cohort of CMC aged 2-18 years derived from population-based administrative data in Quebec, Canada.

The specific aims were to:

1. Describe the clinical characteristics of CMC by driving distance to a pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more)
2. Describe patterns of healthcare utilization of CMC (outpatient primary and specialty care services; and hospital use, including hospital readmissions) by driving distance to a pediatric tertiary care centres (less than 80 kilometres versus 80 kilometres or more).
3. Determine if exposure to a driving distance of more than or equal to 80 kilometres from a pediatric tertiary care centre is associated with increased risk of hospital readmission within 30 days of hospital discharge (primary outcome) for children with varying level of medical complexity.
4. Determine if level of medical complexity and travel distance act as an effect measure modifier of hospital readmission within 30 days of hospital discharge.

Chapter 4: Study methods

4.1 Overview of the study design

The study design was a population-based cohort study of CMC in Quebec, Canada, aged 2-16 years on January 1, 2012 who were actively registered in the provincial medical insurance database (RAMQ) between January 1, 2010 and December 31, 2013, using linked health administrative data provided by the RAMQ. The CMC cohort was identified using the CCC diagnostic algorithm [17 40] between January 1, 2010 and December 31, 2011. Outcomes related to health care utilization were collected between January 1, 2012 and December 31, 2013. Details related to the study timeline are outlined in the Table 4.1.1.

4.1.1 Ethics approval

This study was approved by the McGill University Health Centre Research Ethics Board (see Appendix 1 for copy of approval letter), the *Commission d'accès à l'information* (CAI), and the RAMQ. All information utilized in this study had patient identifiers removed prior to transfer to our research group to respect the protection of personal data.

4.1.2 Description of databases

4.1.2.1 RAMQ database

RAMQ is under the supervision of the Minister of Health and Social Services and manages the provincial healthcare insurance program in Quebec, Canada. Health Insurance Plan Coverage is compulsory for every resident or temporary resident of Quebec who fit the eligibility criteria stated by law. All members have a unique identifier number which links the RAMQ data with different sources or other databases such as hospital discharge data from the *Maintenance et*

Exploitation des Données pour l'étude de la Clientèle Hospitalière (Med-Echo). Coverage ceases if a person dies or emigrates out of the province. Since 1983, the RAMQ has compiled their computer database using the following sources: Beneficiary files, Prescription services and Fee-for-service physician claims. In this study we used RAMQ database information from the following sources which are outlined in more details in Appendix 2:

1) The Beneficiary files database contains socio-demographic information for every registered member and including age, gender, *centre local de services communautaire* (CLSC) and health region.
2) The Fee-for-service physician claims include information for all claims submitted by physicians in inpatient or ambulatory care settings regarding services offered to members of the insurance plan.

Finally, in order to obtain socioeconomic information for our study cohort, we requested the material deprivation index developed by *Institut national de santé publique du Québec* (INSPQ).[75] The material deprivation index was calculated by using 2011 census data from Statistics Canada and linked to the RAMQ database.

4.1.2.2 Med-ECHO database

Med-ECHO is a database with information collected on all hospital stays that occurred in Quebec establishments offering general or specialized care since 1987. This database can be linked using the same unique identifier number as the RAMQ. During each hospitalization, data is collected on administrative admission records, medical diagnoses, hospital services,

intensive care records and interventions received. Data available for each admission is also outlined in more details in Appendix 2.

4.1.3 Construction of the CMC study cohort

The cohort was gathered from the source population of all children in Quebec continuously registered to the RAMQ between January 1, 2010 and December 31, 2013, who were 2 to 16 years old on January 1, 2012, and who had 1 hospitalization or 2 physician claims from January 1, 2010 to December 31, 2011 that included at least 1 complex chronic condition (CCC) diagnostic code. CCC was defined using a combinations of ICD-9 and ICD-10 codes relevant to CMC based on a previously used framework developed by Feudtner *et al.* [40] and adapted by Cohen *et al.* [17] CCC represent medical conditions that are expected to last at least 12 months and are severe enough to require specialized pediatric care in a tertiary care centre. The CCC are further sub-classified into 9 body system categories which include neurologic involvement, cardiovascular, respiratory, renal, gastrointestinal, hematologic and immunologic, other congenital or genetic defects, metabolic and malignancy. ICD-9 and ICD-10 diagnostic codes for each category and sub-classifications are shown in Appendix 3 and Appendix 4.

We excluded from the study cohort children with RAMQ discontinuation due to migration outside the province of Quebec or death during the study period from January 1, 2010 to December 31, 2013 and CMC with missing information in regards to CLSC region (due to the inability to determine driving distance to the closest pediatric tertiary care centre). Children living in Nunavik, Grand-Nord du Quebec and Terres Cries de la Baie James, delineated according to their respective *Centre de santé et services sociaux* (CSSS) were also not included

in the study cohort due to missing information related to a different physician billing structure. See Figure 4.1 for an illustration of the cohort extraction procedure.

4.1.3.1 CMC and levels of medical complexity

Children with CCC that were selected to represent CMC in the study cohort were regrouped in clinically relevant groups based on the CCC body-system sub-classification (neurologic involvement, cardiovascular, respiratory, renal, gastrointestinal, hematologic and immunologic, other congenital or genetic defects, metabolic and malignancy). Children with CCC in the study cohort were divided based on a spectrum of high to low complexity level: NI, multi-CCC and single-CCC, respectively. The groups were mutually exclusive: The NI category included children with CCC for static and progressive neurological and/or functional impairment (included all single-CCC and multi-CCC with neurological involvement); the CCC without NI were separated by using the remaining body system categories, thus CCC affecting more than 1 organ system and CCC affecting a single organ system: multi-CCC and single-CCC, respectively. The 3 groups were further divided according to the need for or absence of technology assistance (TA); TA adding a factor of complexity to its respective group. TA identifies insertion and removal of medical devices required to maintain the health status of a child such as gastrostomy, tracheostomy, and pacemaker among many others.[8 17 33 40 76] Figure 4.2 illustrates the CMC groups and their respective level of medical complexity.

4.1.4 Data collection

For each subject in the cohort the following data were collected:

4.1.4.1 Exposure definition: driving distance to the closest pediatric tertiary care centre

In this study the exposure of interest was defined as the driving distance to the closest of 4 pediatric tertiary care centres in Quebec, Canada (less than 80 kilometres versus 80 kilometres or more). The critical distance selection was based on work from Yantzi *et al* [33] in which 80 kilometres from the hospital was associated with family stress and disruption of the family unit. In order to determine the driving distance for each subject in our study cohort we assigned them a location based on their assigned CLSC. The 3-digit postal code of that CLSC was then used to extrapolate the driving distance to reach all 4 pediatric tertiary care centres in Quebec, Canada. The exact driving distance was determined by using a Geography Information System (SIG) mapping tool (© 2003-2016 Tableau Software All Rights Reserved). The shortest driving distance was then utilized to create a dichotomous variable: CMC living at a driving distance less than 80 kilometres and CMC living at a driving distance of 80 kilometres or more.

4.1.4.2 Outcomes definitions

4.1.4.2.1 Primary outcome: time to readmission within 30 days (also referred to as 30-day readmission)

The primary outcome of this study was time to readmission (in days) within 30 days following discharge from the first index admission. The index admission was defined as the first admission between January 1, 2012 and December 1, 2013 not associated with a death or not preceded by a hospital discharge in the prior 30 days. Hospital-to-hospital transfers or transfers of care to a

different hospital service during index admissions were classified as a continuation of the index admission and not a readmission. Patient locations that were considered as a discharge from an index admission included home, long-term care facilities, rehabilitation centres, CLSC, surgical day care units, medical day care units and psychiatric hospitals. Index admissions less than 24 hours with a procedure or intervention code as primary admission diagnosis were excluded since they most likely represented a scheduled admission for day surgery or minor intervention. Readmission within 30 days was a dichotomous variable described as "Yes or No" depending if it occurred or not for each subject between January 1, 2012 and December 31, 2013. Likewise, index admission was a dichotomous variable described as "Yes or No" depending if it occurred of not for each subject between January 1, 2012 and December 1, 2013 (December 1, 2013 was selected as the limit in our study period so a readmission within 30 days could have the chance to occur until the end of the study period on December 31, 2013). Time to readmission was a continuous variable that represented the numbers of days between the date of discharge from an index admission and the date of readmission within 30 days.

4.1.4.2.2 Secondary outcomes: outpatient and inpatient health care services

Secondary outcomes in this study explored the patterns of health care utilization of CMC and were collected from January 1, 2012 to December 31 2013. Health care services were identified using the Med-Echo database (Appendix 2) and the Fee-for-service physician claims file within the RAMQ database, which includes—amongst other information—specific RAMQ physician billing codes (Appendix 5). The outcomes are regrouped in 2 main categories which include outpatient and inpatient health care services:

1) Inpatient services:

a. having at least one hospital admission (yes/no) and total number of hospital admissions
b. cumulative days in hospital across all hospital admissions

2) Outpatient services:

a. having been seen at least once by a family physician (yes/no) and number of visits
b. having been seen at least once by a pediatrician (yes/no) and number of visits
c. number of distinct physicians providing outpatient care—including all distinct primary and specialty care providers
d. number of outpatient specialist visits
e. number of emergency department visits

4.1.4.3 Covariates

Covariates included age, gender, level of complexity, neighbourhood socio-economic status (SES) and urban versus rural location of residence:

1) Age was determined on January 1, 2012 and was used as a categorical variable divided as follows: 2-4 years, 5-9 years, 10-13 years, and 14-16 years. The categories for the age variable were chosen to match previous research [17 77] and represented 4 clinically relevant groupings—pre-schoolers, early childhood, late childhood and teenagers. The age range starts at 2 years old and ends at 16 years old defined on January 1, 2012 due to the design of the cohort. As shown in the study timeline (Table 4.1.1), this allows for the creation of the cohort using CCC diagnostic codes 2 years before January 1, 2012 as well as allowing for a 2 year outcome window (up until December 31, 2013).

2) Gender was a dichotomous variable with male and female

3) Neighbourhood socio-economic status (SES) was measured by Pampalon's material

deprivation index. [75] This measure is calculated using employment, education and income for each of Statistics Canada's disseminated areas (DA) based on the address of each individual on April 1, 2011 and data from *2011 Census*. Scores for each indicator and by DA are grouped in quintiles to form a neighbourhood SES categories (quintile 1 = most privileged (*Q1*), quintile 5 = most deprived (*Q5*))

4) Residence (urban/rural) was a categorical variable divided into urban, strong metropolitan zone (MIZ), moderate MIZ, weak MIZ and rural. These covariates are based on Statistics Canada's Statistical Area Classification which divides Canadian geographical areas in census metropolitan areas (CMA) census agglomerations (CA) and different metropolitan influenced zones (MIZ). [67] Canada's largest urban centres are characterized as CMA or CA based on population density, while census subdivisions outside of CMA and CA are assigned a MIZ which represents the level of influence from the nearest CMA or CA. In this study, information from this classification was obtained April 1, 2012. CMA and CA were regrouped into the "urban" category; MIZ was categorized as either "strong", "moderate" or "weak", while areas with no MIZ were categorized as "urban".

5) Primary care provider (PCP) was a dichotomous variable based on the presence or absence of a family physician or pediatrician involved in the care of CMC. This PCP covariate was determined on an algorithm used for primary care research by the Institute for clinical evaluative sciences (ICES) in Ontario, Canada [78], which was adapted by *Nakhla and Li.*[79] that uses specific physicians billing codes to identify children in Quebec, Canada, that are enrolled with a primary care provider (Appendix 6).

4.1.4 Databases linkage

The data from the RAMQ and Med-Echo databases were provided in .csv format files, while the driving distance extrapolated from the SIG mapping tool (© 2003-2016 Tableau Software) was provided in an Excel format file (© 2007 Microsoft Office Excel Software). Linking the databases, extracting the cohort of subjects and creating the variables for the exposure, outcomes and covariates were done using R (© R version 3.2.4 software) and SAS (© SAS 9.4 software). To ensure confidentially of the cohort of subjects, their unique RAMQ identifier was removed by the RAMQ and replaced with a new unique identification number that could not be traced back to the original.

4.2 Data analysis

4.2.1 Precision and intervals

Where statistical analysis and hypothesis testing were performed, a 95% confidence interval was calculated using a significance level alpha of 0.05(P <0.05).

4.2.2 Missing values

In this study, unless a death was recorded, subjects were assumed to be covered by the RAMQ as it was a criterion set by the study investigators during the application for the RAMQ database that all subjects be continuously enrolled during the 4 years of the study. Therefore, there were no missing values for all outcomes related to health care services. There were missing values for neighbourhood SES and residence (urban/rural) covariates. These missing values were assumed to be missing at random and analysis was performed on the available data.

4.2.3 Descriptive statistics

4.2.3.1 Proportion of CMC in Quebec, Canada

The proportion of CMC in Quebec was calculated by taking the total number of children that were given a CCC diagnosis on at least 1 hospitalization or 2 physician claims during the period of January 1, 2010 and December 31, 2011. The number of children with the CCC designation was divided by the entire population of children aged 2 to 16 years in Quebec, Canada as of July 1, 2011 (n=1,245,249). [36] This number was provided by Statistics Canada census data and was presented by different age group in Table 4.2.1. Other characteristics of the overall cohort such as proportions of the CMC cohort within each CCC groupings (single-CCC, multi-CCC, NI), need for technology assistance, and subcategories of CCC body-systems (neurologic involvement, cardiovascular, respiratory, renal, gastrointestinal, hematologic and immunologic, other congenital or genetic defects, metabolic and malignancy) were described.

4.2.3.2 Clinical characteristics of the Quebec CMC population

The proportion of children by age group, gender, level of medical complexity (including presence or absence of technology assistance), neighbourhood SES, residence (urban/rural) and presence of primary care provider were computed for the overall cohort, and also according to driving distance to the closest pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more). The denominator used was the number of subjects in each respective driving distance categories.

4.2.3.3 Patterns of health utilization

Measures of health care utilization for CMC according to driving distance to the closest pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more) were compared using chi-square for dichotomous/categorical variables and Mann-Whitney-Wilcoxon test for continuous variables. We reported the medians and IQR for the latter statistics. Percentage frequencies for continuous variables were also presented according to driving distance.

4.2.3.4 Readmission within 30 days

The proportion of CMC with readmission within 30 days was compared according to driving distance to the closest pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more). The denominator used for this proportion was the number of index admissions in each respective driving distance category. Proportions of CMC with a readmission within 30 days were also calculated for each of the following covariates: level of medical complexity, age, gender, neighbourhood SES, residence (urban/rural) and presence of a primary care provider. The denominator used for these proportions was the number of index admissions in each respective category of the covariate in question.

4.2.4 Survival Analysis

4.2.4.1 Kaplan-Meier survival curves and proportional hazard assumption

Kaplan-Meier curves were used along with the log-rank test to compare the probability of 30-day readmission free time following an index admission for CMC living closer to a pediatric tertiary care hospital compared to those farther. [80] All CMC with an index admission were censored at 30 days if no event had occurred. The hypothesis of both curves being the same was rejected if the p-value was below 0.05.

Kaplan-Meier curves were also generated for all covariates included in the hazard model in order to test the proportional hazard assumption. Covariates satisfied this assumption when curves were parallel. Scaled Schoenfeld residuals were also used to test the proportional hazard assumption globally and for each covariate. The proportional hazard assumption was respected when the p-value was below 0.05 as each covariate individually or globally contributed little or no evidence of non-proportionality.

4.2.4.2 Cox proportional hazard model

The Cox proportional hazard model was used to estimate the hazard of time to readmission according to the difference in survival times of CMC living at a driving distance of 80 kilometres or more from a pediatric tertiary care centre compared to those closer.
The hazard rate is the probability of an event, which in this case was a hospital readmission within 30 days following discharge from an index admission, just after time *t*, conditional on survival to time *t* .[81] The Cox model tests for the effect of a defined set of covariates on the event times based on the following equation:

$$h(X, t) = h0(t) \exp \sum_{i=1}^{p} \beta i Xi$$

Where,

h(X, t) hazard function at time *t* for a subject with the set of explanatory variables Xi=1, …, p;

h0(t) unspecified baseline hazard function at time *t* corresponding to a subject with all explanatory variable = 0;

βi logarithm hazard ratio associated with 1 unit increase of the *i*th explanatory variable;

Xi *i*th explanatory variable i=1, …, p. [82]

The model is semiparametric. It generates regression parameters using the maximal partial likelihood criterion for several covariates simultaneously without having to specify the distribution of survival times. Subjects contribute either time-to-event or censored time. For example, in our study subjects could either contribute the number of days until they were readmitted to hospital or if subjects did not experience a readmission, they contributed the number of days before they were censored, i.e., 30 days. Cox proportional hazard regression output is often presented as a hazard ratio which is computed by comparing the hazard rate of a study subject with a particular set of covariates with another study subject with a different set of covariates.

The main assumption of the Cox model is that the hazard ratio is constant over time. In our model this assumption was verified by plotting the Kaplan-Meier survival curves and testing the Schoenfeld residuals as stated in Section 4.2.4.1. [81 82]

4.2.4.3 Cox proportional hazard model for time to readmission within 30 days and association of driving distance to a pediatric tertiary care centre

In our Cox proportional hazard model, the event was the occurrence of a readmission within 30 days of an index admission that occurred at any time point during the period of January 1, 2012 to December 1, 2013. Subjects were censored at 30 days if the event did not occur. The main purpose of this hazard model was to evaluate the association of time to readmission for a driving distance of 80 kilometres or more from the closest pediatric tertiary care centre, compared to those residing closer to that reference point.

4.2.4.3.1 Covariates adjustment

Based on literature review and clinical experience covariates were identified as potential risk factors .[73] These risk factors were evaluated for confounding by examining differences in regression parameters while performing univariate and multivariate analyses and creating a directed acyclic diagram (Figure 4.3). Our final model as shown below included level of medical complexity (CCCTA :0-5), age(Age_cat: 0-3), gender(Gender: 0-1), neighbourhood SES(SES_q: 0-4), residence (urban/rural)(SGC_cat: 0-4) and presence of a primary care provider (PCP: 0-1). See Table 4.2.2-4.2.5 for all variable codes used for statistical computing.

Cox proportional hazard model #1	
t: time h1: 30day_readmin= 1 h0:baseline hazard exp($\beta' x$): log hazard rate for x x:covariates	<u>Full final model:</u> (t, h1) = h0(t) * exp (*β1*(Distance=1) + *β2*(CCCTA_cat=1) + *β3*(CCCTA_cat=2) + *β4*(CCCTA_cat=3) + *β5*(CCCTA_cat=4) + *β6*(CCCTA_cat=5) + *β7*(Age_cat=1) + *β8*(Age_cat=2) + *β9*(Age_cat=3) + *β10*(Gender=1) + *B11*(SES_q=1) + *B12*(SES_q=2) + *B13*(SES_q=3) + *B14*(SES_q=4) + *β15*(SGC_cat=1) + *β16*(SGC_cat=2) + *β17*(SGC_cat=3) + *β18*(SGC_cat=4) + *β19*(PCP=1) reference category for each covariate coded as $x=0$

4.2.4.4 Hazard model for time to readmission within 30 days and with interaction terms

In order to test whether the association between a driving distance of 80 kilometres or more from a pediatric tertiary care centre and the rate of hospital readmission varied by level of medical complexity we included interaction terms in the final model.

Cox Proportional Hazard Model #2	
t: time h1: 30day_readmin= 1 h0:baseline hazard exp($\beta' x$): log hazard rate for x x: covariates	<u>Interaction model with interaction terms (**in bold**)</u> (*t*, h1) = h*o*(*t*) * exp (*β1*(Distance=1) + *β2*(CCCTA_cat=1) + *β3*(CCCTA_cat=2) + *β4*(CCCTA_cat=3) + *β5*(CCCTA_cat=4) + *β6*(CCCTA_cat=5) + *β7*(Age_cat=1) + *β8*(Age_cat=2) + *β9*(Age_cat=3) + *β10*(Gender=1) + *B11*(SES_q=1) + *B12*(SES_q=2) + *B13*(SES_q=3) + *B14*(SES_q=4) + *β15*(SGC_cat=1) + *β16*(SGC_cat=2) + *β17*(SGC_cat=3) + *β18*(SGC_cat=4) + *β19*(PCP=1) + ***β20*(CCCTA_cat=1*Distance=1) + *β21*(CCCTA_cat=2*Distance=1) + *β22*(CCCTA_cat=3*Distance=1) + *β23*(CCCTA_cat=4*Distance=1) + *β24*(CCCTA_cat=5 *Distance=1)** reference category for each covariate coded as $x=0$

4.2.4.4 Estimate of number of events needed

Based on previously published data, 22% of CMC lived at a distance of 80 kilometres or more from a tertiary care pediatric centre. [66] Additionally, CMC that only have a follow-up visit in the first 3 days post discharge, which is often the case for children living remotely, have an increase odds of readmission of 1.67. [42] Taking the latter information into account, 197 events were needed (alpha 0.05 and beta 0.2) in order to detect an association between the 2 groups based on driving distance to specialized care when performing survival analysis. [83]

Figures and Tables (Chapter 4)

Table 4.1.1 Study timeline

Study events	Year 1 Jan1-Dec31, 2010	Year 2 Jan1-Dec31, 2011	Year 3 Jan1-Dec31, 2012	Year 4 Jan1-Dec31, 2013
Study cohort extraction				
Determination of baseline characteristics:				
Neighbourhood SES		←Apr 1, 2011		
Level of medical complexity, age, gender			←Jan 1, 2012	
Residence (urban/rural)			←Apr 1, 2012	
Health care utilization outcomes:				
Index admissions*				
30-day readmissions				
Remaining health care utilization outcomes				

*Time period for extraction of index admission: January 1, 2012 to December 1 2013

January (Jan), December (Dec), April (Apr)

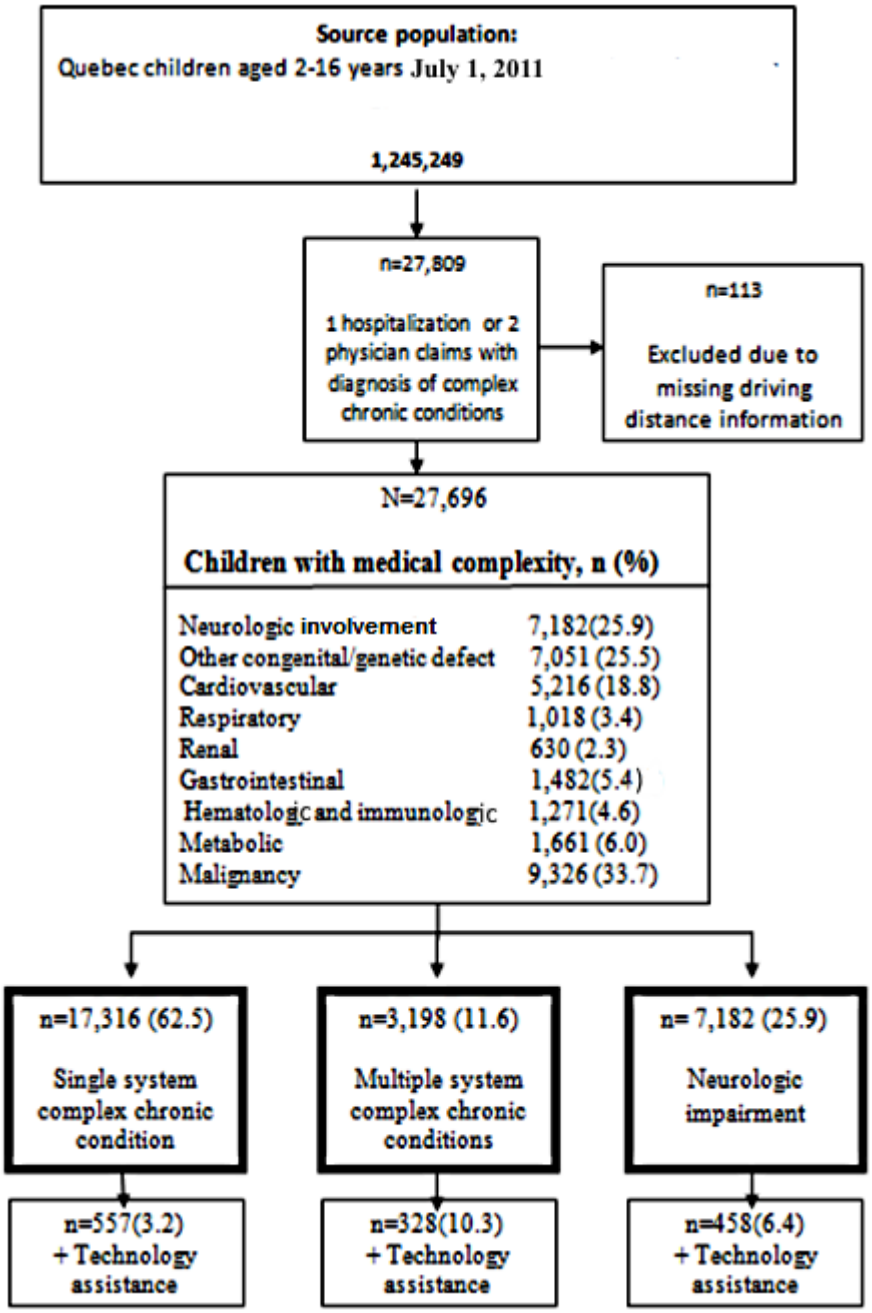

Figure 4.1 Study cohort extraction

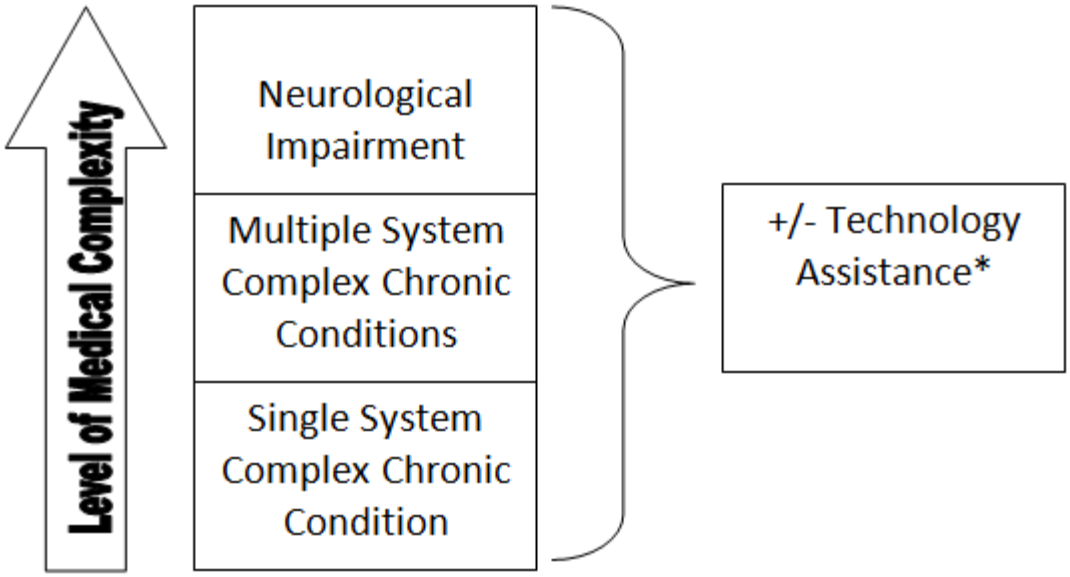

Figure 4.2 Schema of CMC study cohort by level of medical complexity

*Technology assistance representing an added factor of medical complexity to its respective CMC category

Children with medical complexity (CMC)

Table 4.2.1 Population of Quebec children and teenagers by age group, July 1 2011[36]	
Age(years)	**Population(n)**
2 to 4	260,422
5 to 9	389,951
10 to 13	319,012
14 to 16	275,864
Total	1,245,249

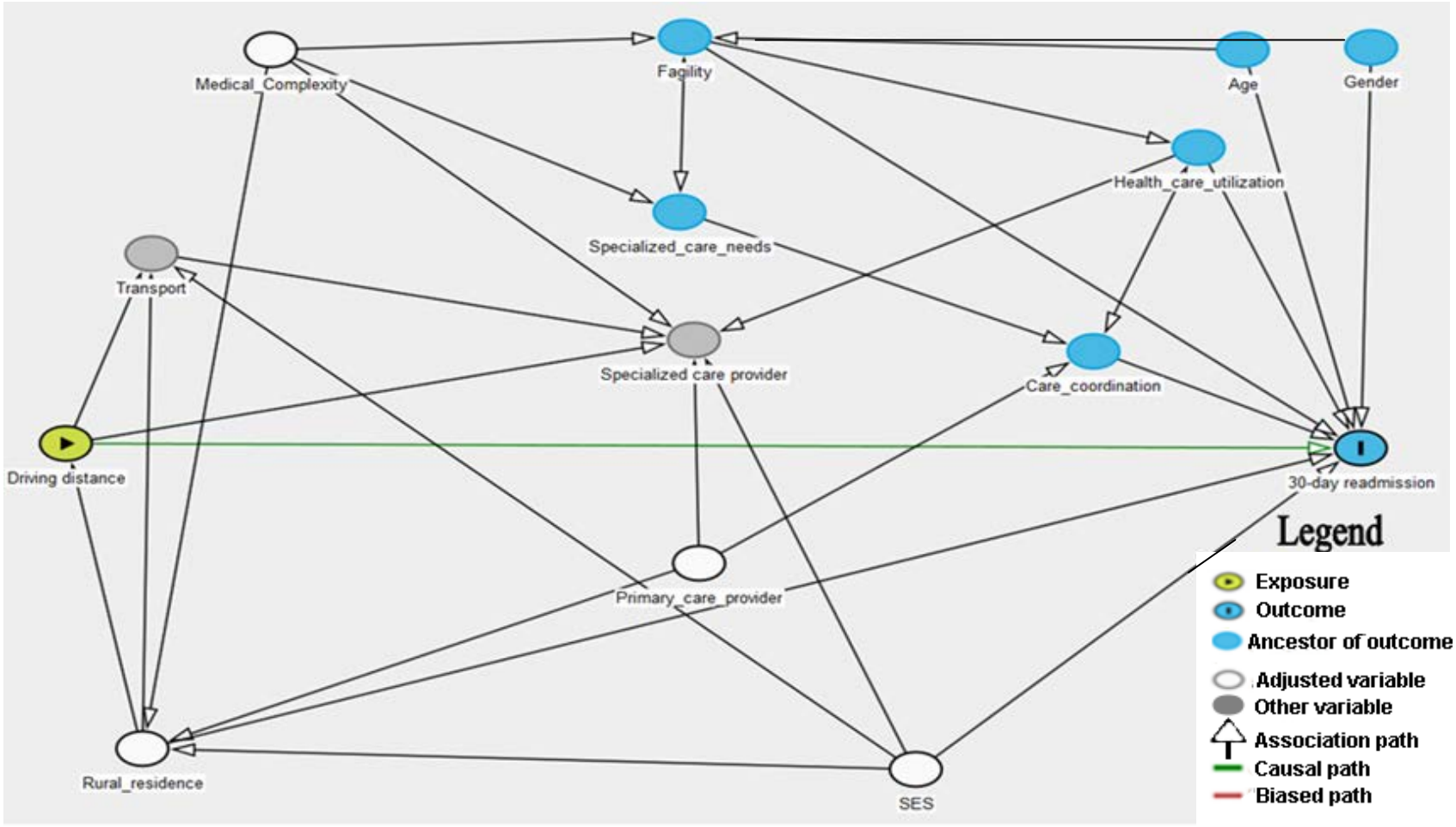

Figure 4.3 Directed Acyclic Graph of the hypothesized association of a driving distance of 80 kilometres or more from a pediatric tertiary care centre on time to 30-day readmission.

Neighbourhood SES (SES)

Table 4.2.2 Statistical computing coding for exposure variable: driving distance to the closest tertiary care centre

Variable Description	Database Name	Coding	Type
Driving distance to pediatric hospital	Distance	<80 kilometres=0 ≥80 kilometres=1	Dichotomous

Table 4.2.3 Statistical computing coding for CMC groups and level of medical complexity

Variable Description	Database Name	Coding	Type
CMC categories	CCC_cat	Single-CCC=0 Multiple-CCC=1 NI=2	Categorical
Technology Assistance	TA	Presence=1 Absence=0	Dichotomous
Level of Medical Complexity + TA	CCCTA_cat	Single-CCC + No TA=0 Single-CCC + TA = 1 Multi-CCC + No TA =2 Multi-CCC + TA=3 NI + No TA = 4 NI + TA = 5	Categorical

Children with medical complexity(CMC), complex chronic condition (CCC), neurologic impairment (NI), technology assistance (TA)

Table 4.2.4 Statistical computing coding for demographic characteristics

Variable Description	Database Name	Coding	Type
Age Category	Age_cat	2-4 years=0 5-9 years =1 10-13 years =2 14-16years =3	Categorical
Gender	Gender	Female=0 Male=1	Dichotomous
Neighbourhood SES	SES_q	Q1=0 Q2=1 Q3=2 Q4=3 Q5=4	Categorical
Urban versus Rural residence	SGC_cat	Urban=0 Strong MIZ=1 Moderate MIZ=2 Weak MIZ=3 Rural=4	Categorical
Presence of a PCP	PCP	No=0 Yes =1	Dichotomous

Children with medical complexity(CMC), neighbourhood socioeconomical status (SES), census metropolitan areas (CMA), quintiles of 20% of population (Q1-Q5), census agglomeration (CA), metropolitan influenced zones(MIZ), primary care provider (PCP)

Table 4.2.5 Statistical coding for outcome measures for health care utilization services

Variable Description	Database Name	Coding	Type
30-day readmission	Readmin_30day	Yes=1 No=0	Dichotomous
Index Admission	Index_admin	Yes=1 No=0	Dichotomous
Hospital stays	Hosp_stay	Number of hospital stays	Continuous
Cummulative hospital days	Hosp_days	Total number of days in hospital	Continuous
Family physician	FP	Yes=1 No=0	Dichotomous
Family physician visits	FP_visits	Number of visits	Continuous
Pediatrician	Peds	Yes=1 No=0	Dichotomous
Pediatrician visits	Peds_visits	Number of visits	Continuous
Specialists visits	Spec_visits	Number of visits	Continuous
Distinct physicians	Distinct_phys	Number of physicians	Continuous
Emergency Department visits	ED_visits	Number of visits	Continuous

Chapter 5: Results

5.1 Description of the CMC cohort (Figure 4.1)

In total there were 27,696 subjects in our study sample between the age of 2 and 16 years on January 1, 2012. Using census information for the denominator of the population [36], CMC constituted 2.2% of the total population of children aged 2 to 16 years in Quebec. The median age was 11 years (IQR 8) and 51.2% were female (Table 5.1). Within the group of CMC, 62.5% were identified as single-complex chronic condition (CCC), 11.6% as multi-CCC, and 25.9% as neurologically impaired (NI), while 4.9% of CMC were dependent on technology assistance (TA). In decreasing order, the cohort of CMC included all of the 9 body-systems of Feudtner's CCC subgroups (subgroups are not mutually exclusive as patients could have involvement of more than 1 body-systems): malignancy (33.7%), neurological involvement (25.9%), congenital or genetic defects (25.5%), cardiovascular disorders (18.7%), metabolic disorders (6.0%), gastrointestinal disorders (5.4%), hematologic or immunologic disorders (4.6%), respiratory disorders (3.4%) and renal disorders (2.3%).

5.2 Clinical characteristics of CMC by driving distance to a pediatric tertiary health care centre (less than 80 kilometres versus 80 kilometres or more) (Table 5.1)

When our CMC cohort was divided according to the exposure, driving distance to the closest pediatric tertiary care hospital, we found that 221,151 subjects (76.4%) resided within 80 kilometres, while 6,546 subjects (23.6%) lived beyond that distance. The proportions of CMC according to level of medical complexity and driving distance are displayed in Figure 5.1. Demographic characteristics were similar in the 2 driving

distance categories, except for neighbourhood SES and residence (urban/rural). Neighbourhood SES had an inverse distribution of quintiles according to driving distance. CMC residing farther from a pediatric tertiary care center had the largest proportion of children in the lowest quintile (22.0 %) while CMC residing closer had the largest proportion of children in the highest quintile (24.0%). Likewise, CMC residing farther had a larger proportion living in rural areas compare to those residing closer (10.6% versus 1.5%, respectively) and a lower proportion living in urban areas (49.9% versus 81.3%, respectively).

Table 5.1 Characteristics of CMC by driving distance to a pediatric tertiary care centre			
Variables	**Overall n (%)**	**<80km driving distance n (%)**	**≥80km driving distance n (%)**
Total	27696	21151(76.4)	6545(23.6)
Age group (years)			
2-4	5643(20.4)	4322(20.4)	1321(20.2)
5-9	7245(26.1)	5571(26.3)	1674(25.6)
10-13	7222(26.1)	5493(26.0)	1729(26.4)
14-16	7586(27.4)	5765(27.3)	1821(27.8)
Gender, female	14169(51.2)	10790(51.0)	3379(51.6)
Level of medical complexity			
Single CCC			
With TA	557(2.0)	416(2.0)	141(2.1)
Without TA	16759(60.5)	12753(60.3)	4006(61.2)
Multiple CCC			
With TA	329(1.2)	246(1.2)	83(1.3)
Without TA	2869(10.4)	2258(10.7)	611(9.3)
NI			
With TA	461(1.7)	361(1.7)	100(1.5)
Without TA	6721(24.3)	5117(24.2)	1604(24.5)
Neighbourhood SES			
Q1 (highest)	5968(21.5)	4962(24.0)	1006(15.8)
Q2	5555(20.0)	4225(20.4)	1330(20.9)
Q3	5357(19.3)	4006(19.4)	1351(21.2)
Q4	5095(18.4)	3818(18.5)	1277(20.1)
Q5 (lowest)	5063(18.3)	3662(17.7)	1401(22.0)
Residence(urban/rural)*			
Urban	20420(73.7)	17161(81.3)	3259(49.9)
Strong MIZ	2253(8.1)	919(4.4)	1334(20.4)
Moderate MIZ	1672(6.0)	1304(6.2)	368(5.6)
Weak MIZ	2286(8.3)	1405(6.7)	8811(13.5)
Rural	1016(3.7)	325(1.5)	691(10.6)
PCP*, yes	18207(65.7)	14016(66.3)	4191(64.0)

* See **Section 4.1.4.3** in text for definitions of neighbourhood SES, residence and primary care provider

Children with medical complexity (CMC), complex chronic condition (CCC), technology assistance (TA), neurologic impairment (NI), neighbourhood socio-economic status(SES), quintiles(Q), metropolitan influenced zones(MIZ), primary care provider(PCP)

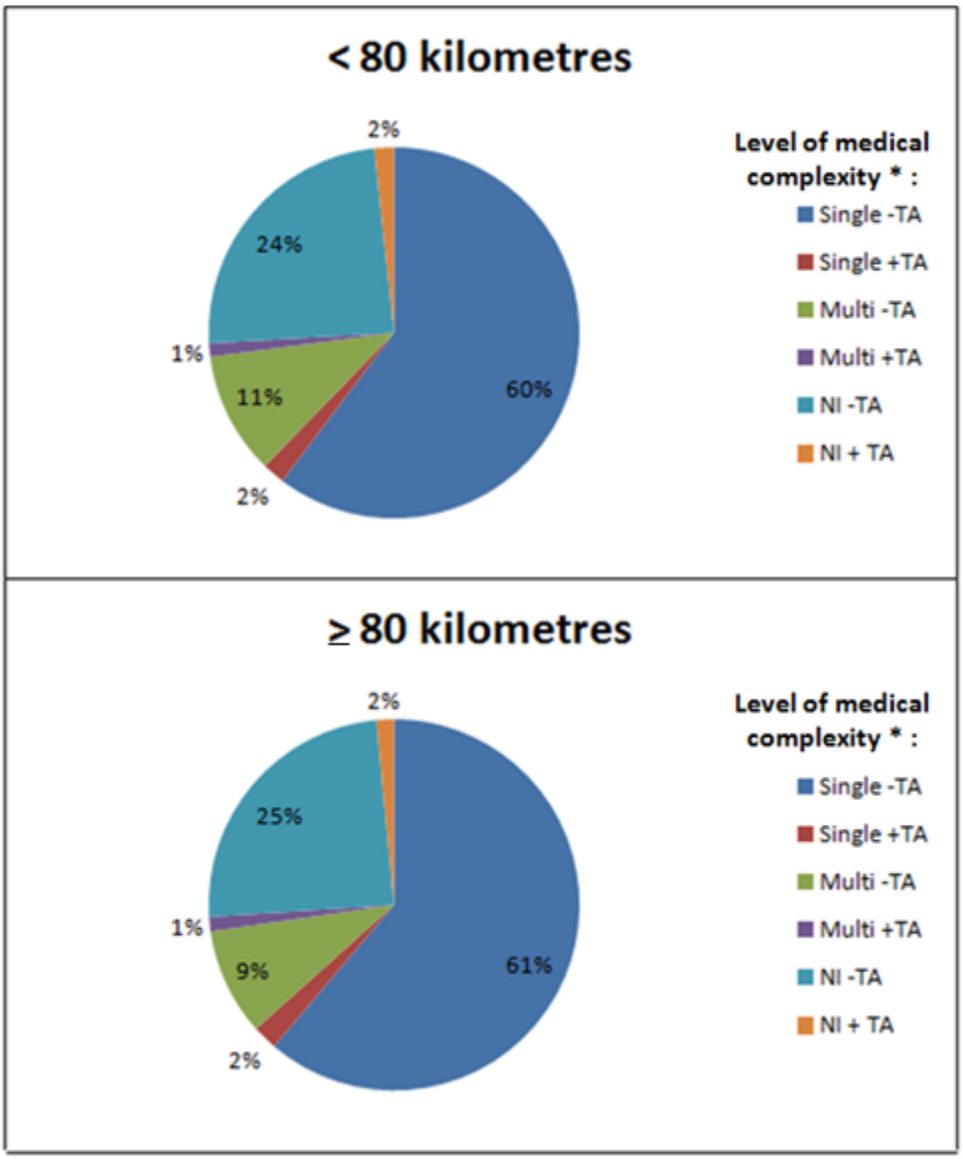

Figure 5.1 Distribution of CMC cohort by level of medical complexity according to driving distance to a pediatric tertiary care hospital

*Single system complex chronic condition (CCC) without technology assistance (Single –TA); single system CCC with TA (Single +TA); multiple system complex CCC without TA (Multi –TA); multiple system complex CCC with TA (Multi +TA); neurologic impairment (NI) CCC without TA (NI –TA); NI CCC with TA (Multi +TA)

5.3 Patterns of healthcare utilization of CMC by driving distance to a pediatric tertiary care centre (less than 80 kilometres versus 80 kilometres or more) (Table 5.2)

Health care services were compared according to driving distance to the closest pediatric tertiary care (less than 80 kilometres versus 80 kilometres or more) in the time period from January 1, 2012 and December 31, 2013.

5.3.1 30-day readmission (primary outcome) and driving distance to the closest pediatric tertiary care centre

Overall, within our CMC cohort, there was a total of 6,724 (24.3%) subjects with an index admission (at least 1 hospitalization during the study outcome period). As shown in Table 5.2, there was a significant difference between the proportion of CMC that experienced an index admission, living closer (23.9%) compared to living farther (25.5%). A total of 514 (7.6%) subjects with index admissions had a subsequent readmission within 30 days of their discharge. There was no significant difference between the readmission rate for CMC living closer versus those living farther (7.5% vs. 8.1%, respectively).

5.3.2 Inpatient and outpatient health services utilization (secondary outcomes) according to driving distance to a pediatric tertiary care centre

Inpatient and outpatient health services use was explored according to driving distance during the 2-year study outcome period included total number of hospital admissions, cumulative days in hospital, proportion seen by a family physician and number of visits, proportion seen by a pediatrician and number of visits, number of distinct physicians, number of specialist visits, as well as number of emergency department visits (Table 5.2).

In terms of inpatient services, there was a significant difference between the number of hospital admissions and no significant difference for cumulative days in hospital between those living closer and further from a pediatric tertiary care centre. Overall, a larger percentage of CMC living at 80 kilometres or more to pediatric tertiary care centre had at least 1 hospital admissions (index admission). Moreover, when looking at the distribution of percentage frequencies a smaller percentage of CMC living farther had 1 or 2 hospital admissions but a larger percentage of these CMC had 3 or more hospital admissions compared to those living closer (Figure 5.2).

All findings for outpatient services showed significant differences between driving distance categories (Table 5.2). The distributions of family physician visits, pediatrician visits, number of distinct physicians, specialty visits and emergency department visits are presented as percentage frequencies in Figure 5.4, 5.5, 5.6, 5.7 and 5.8 respectively. In general, a smaller percentage of CMC living farther to a pediatric tertiary care centre were seen at least once by a family physician or pediatrician compared to those closer. When examining percentage frequencies, a larger percentage of CMC living farther had 1 or 2 visits to the family physician and 1-6 visits to the pediatrician, but a smaller percentage of CMC living farther had visits above the latter thresholds, compared to those living closer. Likewise, CMC living farther had less visits with specialists when the frequency exceeded 6 visits compared to those living close. However, percentage frequencies for emergency department visits, generally showed, those living farther had a larger percentage of CMC with a frequency of 3 emergency department visits or more.

Table 5.2 Health care utilization of CMC according to driving distance to a pediatric tertiary care centre			
Health services outcomes	**<80km driving distance (n= 21151)**	**≥80km driving distance (n=6545)**	**p-value****
Inpatient Proportion with 30-day readmission, n (%)*	379(7.5)	135(8.1)	0.4
Proportion with ≥ 1 hospital admission (index admissions), n(%)	**5054(23.9)**	**1670(25.5)**	**0.008**
Number of admissions, median (IQR)	**1(1-2)**	**1(1-2)**	**0.02**
Cumulative days is hospital, median (IQR)	2(1-6)	2(1-6)	0.6
Outpatient Proportion with FP visits, n (%)	**12763(60.3)**	**3542(54.1)**	**<0.001**
Median number of FP visits, (IQR)	**2(1-4)**	**2(1-3)**	**<0.001**
Proportion with pediatrician visits, n (%)	**18449(87.2)**	**5549(84.8)**	**<0.001**
Median number of pediatrician visits, (IQR)	**4(2-8)**	**4(2-7)**	**<0.001**
Median number of distinct physicians, (IQR)	**3(2-6)**	**3(2-5)**	**<0.001**
Median number of specialists visits, (IQR)	**4(2-8)**	**4(2-7)**	**<0.001**
Median number of emergency department visits, (IQR)	**2(1-3)**	**2(1-3)**	**<0.001**

*Denominator =total number of index hospitalizations in each driving distance category; for <80km, n=5054 for ≥80km, n= 1670

**Significance testing done with chi-square test for categorical variables and Mann-Whitney-Wilcoxon test for continuous variabes

Children with medical complexity (CMC), interquartile range (IQR), family physician (FP)

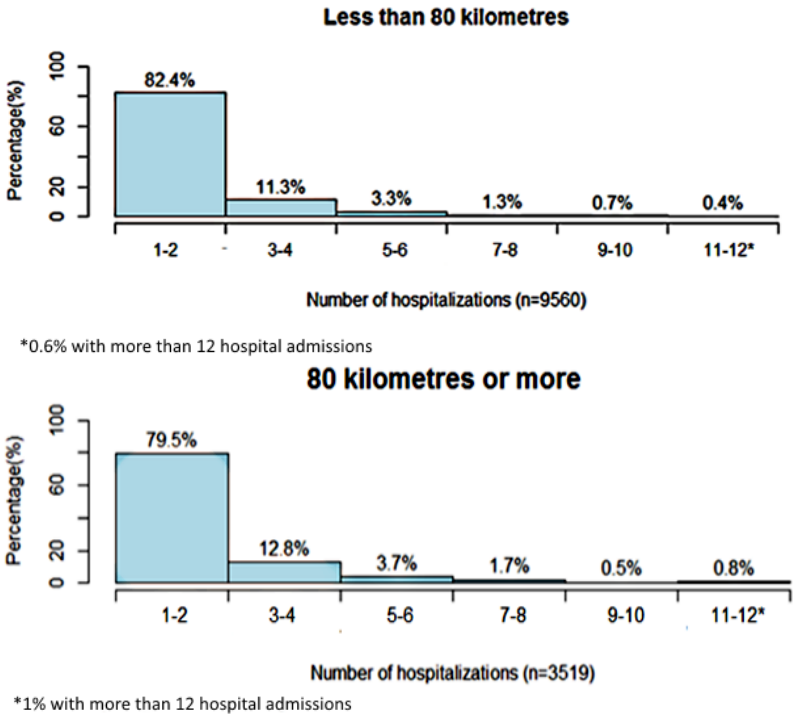

Figure 5.2 Percentage frequencies of hospital admissions in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

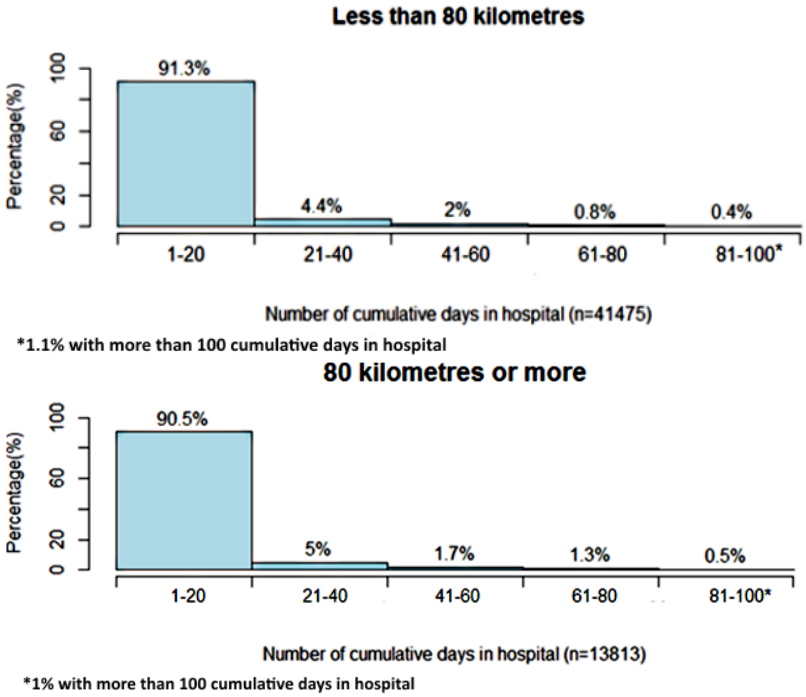

Figure 5.3 Percentage frequencies of cumulative days in hospital in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

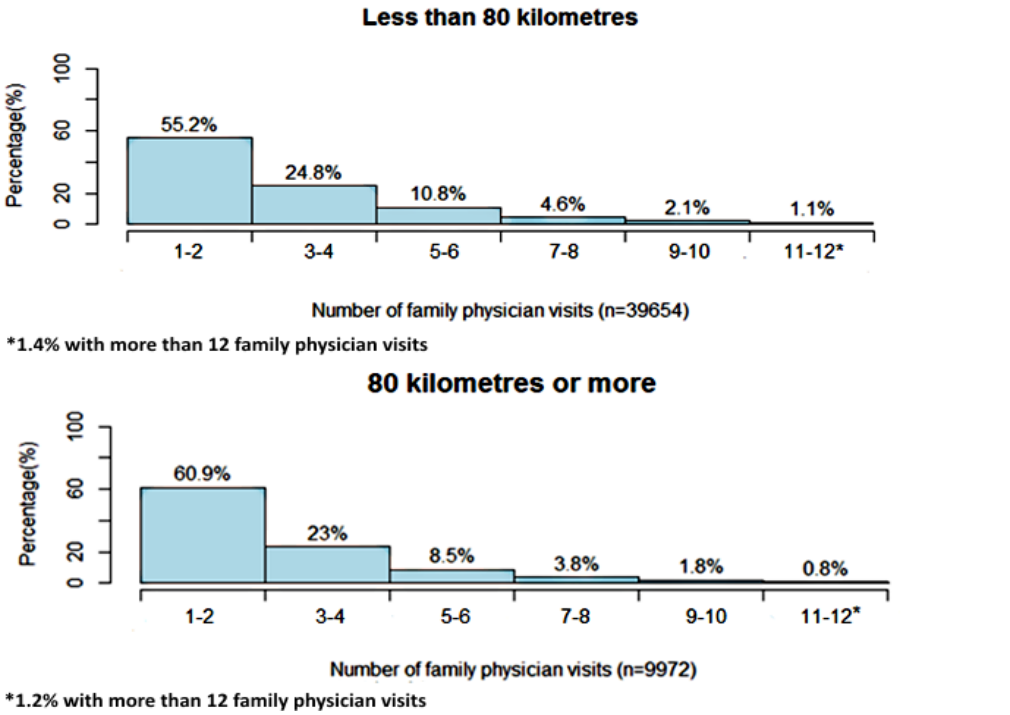

Figure 5.4 Percentage frequencies of family physician visits in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

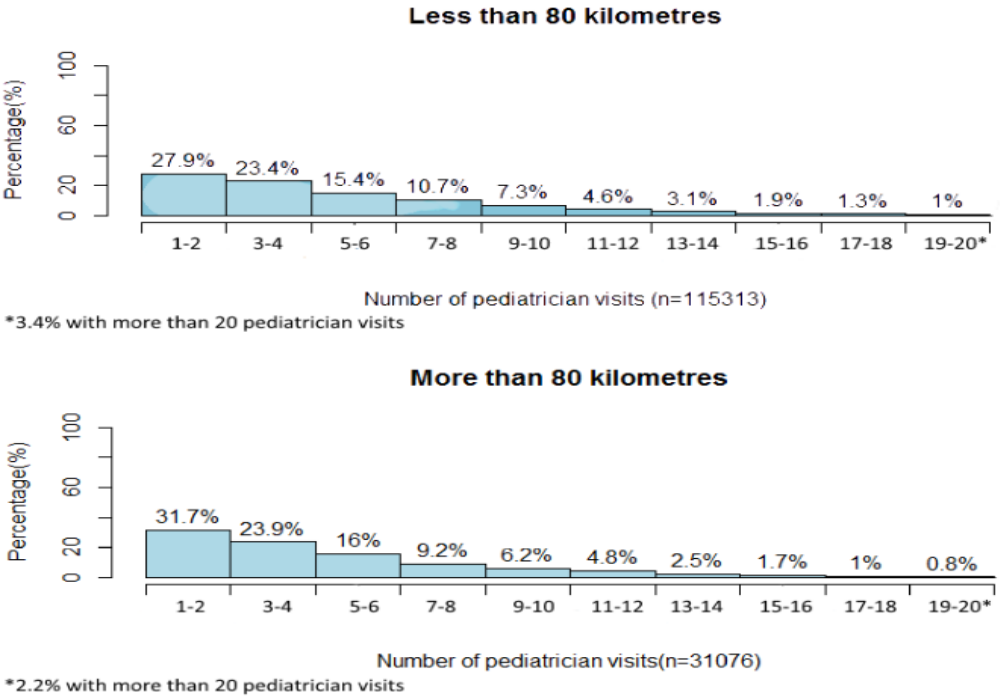

Figure 5.5 Percentage frequencies of number of pediatrician visits in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

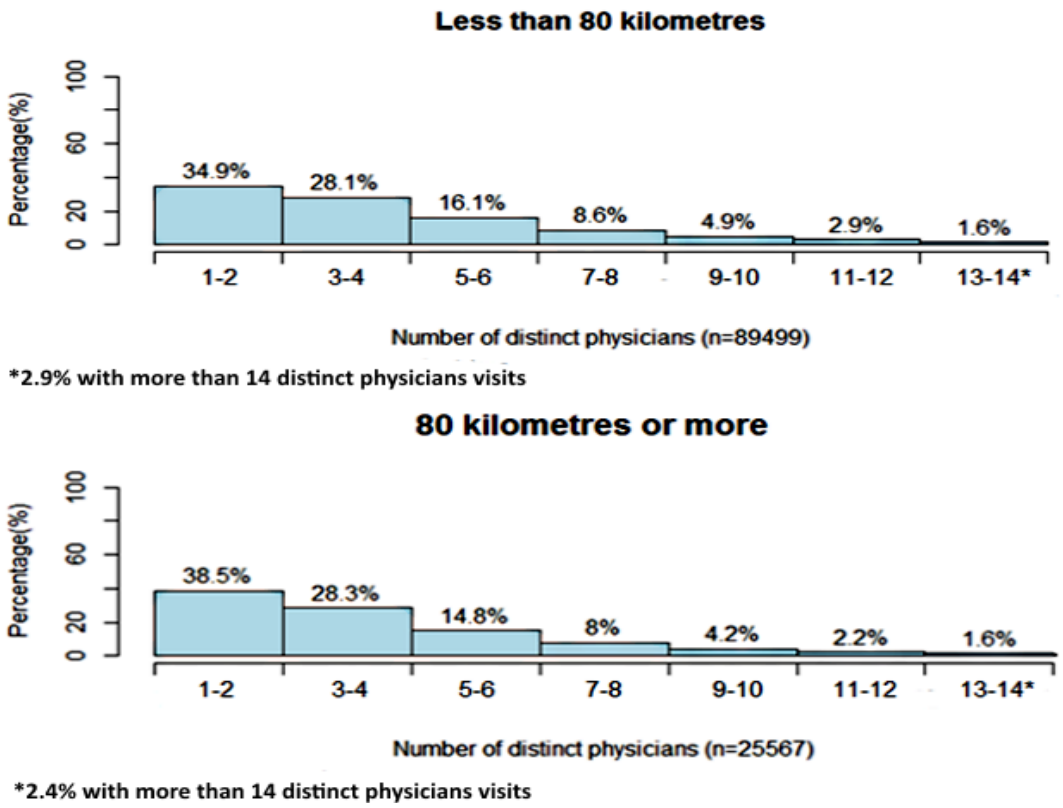

Figure 5.6 Percentage frequencies of number of distinct physicians in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

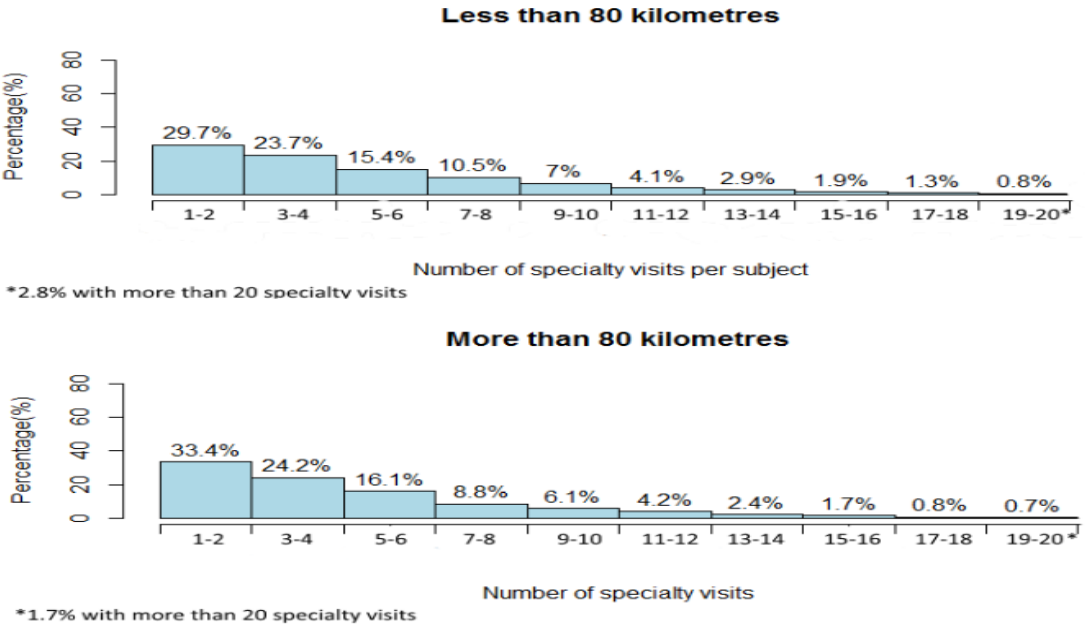

Figure 5.7 Percentage frequencies of number of specialist visits in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

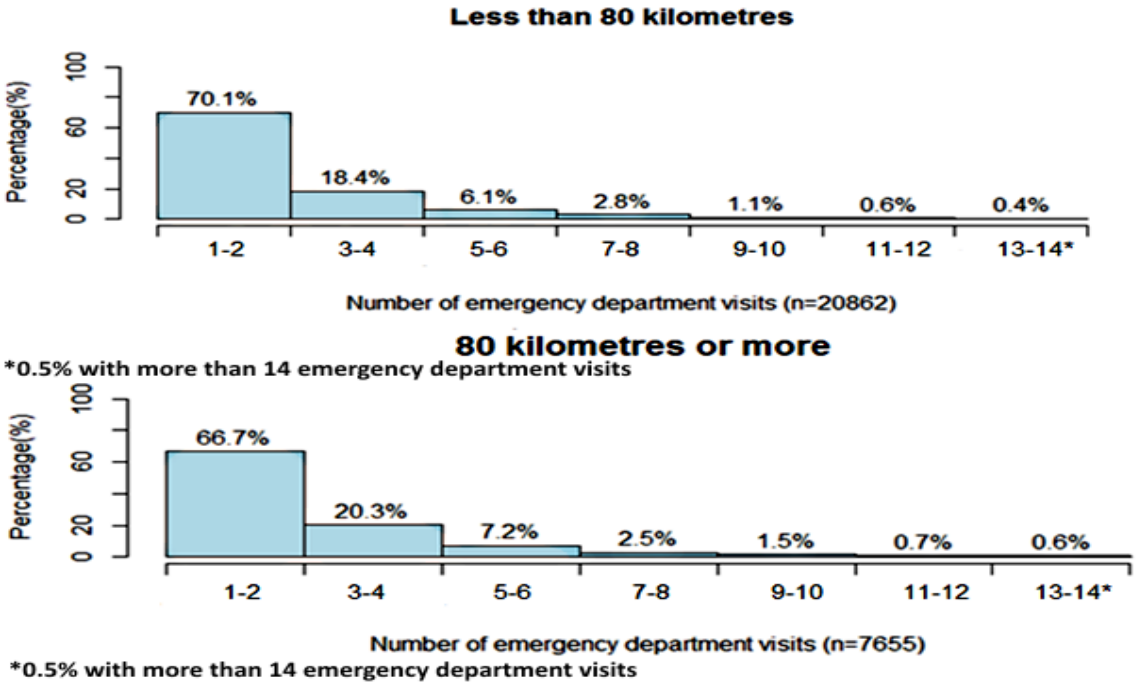

Figure 5.8 Percentage frequencies of emergency department visits in children with medical complexity over 2 years according to distance to a pediatric tertiary care centre

5.4 Hazard models for time to readmission within 30 days

See Appendices 8-11 for exact R statistical software output for logrank test, Cox proportional hazard model #1, Schoenfeld residuals and Cox proportional hazard model #2 (including interaction terms)

5.4.1 Association between time to readmission within 30 days and driving distance to a pediatric tertiary care centre

The Kaplan-Meier curves for 30-day readmission free probability by driving distance (without taking into account any other predictors) are depicted in Figure 5.9. The logrank statistic was 0.6 with a p-value of 0.43 indicating no significant difference between those 2 groups. The Kaplan-Meier curves were also used to test for proportionality for all covariates (driving distance to a pediatric tertiary care centre, level of medical complexity, age, gender, neighbourhood SES, residence (urban/rural) and presence of a primary care provider) included in the Cox proportional hazard model. We could appreciate that the curves seemed to satisfy the proportional hazard assumption as they were generally parallel (Figures 5.9-5.15). We also used the Scaled Schoenfeld residuals over time to test the proportional hazard assumption. P-value for individual covariates and globally was below 0.05 which signifies the assumption holds since they contributed little or no evidence of non-proportionality.

The final Cox proportional hazard model for time to readmission within 30 days included driving distance, level of medical complexity, age, gender, neighbourhood SES, residence (urban/rural) and presence of PCP. Proportions of 30-day readmissions for each covariate above, (denominator equal to the number of index hospitalizations in the respective category of each covariate), unadjusted and adjusted hazard ratios are presented in Table 5.3. Driving distance

was not associated with time to readmission within 30 days (adjusted hazard ratio of 1.07, 95% CI of 0.86-1.34, for distance ≥80km). However, the adjusted model showed statistically significant higher hazard ratios for all categories of medical complexity compared to single CCC without TA, including multi-CCC with TA, which had the highest hazard ratio at 3.12 (95% CI of 2.14-4.55). The 4th neighbourhood SES quintile was also statistically significant with a hazard ratio of 1.52 (95% CI of 1.13-2.05).

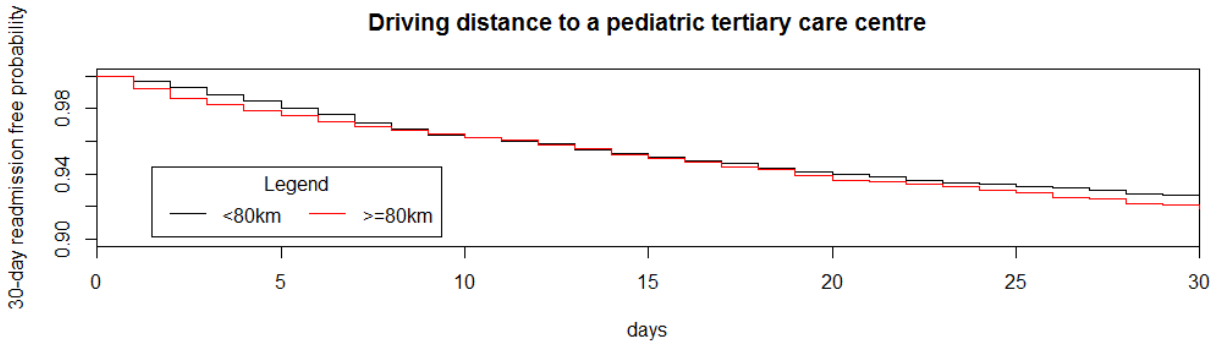

Figure 5.9 Kaplan-Meier curves for 30-day readmission free probability following index admission for *driving distance* covariate in hazard model

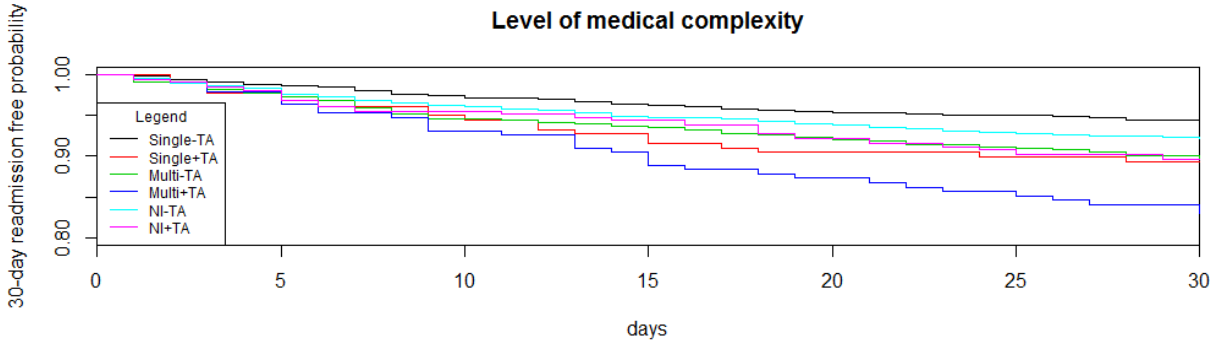

Figure 5.10 Kaplan-Meier curves for 30-day readmission free probability following index admission for *level of medical complexity covariate* in hazard model

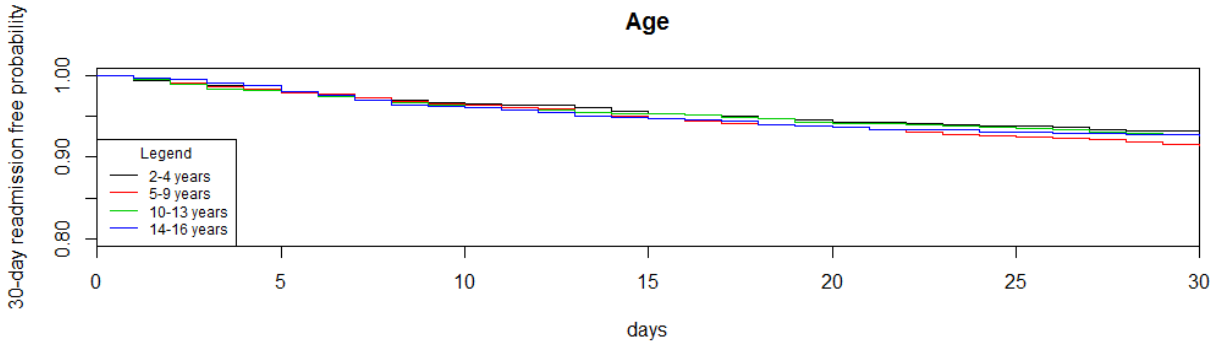

Figure 5.11 Kaplan-Meier curves for 30-day readmission free probability following index admission for *age* covariate in hazard model

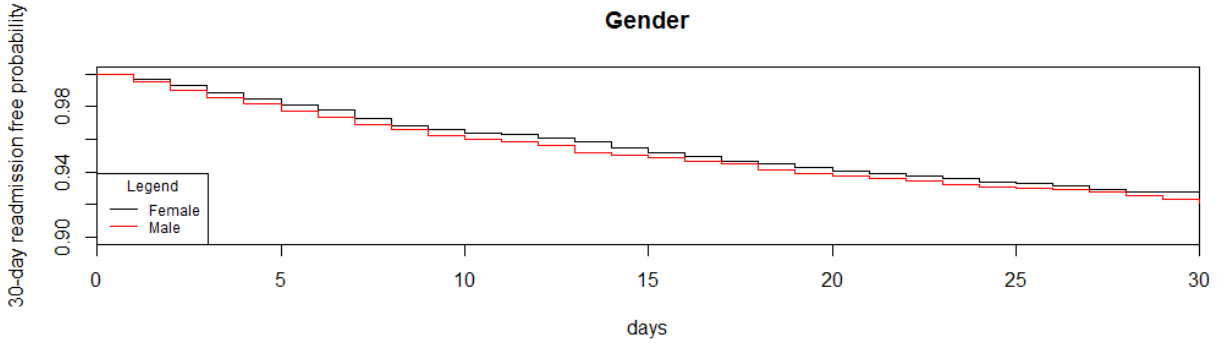

Figure 5.12 Kaplan-Meier curves for 30-day readmission free probability following index admission for *gender* covariate in hazard model

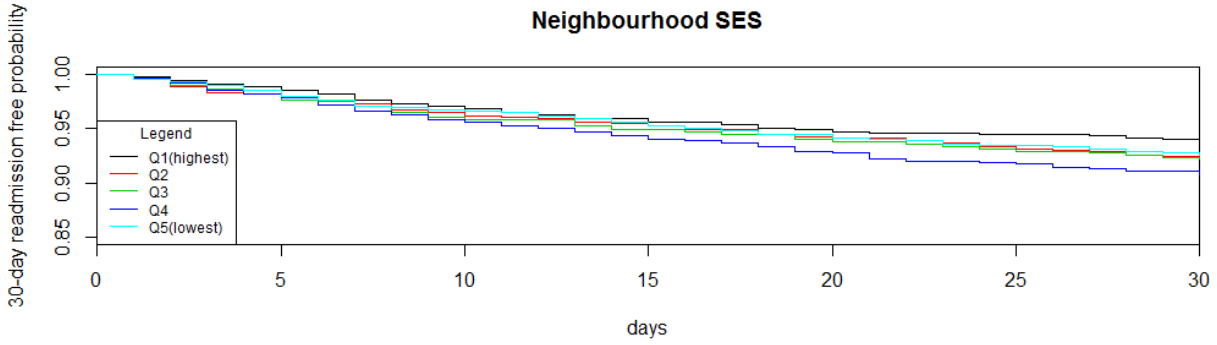

Figure 5.13 Kaplan-Meier curves for 30-day readmission free probability following index admission for *neighbourhood SES* covariate in hazard model

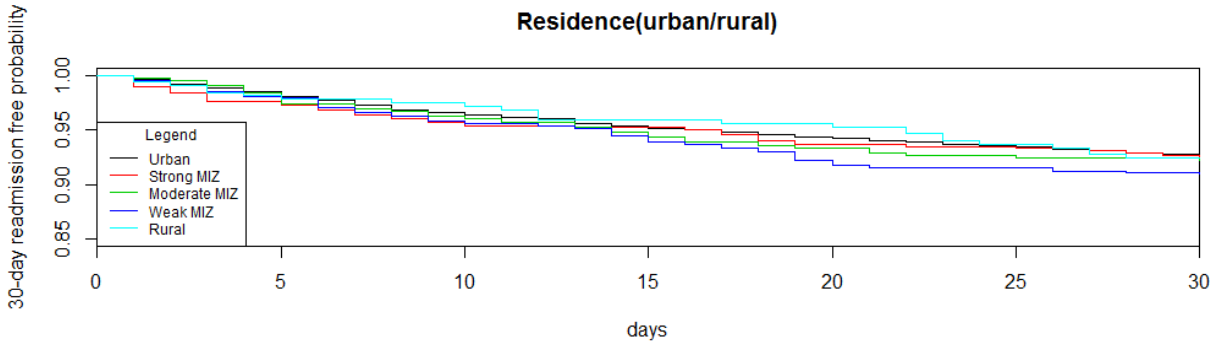

Figure 5.14 Kaplan-Meier curves for 30-day readmission free probability following index admission for *residence (urban/rural)* covariate in hazard model

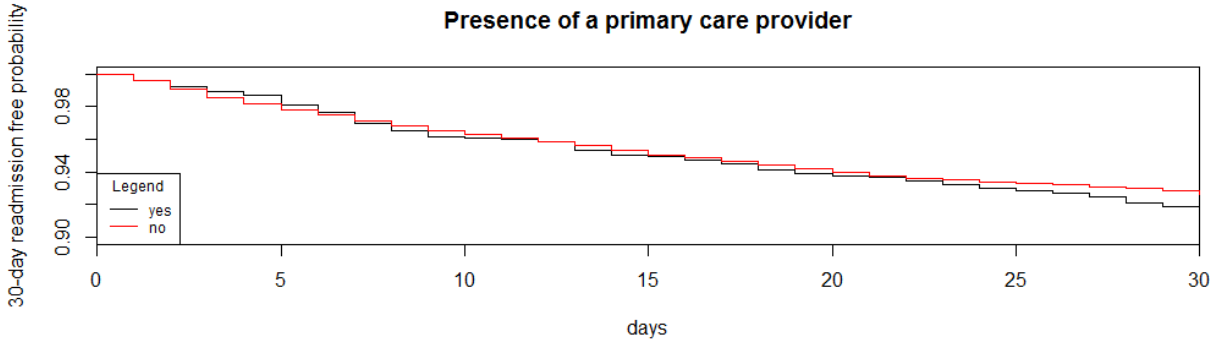

Figure 5.15 Kaplan-Meier curves for 30-day readmission free probability following index admission for *presence of primary care provider covariate* in hazard model

Table 5.3 Unadjusted and adjusted hazard ratios of time to readmission within 30-days (N=514) following index hospitalization (N=6724) according to driving distance to a pediatric tertiary care centre			
Variables	**30-day Readmissions n, (%)**	**Unadjusted HR (95% CI)**	**Adjusted HR (95% CI)**
Distance (kilometres)			
<80	379(7.5)	Reference	Reference
≥80	135(8.1)	1.08(0.89-1.32)	1.07(0.86-1.34)
Level of Medical Complexity			
Single without TA	185(5.8)	Reference	Reference
Single with TA	19(10.7)	**1.89(1.18-3.04)**	**1.77(1.08-2.88)**
Multi without TA	32(10.2)	**1.80(1.40-2.32)**	**1.84(1.43-2.37)**
Multi with TA	91(16.8)	**3.05(2.10-4.44)**	**3.12(2.14-4.55)**
NI without TA	154(10.7)	**1.36(1.10-1.68**	**1.33(1.07-1.66)**
NI with TA	33(7.8)	**1.89(1.31-2.74)**	**1.75(1.19-2.58)**
Age (years)			
2-4	94(6.9)	Reference	Reference
5-9	166(8.5)	1.25(0.97-1.61)	1.27(0.99-1.61)
10-13	103(7.7)	1.13(0.85-1.49)	1.11(0.85-1.45)
14-16	151(7.3)	1.08(0.83- 1.39)	1.15(0.89-1.48)
Gender			
Female	243(7.4)	Reference	Reference
Male	271(7.9)	1.07(0.90-1.27)	1.05(0.88-1.25)
Neighbourhood SES*			
Q1(highest)	76(6.0)	Reference	Reference
Q2	99(7.8)	1.30(0.97-1.76)	1.30(0.96-1.76)
Q3	103(7.8)	1.30(0.97-1.75)	1.30(0.96-1.75)
Q4	118(9.2)	**1.55(1.16-2.07)**	**1.52(1.13-2.05)**
Q5(lowest)	106 (7.4)	1.24(0.92- 1.66)	1.22(0.89-1.66)
Residence(urban/rural)*			
Urban	352(7.5)	Reference	Reference
Strong MIZ	47(7.5)	1.01(0.75- 1.37)	0.91(0.65-1.26)
Moderate MIZ	33(7.8)	1.05(0.74 -1.51)	0.99(0.69-1.43)
Weak MIZ	56(9.1)	1.23(0.93-1.63)	1.11(0.82-1.50)
Rural	24(7.6)	1.01(0.67- 1.53)	0.85(0.54-1.36)
PCP*			
Yes	198 (8.2)	Reference	Reference
No	316(8.2)	0.89(0.75-1.07)	0.93(0.76-1.12)

* See **Section 4.1.4.3** in text for definitions of neighbourhood SES, residence and primary care provider

Hazard ratio (HR), confidence interval (CI), complex chronic condition (CCC), technology assistance (TA), neurologic impairment (NI), socio-economic status (SES), quintiles (Q1-5), metropolitan influenced zones (MIZ), primary care provider(PCP)

5.4.2 Level of medical complexity and travel distance as an effect measure modifier of time to 30-day hospital readmission

A Cox proportional hazard regression model for time to 30-day readmission with interaction terms for driving distance and level of medical complexity was used to assess if there was a measure modification effect between those covariates. All variables from the previous final Cox model were also included (age, gender, neighbourhood SES, residence (urban/rural) and presence of PCP). As seen in Table 5.4, none of the interaction terms were statistically significant. For information on all hazard ratios in the interaction model refer to Appendix 7.

Table 5.4 Interaction terms for adjusted hazard ratios of time to readmission within 30-days following index hospitalization according to driving distance to a pediatric tertiary care centre and level of medical complexity [#]

Interaction terms	HR (95% CI)
≥80km driving distance*Single CCC with TA	0.39(0.11-1.42)
≥80km driving distance*Multiple CCC without TA	0.76(0.42-1.37)
≥80km driving distance*Multiple CCC with TA	0.75(0.32-1.79)
≥80km driving distance*NI CCC without TA	0.79(0.49-1.28)
≥80km driving distance*NI CCC with TA	0.95(0.38-2.35)

[#]Model also includes distance, level of medical complexity, age, gender, neighbourhood SES, Residence (urban/rural) and Primary Care Provider

Hazard ratio (HR), confidence interval (CI), kilometers (km), complex chronic condition (CCC), technology assistance (TA), and neurologic impairment (NI)

Chapter 6: Discussion

6.1 Discussion

The framework definition for children with medical complexity (CMC) characterizes this group of patients as having severe chronic conditions associated with medical fragility and functional limitations, as well as substantial care needs and health care use. [4] An increasing number of clinical-care programs have been put in place in Canada [30] and in many other developed countries around the world [84] to improve care delivery for CMC and decrease unwarranted utilization of services. Yet, CMC are still identified as the group with the highest rate of pediatric hospital readmission. [6] This study aimed at understanding factors, and in particular geographical barriers, driving readmissions in CMC. Based on work from Yantzi *el al.*, [33] we looked at the association between driving distance to specialized care and 30-day readmission. The current study is the first population-based study to examine characteristics of CMC and the pattern of their health service use in Quebec, Canada. The key findings were that close to a quarter of CMC lived 80 kilometres or more from a pediatric tertiary centre, and that this group of CMC had less outpatient visits to family physicians, pediatricians or specialists—especially when the number of visits reached a certain threshold—compared to those living within a driving distance of 80 kilometres. However, the remote CMC utilized more unplanned/unscheduled services such as emergency department visits and repeated hospital admissions compared to their closer counterparts. Yet, no association was found for the hazard of time to readmission within 30 days following an index hospital admission for CMC living at a driving distance of 80 kilometres or more compared to those living at a driving distance of less than 80 kilometres from the closest pediatric tertiary care centre.

CMC in Quebec, Canada

CMC make up a small proportion of children in Quebec. Our cohort consisted of 27,696 CMC, representing approximately 2.2% of the total population of children aged 2 to 16 years old in Quebec in this time period. [36] CMC were included in this study if they had at least 1 hospitalization or 2 health service claims with ICD-9 or ICD-10 codes consistent with CCC diagnoses in the period of January 1, 2010 to December 31, 2011. Our proportion of CMC is higher than the 0.67% reported by Cohen *et al.* [17] in Ontario, Canada which utilized the same CCC classification as ours; however, their cohort consisted solely of patients who had a hospital admission during a 2-year period. In contrast, Kuo *et al.* reported [16] 0.4% as the proportion of complex children with special health care needs (CSHCN) from all the children in the United States. This latter study was based on the National Survey of CSHCN (n=40,723) and more medically complex CSHCN were identified based on caregiver reports of child and family care needs, dependence on technology assistance and involvement of 2 or more specialists.

Within the cohort of CMC in our study, 62.5% were identified as single-CCC, 11.55% as multi-CCC, and 25.9% as neurologically impaired (NI). Close to 5% of CMC were dependent on technology assistance (TA). The 3 most important sub-categories of CCC body-systems in our cohort included patients with malignancy (33.7%), neurological impairment (25.9%) and congenital or genetic defect (25.5%) as shown in Figure 4.1. All proportions of the groups of children with CCC (single-CCC, multi-CCC and NI) were consistent with the Cohen *et al.* study [17] except for the percentage with TA, which was slightly lower in our study. Again, the disparity in TA could be attributed to the fact that the hospitalized CMC in Cohen's study were likely more fragile and medically complex than CMC selected from both community and hospital

settings as was the case in our study. [17] For example, in a study investigating the characteristics of hospitalizations for CMC enrolled in a structured clinical-care program, Berry *et al.* stated that 69% of patients had technology assistance. [44] This high percentage was likely explained by the fact that the latter cohort of CMC were hospitalized as well as followed by a dedicated complex care program. These two criteria most likely selected a sample of more complex patients with more intense care needs dependent on vital therapies such as technology assistance.

Baseline characteristics of CMC and the 80 kilometre cut-off

When our CMC cohort was divided according to the exposure, driving distance from the closest pediatric tertiary care hospital, we found that 76.3% of CMC resided within 80 kilometres, while 23.6% were outside that distance. These results were similar to another Cohen *et al.* study which reported that between 20.1-22.8% of single-CCC and multi-CCC were living at a distance of more than 80 kilometres from a specialized hospital. [66] The baseline characteristics of the CMC were similar by exposure categories, except for neighbourhood SES, and location of residence (urban/rural).

Neighbourhood SES and residence (urban/rural) and differences by exposure

The neighbourhood socio-economic status (SES) represented a family's employment, income and education based on small geographic units of 400 to 700 persons from census data, also known as disseminated areas. The distribution of those units was separated in quintiles representing 20% of the population. [85] It is important to understand that neighbourhood SES is not an individual-level measure but a small-area measure of socio-economic conditions. These quintiles can be appreciated when looking at the distribution of neighbourhood SES for the

overall population of CMC in Quebec in Table 5.1. CMC living closer to pediatric hospitals have an inverse neighbourhood SES distribution compared to those living farther. The largest proportion of children were among the higher neighbourhood SES quintiles for CMC living <80km, while most children were among the lower neighbourhood SES quintiles for CMC living ≥80km. These findings are similar to those reported by Pampalon in his study of health differences in for the general/total population of Quebec. [32]

The residence (urban/rural) variable represented the standard geographic distribution of Statistics Canada. [67] The "urban" group was categorized by a population density of at least 10,000 inhabitants, while the "MIZ" groups represented different levels of metropolitan influence zones (MIZ) that were calculated according to the number of workers that commuted to those urban areas. Finally the "rural" group represented areas with no MIZ. Therefore, the residence(urban/rural) variable did not correspond to a specific distance from the most populated areas but more as a measure of population density or the influence of those more densely-populated areas on their neighbouring towns. This is an important distinction to make when interpreting our results. Although CMC located at a driving distance ≥80 kilometres to the closest tertiary care centre might have to travel significant distances to reach specialized *pediatric* services, half of those CMC still resided in urban areas and another 20% lived in areas of strong MIZ, where other general health professionals and services may be accessible such as family physicians, pediatricians, as well as general hospitals serving mainly the adult population.

Fewer younger children and more older children in CMC

Both exposure levels had the smallest proportion of CMC within the 2-4 year old age group (approximately 20%) compared to the 3 remaining age groups (range 25-28%). This is in contrast to other studies, where the younger age group was usually the most prevalent. [8 16 17 44 77] This contrast may be explained by differences in study design. For example, in these latter studies, children under the age of 2 were also included, which was not the case in the current study. Compared to older children, those under 2 years of age likely represent a larger proportion of infants that have graduated, or are still receiving care, in a neonatal intensive care unit for conditions often associated with transient medical complexity such as prematurity. [86] Another explanation could be that the age distribution of our CMC cohort follows the age distribution of Quebec children. In Quebec, when the total population of children was categorized by the same age groups as our study, the 2-4 year olds also represented the smallest group. [36]

Primary care provider in the care of CMC: Glass half full or half empty?

The primary care provider variable was based on an algorithm used for primary care research by ICES in Ontario, Canada, [78] adapted by adapted *Nakhla and Li,.*[79] which identified subjects if they were regularly followed by a family doctor or pediatrician for primary care services according to specific physician billing codes. In our study, proportions of children with a primary care provider were similar according to driving distance exposure (66.3 % and 64%, for CMC at a driving distance of less than 80 kilometres versus more than 80 kilometres respectively). These findings may be difficult to compare to what has been reported in the literature, since previous studies usually differentiate areas by urban versus rural as opposed to

driving distance to the closest pediatric tertiary care centres. For instance, Pampalon *et al*. reported that in the general population of Quebec, urban citizens are less likely to have a family physician compared to residents living in any of the MIZ. [32] Yet, a study looking specifically at children in Ontario, Canada, reported that 32.8% of children living in areas with low physician supplies, which are often more distant locations, had no primary care provider (family physician or pediatrician) compared to 6.3% in the high physician supplies areas. [87]

Nonetheless, primary care providers in Quebec seem to be involved with the majority of CMC regardless of their location of residence. These findings are similar to those reported by Kuo *et al.* in which they found that despite enrolment in a structured clinical program for CMC, 55.6% still utilized a primary care provider as their main point of contact. [50] Are these findings reassuring or concerning? On the one hand, the majority of CMC have a regular primary care provider; on the other hand, one third of CMC do not. Structured clinical complex care programs in Quebec may provide to a certain extent primary care services in a "one-stop-shop" model of care. However, due to lack of resources, CMC enrolled in such programs only represent a small proportion of the overall CMC population of Quebec. For example, in Quebec, the Montreal Children's Hospital complex care service has one of the largest cohorts of CMC in the province, but only follows a maximum of 400 patients (personal communication, Dr H. Patel).

Another interesting dimension to our findings is that, overall, 66% of CMC had a regular primary care provider; while 60% and 87% of CMC were seen at least once by a family physician or a pediatrician, respectively. The differences between these visits and the ones that

identify primary care providers are related to physician billing codes. Physician billing codes used to identify family physician/pediatrician visits included codes related to primary care services, but could also refer to care provided via consultation in the office, follow-up or consultation within a hospital setting, multidisciplinary meeting/parent meeting for a complex pathology (office or hospital) and care provided at home. Are primary care providers, but more specifically pediatricians, only involved transiently with CMC to provide primary care or specialty consultation? Could CMC be mostly assessed by primary care providers within hospital settings? These questions cannot be answered with our current data. However, these are questions that could potentially be explored in future research.

In brief, the constitution of our study population seemed to be consistent with previously published data. Any differences may be explained by the fact that our study is unique by including in our study sample both inpatient and outpatient CMC, as well as examining CMC from a population-based point of view with a health administrative database. Lastly, no major demographic differences were identified between CMC in the 2 exposure groups, except for lower quintiles of neighbourhood SES and less metropolitan influence in the group corresponding to a distance of more or equal to 80 kilometres from a pediatric tertiary care centre.

30-day readmission and the association with driving distance

Overall, the rate of 30-day readmission over the 2 years of our study was 7.6%. The rate of readmission reported in the literature for CMC is quite variable and ranges from 6.3 to 25.4%. [17] [21] [42] [44] In our study there were 379(7.5%) readmissions in the group living at a driving distance

of less than 80 kilometres and 135(8.1%) readmissions in the group at more than 80 kilometres. These proportions were not significantly different. In both categories of driving distance exposure, multi-CCC with TA had the most 30-day readmissions. In fact, the presence of TA increased the proportion of readmissions in each CCC category. When Cox proportional hazard regression was performed for readmission based on driving distance, results were similar. The adjusted hazard ratio for 30-day readmission was 1.07(95% CI 0.86-1.34) for CMC living beyond 80 kilometres compared to those closer to pediatric centres. All CCC categories were associated with a significant increased hazard ratio for 30-day readmission compared to the reference (single CCC without TA), but the highest adjusted hazard ratio was for multi-CCC with TA at 3.12(95% CI 2.14-4.55). Finally, the 4th quintile for neighbourhood SES—the second lowest—was associated with a significant hazard ratio of 1.52(95% CI 1.13-2.05).

Thus, driving distance was not associated with 30-day readmission in CMC. These results can be compared to by Peltz *et al*. who studied 30-day readmissions in the general pediatric population from 41 children's hospitals in the United States. Over 1 year and 672,190 pediatric admissions, they found a slight increase in odds of 30-day readmission for rural children compared to urban ones with an odds ratio of 1.1(95% CI 1-1.1). Overall, rural children lived at a median distance of 110 kilometres from the children's hospitals (IQR between 77 and 167 kilometres) and also resided in lower income areas. [88] As this latter study can attest, distance has the potential to affect 30-day readmissions, but in what context? Could there be additional factors that come in play when studying 30-day readmission in the CMC population? Lorch *et al.* demonstrated an association between pediatric 21-day readmission and a travel time greater than 15 minutes compared to a shorter travel time, but only for conditions of lower severity such as

asthma, gastroenteritis and urinary tract infection with odds ratios of 1.46(95% CI 1.04-2.04), 2.64(95% CI 1.43-4.90), 4.5(95% CI 2.0-10.2), respectively. That same association was not seen for more severe conditions such as meningitis (odds ratio 1.24, 95% CI 0.49-3.12). Additionally, sicker children with other severe diagnoses like seizures or co-morbid conditions were more likely to undergo longer travels in order to reach tertiary pediatric hospitals during a clinical deterioration. [89] Although the Lorch *et al*. study does not address individual CMC directly, could families of sicker children with medical complexity be willing to travel longer distances to reach specialized care and attend timely follow-up assessments irrespective of where they reside, especially when their child has been recently sick and hospitalized? Could this tendency act as a protective factor against risk of hospital readmissions? As previously stated by Yantzi *et al.* when referring to children with chronic diseases: "Parents will often do whatever it takes to help their child, which in this case involves taking the child to the hospital that will provide the most comprehensive level of care". [33]

Lower SES may be a more important barrier to timely health care access rather than distance in the context of a recent hospital discharge for CMC. In our study, we found that the second lowest SES quintile (Q4) was associated with a significant increased hazard ratio of 30-day readmission. This association was not observed for any other SES quintile, including the lowest. Although this result may be attributed to chance, those who work closely with families of children with chronic diseases have reported that families representing that fourth SES quintile often receive less government support compared to the lowest income families (personal communication, Dr H. Patel). Indeed, the RAMQ has in place a *Public Drug Insurance Plan* for individuals and families identified by the *Ministère de l'emploi et de la solidarité sociale* for

requiring social assistance. [90] Families with a certain level of income, even if very modest, may not be eligible for such programs which means they might have more out-of-pocket expenses for medications and equipment required by their ill child. As reported by parents of CMC: "Sometimes I feel like giving up and going on welfare [...] No matter how much money we made, we could not subsidise these kinds of bills". [19] Therefore, these families experience additional financial strains which may impact the care of CMC in different ways, including limited time and transportation to specialized facilities or even affording medications/therapies that could potentially prevent further admissions. These observations require validation in future research.

Another possible explanation for the lack of association between travel distance and readmission is that optimal chronic care management, care coordination, continuity of care between tertiary care centres and community providers, as well as primary and specialty care access, [23 42] which have all been linked with a decrease in preventable admissions for CMC, are suboptimal throughout the province no matter how close or far CMC live from pediatric tertiary care centres. As shown in our results and many other studies, [42 73] medical complexity is an important risk factor for readmission in itself and without a robust discharge plan in place, 30-day readmissions may be to some extent inevitable.

In sum, our 30-day readmission outcome may not have been the best measure to capture the struggles related to geographical barriers, as worried parents, even those living more remotely, in times of illness significant enough to have required treatment in hospital, may be particularly motivated to drive greater distances in order to receive expert follow-up care, which

consequently may have a protective effect on risks of readmissions. Concurrently, optimal discharge planning for CMC, which includes care coordination, communication between providers and outpatient specialty and primary care follow-up, may be lacking across the board, affecting rates of 30-day readmission no matter where CMC reside. Our data also points to the importance of social determinants of health when considering risks of 30-day readmission, as disadvantaged populations may be at disproportionally greater risks of readmissions compared to the general population. [91]

Other patterns of health care utilization for CMC

Considering driving distance from pediatric tertiary care centres does not seem to be associated with 30-day readmission, how does distance influence other aspects of health care utilization? There have been previous reports centred on patterns of health care use for CMC at the population-level [17]and between different types of hospitals (tertiary care, community, regional),[77] as well as studies on unmet care needs [60 61] and access to a medical home [51 59] based on geographical variation. However, to our knowledge we are the first study addressing health service use of CMC other than readmission, based on distance from pediatric tertiary care.

CMC health services and the 80 kilometres cut-off

As shown in Table 5.2, we can see significant unadjusted differences based on driving distance for many secondary outcomes including: number of hospitalizations, number of specialist visits, number of distinct physicians and number of emergency department visits. At first glance, these differences might not seem important but on careful examination the distribution of service use varies, especially when examining the amount of services above a certain threshold. As shown

in Figure 5.2, the majority of children, no matter their location of residence, had between 1 and 2 hospitalizations. However, when we looked at children that had 3 or more hospitalizations, there was a slightly higher percentage of CMC living at a distance beyond 80 kilometres from pediatric hospitals compared to those living closer (21% versus 18%, respectively). This difference may be clinical relevant as, in general, the overwhelming majority of children that experience a hospitalization only have one per year. [92] It is interesting to note that there was no difference in cumulative length of stay based on distance from the closest specialized pediatric care centres which contradicts many previous studies; although our results may not be directly comparable since most studies solely study the length of stay relative to the index admission. [88 89] On the other hand, as shown in Figure 5.7, a lower proportion of CMC at a driving distance of ≥80km had more than 6 specialty visits compared those living at <80km (26.3% versus 31.2 % respectively). This tendency towards fewer specialty visits at greater distances may well be related to lower specialty physician supply in those areas. On the other hand, six visits was the median number of distinct medical specialties regularly following CMC in a study from Cohen *et al.* where distance was not taken into account. [17] With so many specialists involved, especially for children with more complex needs, we can easily imagine how the number of visits can add-up quickly over a two year period. Again, in our study, the majority of children saw between 1 and 4 distinct physicians, but for children living at a distance beyond 80 kilometres versus those closer, a smaller percentage had been assessed by 5 or more distinct physicians (less than 33.2% versus 37% respectively). Finally, as shown in Figure 5.8, a higher proportion of CMC living at a distance beyond 80 kilometres had more than 3 visits to the emergency department compared to those closer (33% versus 30% respectively).

Health service research for CMC cannot be complete without examining the visits with family physicians and pediatricians according to driving distance. A smaller percentage of CMC living above 80 kilometres from a pediatric hospital were seen at least once by a family physician or pediatrician (Table 5.2). Differences were also seen in the frequency of those visits according to the distance from a pediatric tertiary care hospital. As seen in Figure 5.4 and Figure 5.5, a larger percentage of CMC living farther had between 1-2 visits to the family physician and 1-6 visits to the pediatrician compare to those closer; however, a smaller percentage of CMC living farther had a number of visits above that threshold.

When our health service utilization results are compared to Cohen *et al.*'s cohort of hospitalized CMC in Ontario, Canada, the extent of utilization seen in our study is slightly lower. For example, overall CMC had a median of 13 distinct physicians (IQR 8-20), a median of 2 emergency department visits (IQR 1-5) and a median of 12 primary care visits (IQR 6-20).[17] These differences could be explained by minor differences in the structure and performance of 2 distinct provincial health care systems but also because Cohen *et al.*'s cohort was likely more medically complex due to the fact that they were recruited in hospital.

Overall, it seems there are many differences in the patterns of health care utilization of CMC based on driving distance from the closest pediatric tertiary care hospital on unadjusted analyses. These differences are even more accentuated above a certain threshold. For example, the majority of CMC, no matter where they reside, utilized the same median number of baseline services over 2 years (1-2 family physician visits, 1-6 pediatrician visits, 1-6 special specialist visit, however, beyond those baseline services, CMC living farther have fewer scheduled visits

(family physician, pediatrician, specialists) and more presumably unplanned services (emergency department visits or hospital admission) and vice-versa for CMC living closer. Could a lower frequency of outpatient visits for regular follow-up explain the higher occurrence of emergency visits and hospital admissions? It may be possible that repeated hospital admissions (or in other words, repeated hospital readmissions beyond 30 days) and frequency of emergency department visits, may be more sensitive measures to capture the disturbances in daily routine associated with longer travels, as families of CMC could be less willing to travel longer distances in periods of relative medical stability compared to when their child had been recently sick and hospitalized within 30 days. This possible lack of routine follow-up for issue-based care but also preventive care may leave CMC more susceptible to acute deteriorations requiring management as an inpatient or in an emergency department. When families of CMC report that "even a trip to the grocery store has to be extremely choreographed", [19] one cannot deny that such travels—but more specifically the frequency of those travels—may represent an additional burden for families living farther from specialized tertiary centres. [33]

Nonetheless, other factors may come into play and influence patterns of health utilization for CMC, as it is unclear why CMC living farther from tertiary centres also had less frequent visits to the family physician and pediatrician compared to those closer. As stated previously, half of the CMC living at a distance of ≥ 80km from a pediatric tertiary care centre were located in areas considered urban, where in theory physician supplies should be adequate and visits to local providers, such as family physicians or pediatricians, should not necessitate significant travel distances due to geographic proximity. Could these differences be related to individual CMC characteristics such a functional limitation which was not captured by our CCC classification

system, geographic disparities in the supports and resources from local health authorities, or even the efficiency of public transport (more available in larger urban centres)? These questions cannot be answered within the data available in our study but could possibly be explored in future research.

Health service use in CMC: a typical trend

This study gives us a first glimpse of who the children with medical complexity are in Quebec and what their aggregated health care utilization experience is. Although travel distance was not associated with 30-day readmission, we saw many other differences according to travel distance such as the distribution of the total number of hospital admissions, primary care visits and specialty visits, as well as emergency department visits. What is also striking is that, no matter where CMC reside, a certain percentage of these children are subject to an excessive number of hospital admissions. In our study, approximately 1% of the overall proportion of admitted CMC had 12 or more hospitalizations (Figure 5.2). The latter is a typical trend seen with CMC: as a small proportion of children are responsible for a very large share of the costs and services. [17] This trend was also found in a study from Berry *et al*. examining the 579,504 admissions that occurred within 37 children hospitals situated in the United States, as 2.9% of children were responsible for 18.8% of the total number of admissions. Are these CMC so acutely sick that readmissions to hospital are inevitable? Could these admissions have been prevented with better homecare support and outpatient follow-up? Future research focusing on the perspectives of families, as well as health care providers in combination with information from administrative health care databases should be a priority in order to gain more information on preventable readmissions in CMC.

In summary, reducing potentially preventable admissions for CMC has been a challenge but with every new study dedicated to this vulnerable population we continue to learn and adapt our clinical approach. The most relevant findings from this study are that differences in the frequency of services are present according to driving distance from a tertiary care hospital when it comes to the frequency of health services such as visits to family physicians, pediatricians, specialized, emergency department, as well as hospital admissions, which are more noticeable above a certain threshold of health services such as 3 or more hospital admissions or 6 or more specialty visits. Although, it is clear these differences cannot be fully explained solely by geographical barriers such as driving distance. The next step would be to understand the barriers for community primary care providers such as family physicians and pediatricians in caring for CMC, and how these relate to readmissions and health care utilization. We could then consider developing initiatives to support primary care provider in the care of CMC such as education that starts at the residency level, CMC-specific training opportunities, allied health resources that includes patient care coordinators but most importantly, fostering partnerships between pediatric tertiary care hospitals and community health centres in order to facilitate communication, continuity of care, as well as translation of knowledge, services and clinical frameworks for CMC.

6.2 Strengths and limitations

This study had strengths and brought new information to the surface in regards to CMC in Quebec, Canada. Firstly, this study was the first population-based study in Canada examining health service utilization of CMC in a large cohort defined by CCC criteria derived from children in Quebec. Moreover, the RAMQ data linked health care used various settings so health services could be captured across inpatient and outpatient services. The observations gathered with our

database also permitted us to explore, link and adjust for important variables such as a usual primary care provider based on an algorithm that highlights physician billing codes for primary care services, neighbourhood SES which was related to income, employment and education, as well as location of residence (rural/urban) that incorporated a well utilized system, the Statistics Canada's Statistical Area Classification (CMA, CA, MIZ). Finally, this study utilized the distance parameter from the study from Yantzi *et al.* [33] in which a distance of "80 kilometres or more" to a hospital was associated with family stress and disruption of the family unit and portrayed health service use based on this critical distance of 80 kilometres which added an other dimension to that concept .

Several limitations of our study design need to be discussed, as these may have influenced our study results. Our population of CMC under study, as well as all observations (including the driving distance exposure and outcomes related to 30-day readmission and other determinants of health care utilization) were extracted from a retrospective health administrative database. Administrative databases are compiled for administrative purposes and not necessarily research, therefore the information gathered may be subject to misclassification, omissions, and poor coding quality. Moreover, CMC may not have been captured in our study sample if they did not have either 1 hospitalization or 2 physicians' claims with a CCC-related diagnosis. However, even if a child had a less complex or morbid CCC, it is unlikely that this child would not have had 2 physician visits over 2 years. The complex chronic condition (CCC) framework used to define children with medical complexity in this study is widely utilized in health care service research but currently not rigorously validated due to the complex nature of the algorithm [40]. CCC does not include all chronic conditions and may not be able to differentiate medical

complexity arising from multiple diagnoses of lower severity. Likewise, CCC cannot capture the full extent of functional limitation and psycho-social complexity associated with the CMC definition framework and may include diagnoses of transient complexity such as those related to malignancy. On that same theme, the algorithm to detect enrollment under a primary care provider from *Nakhla and Li* [79] was created in collaboration with clinicians and knowledge users, and was been adapted from algorithms used for primary care research by ICES in Ontario, Canada. [78] Billing codes for the above algorithm, as well as family physicians, pediatricians and specialty service claims were selected based on a review of the *Manuel de facturation des omnipracticiens* [93] and *Manuel de facturation des spécialistes* [94] from the RAMQ; they have not been previously validated. At this time they exhibit face validity only.

This study was limited by the information that was available. For example, other variables that may have influenced the variability in the rate of 30-day readmission were not integrated in our analysis due to the unavailability of the RAMQ database. However, these variables could be considered in future research and include: 1) The risk of 30-day readmission for repeated index admissions over time. Some studies have suggested that the number of hospital admissions prior to the index hospitalization increases odds of 30-day readmission; therefore, following subjects longitudinally for repeated index admissions could have portrayed a different picture. [43 73] In this current study subjects were censored following their first index admission; 2) Length of the hospital stay of the index admission. Length of hospital stay, which is most often reported in days, has been shown to increase risk of 30-day readmission and is often included in models predicting risk of 30-day readmission in the pediatric population; [43 88 89] 3) Parental perception of their child's health at hospital discharge. Parental perception has been shown by Berry *et al.* to

decrease the odds of 30-day readmission (adjusted odds ratio of 0.2 with 95% CI of 0.1-0.6) when parents strongly agreed with the statement "I felt that my child was healthy enough to leave the hospital" just before going home; [95] 4) Health service utilization specifically in the 30 days following hospital discharge. Post-discharge care by primary care providers, specialists and homecare services may contribute to reducing hospital readmissions; 5) Location of index hospitalization. The study population consisted of patients with CCC diagnoses, and by definition these children have conditions severe enough to require specialized pediatric care in a tertiary care centre. Therefore, we assumed that most of the hospital admissions would occur within one of the 4 pediatric hospitals situated in Quebec. However, there is always a possibility that children could have been hospitalized closer to home in a community or regional hospital. Location of hospitalization is very important as many studies have noted a great variability of 30-day readmission according to the type of hospital. [43 68] For example, Feudtner *et al.*, contrary to popular belief, found that better performing medical institutions, such as tertiary care centres, had higher rates of readmissions. The exact reason is unclear but possible explanations include: greater severity of admitting diagnoses, higher complexity of the baseline patient population, increased access to specialized care and interventions, suboptimal communication between specialized centres and the community; [43] 6) Planned hospital readmission. Compared to other administrative databases available in other Canadian provinces, planned and unplanned readmissions could not easily be differentiated in the Med-Echo database. Therefore, some of the readmissions accounted for could have been planned to administer vital therapies such as chemotherapy for a child with a malignancy. Future research could consider the exploration of admitting diagnoses and how it relates to readmissions.

Furthermore, our study framework was based on work from Yantzi *et al*. suggesting that 80 kilometres from hospital is a critical distance associated with significant familial stress and disruption of the daily routine. [33] However, it is possible that patterns of health care use varied according to different distances from pediatric tertiary care centres. In future studies, various driving distance categories will be considered using a more geographically refined measure. Likewise, other geographical barriers such as travel time and difficulties related to transportation that were not accounted for in this study may be included.

Finally, when examining our secondary outcomes (other inpatient and outpatient services including number of hospitalizations, pediatrician visits, specialty visits), statistical significance was observed for medians and percentages that appeared similar between the two driving distance exposure groups. This could be attributed to our large study population; however, even these small differences represented hundreds of patients ((ie. 1% of 27,696 (our study sample) represented 277 children)). These findings will be confirmed in future research using adjusted statistical models.

6.3 Conclusion

This provincial population-based evaluation of CMC pattern of health care utilization based on travel distance from pediatric tertiary care centres using a comprehensive database, although limited by a few issues intrinsic to health administrative data and CMC research, has highlighted many interesting findings that help us understand health service use in this complex, vulnerable population. For instance, although 30-day readmission was not associated with travel distance, we observed differences in use of health care services, especially when utilization of specialized

services reached a certain threshold. Along with these differences, we found that 66% of CMC had a usual provider of primary care which still left a third of CMC without a family physician or pediatrician serving as a main point of contact with the health care system and specialized care.

Ongoing research focused on CMC is required in Quebec but also on a national platform to address the needs of this complex population and barriers faced by families and health care providers. For instance, in this study we made reference to many factors that could possibly influence hospital readmissions and other patterns of health care utilization such as medical complexity, functional limitation, illness severity, family stressors, community/government support, financial strains, transportation difficulties and other social determinants. Using data from an administrative database permitted us to scratch the surface of the underlying mechanisms influencing patterns of health care utilization in CMC. The next step would be to obtain the point of view of families and primary care providers caring for CMC in the community to add to the findings of this current study to understand the full picture, in order to develop programs and infrastructure that not only reduce unnecessary utilization of health care services such as readmissions but also improve quality of care for CMC.

Chapter 7: References

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APPENDIX

Appendix 1 Ethics approval letter

Centre universitaire de santé McGill
McGill University Health Centre

Comité d'éthique de la recherche CUSM
MUHC Research Ethics Board
2155 rue Guy, suite 790.2
Montreal, QC, H3H 2R9, CANADA
Tel: 514 934- 1934
Email: [illegible]

April 14, 2016

Dr. P. Li
Pediatric
MUHC - Montreal Children's Hospital
1001 boul. Decarie, #
Montreal, Qc H4A 3J1

Re: # 12-297-PED A Population-based Study on the Association of Primary Care Reforms with Health Services Utilization and Quality of Care Outcomes among Vulnerable Children in Quebec, Canada

Dear Dr. Li,

We are writing in response to your correspondence requesting Research Ethics Board review of an amendment for the research study referenced above.

We are pleased to inform you that approval for the following documents was provided on April 14, 2016 via delegated review and will be reported at the REB meeting on April 25, 2016:

- MUHC eReview Revision to an Approved Study form (protocol amendment to include 2 sub-studies that will analyze RAMQ data)
- Protocol - addendums :
  - Sub-study - Analysis on Status of Primary Care Provider and Health Care Services Use in Children with Recurrent Acute Otitis Media, version 1, April 12, 2016
  - sub-study – Travel Distance to Tertiary Care Centres in Patterns of Health care Utilization Among Children with Medical Complexity in Quebec: A population-based cohort study, (no version/date)

Should any revision to the study, or other unanticipated development occur prior to the next required review, you must advise the REB without delay. Regulation does not permit initiation of a proposed study modification prior to REB approval for the amendment.

Sincerely,

JA/ McDonald, MD, FRCPC
Co-chairperson,
MUHC Research Ethics Board – pediatric panel

Pediatric Panel
MUHC - Montreal Children's Hospital (Ste-Catherine Pavilion)
[illegible]

Appendix 2 Information requested from RAMQ database

Table 1 : Beneficiary Files	
No séq.	*Les renseignements autorisés*/authorised information
1	01- *Numéro banalisé de l'individu*/individual identifier number
2	02- *Année et mois de naissance de la personne assurée*/Year and month of birth of registered person
3	03- *Sexe de la personne assurée*/Sex of registered person
4	04- *Région sociosanitaire de la personne assurée*/health region of the registered person
5	05- *Territoire centre local de services communautaire la personne assurée*/Region of local community service centre of the registered person
6	07- *Date index* /Index data (AAAA-MM-JJ)
7	08- *Année et mois du décès de la personne assurée*/Year and month of death of the registered person
8	08 – *Code Zone d'Influence métropolitaine*/Metropolitan influenced zone code

Table 2 : Material and Social Deprivation Index	
No séq.	*Les renseignements autorisés*/authorised information
1	1- *Numéro banalisé de l'individu*/individual identifier number
2	2- *Année*/year
3	3- *Quintile des composantes matérielle (Quintmat)/*Quintile of material deprivation index
4	4- *Quintile des composantes sociale (Quinsoc*)/Quintile of social deprivation index
5	5- *Centile des composantes matérielle (Centmat*)/Centile of material deprivation index
6	6- *Centile des composantes sociale (Centsoc*)/Centile of social deprivation index
7	7- Base
8	8- *Groupe*/Group

Appendix 2 (…continued)

Table 3 : Fee-for-service Physician Claims	
No séq.	*Les renseignements autorisés*/authorised information
1	01- *Numéro banalisé de l'individu*/individual identifier number
2	02- *Classe du professionnel*/professional class
3	03- *Numéro banalisé du professionnel*/professional identifier number
4	04- *Spécialité du professionnel*/professional speciality
5	05- *Code d'entente de facturation de la demande de paiement*/Billing agreement code for payment claim
6	06- *Code de groupe d'actes*/Service group code
7	07- *Code d'acte*/Service code
8	08- *Rôle dans l'exécution de l'acte*/Service execution role
9	09- *Date du service* /Date of service (AAAA-MM-JJ)
10	10- *Code de diagnostic*/Diagnosis code
11	11- *Type d'établissement*/establishment type
12	12- *Numéro banalisé de l'établissement*/establishment identifier number
13	13- *Code de localité du lieu de dispensation banalisé*/Location code of service delivered
14	14- *Région sociosanitaire du lieu de dispensation*/health region of service delivered
15	18- *Montant facturé*/billed amount
16	21- *Classe du professionnel référent*/Class of referral professional
17	22- *Numéro banalisé du professionnel référent*/Identifier number of referral professional
18	23- *Spécialité du professionnel référent*/Specialty of referral professional

Appendix 2 (…continued)

Table 4.1 : Med Écho-Diagnostics	
No séq.	*Les renseignements autorisés*/authorised information
1	1- *Numéro séquentiel banalisé du séjour hospitalier*/serial number of hospital stay
2	2- *Numéro banalisé de l'individu*/individual identifier number
3	3- *Type de diagnostic*/type of diagnosis
4	4- *Numéro séquentiel du diagnostic*/serial number of diagnosis
5	5- *Numéro séquentiel du système de classification*/serial number of diagnosis classification system
6	6- *Code de diagnostic médical clinique*/clinical medical diagnosis code
7	7- *Code de caractéristique du diagnostic/*diagnosis characteristic code

Table 4.2 : Med Écho-Interventions	
No séq.	*Les renseignements autorisés*/authorised information
1	01- *Numéro séquentiel banalisé du séjour hospitalier*/serial number of hospital stay
2	02- *Numéro banalisé de l'individu*/individual identifier number
3	03- *Numéro de l'intervention*/intervention number
4	04- *Date de l'intervention*/intervention data (AAAA-MM-JJ)
5	06- *Numéro séquentiel du système de classification*/serial number of diagnosis classification system
6	07- *Code d'intervention santé*/health intervention code
7	08- *Code de l'attribut de situation d'intervention*/attribute code of intervention situation
8	09- *Code de l'attribut du lieu d'intervention*/attribute code of intervention location
9	10- *Code de l'attribut d'étendue de l'intervention*/attribute code of the extent of intervention

Appendix 2 (…continued)

Table 4.3 : Med Écho-Administrative hospital records	
No séq.	*Les renseignements autorisés*/authorized information
1	01- *Numéro séquentiel banalisé du séjour hospitalier*/serial number of hospital stay
2	02- *Numéro banalisé de l'individu*/individual identifier number
3	03- *Numéro banalisé de l'établissement* /serial number of establishment
4	04- *Région sociosanitaire de l'établissement*/health region of establishment
5	05- *Date d'admission*/admission date (AAAA-MM-JJ)
6	06- *Date de départ*/discharge date (AAAA-MM-JJ)
7	07- *Type de soins*/type of care
8	16- *Numéro banalisé de l'établissement de provenance du ministère de la santé et services sociaux*/serial number of establishment from ministry of health and social services
9	17- *Type de lieu de provenance*/type of previous location
10	18- *Date d'arrivée à l'urgence*/arrival date to emergency (AAAA-MM-JJ)
11	19- *Nombre de jours d'absence*/days of absence
12	20- *Nombre de jours séjour hospitalier*/days in hospital
13	21- *Numéro banalisé de l'établissement ministère de la santé et services sociaux*/serial number of establishment from ministry of health and social services
14	22- *Type de lieu de destination*/type of discharge destination
15	24- *Type de décès*/type of death

Table 4.4 : Med Écho-Services	
No séq.	Les renseignements autorisés/authorised information
1	1- *Numéro séquentiel banalisé du séjour hospitalier*/serial number of hospital stay
2	2- *Numéro banalisé de l'individu*/individual identifier number
3	3- *Numéro de séjour du service hospitalier/*serial number of hospital service during hospital stay
4	4- *Code de service*/service code
5	6- *Classe du dispensateur lors du service*/class of service provider
6	7- *Code de spécialité du dispensateur lors du service*/service provider specialty code
7	8- Nombre de jours dans le service/number of days on service

Appendix 2 (…continued)

Table 4.5 : Med Écho-Intensive care	
No séq.	*Les renseignements autorisés*/authorised information
1	1- *Numéro séquentiel banalisé du séjour hospitalier*/serial number of hospital stay
2	2- *Numéro banalisé de l'individu*/individual identifier number
3	3- *Numéro de séjour aux soins intensifs*/number of intensive care stay
4	4- *Code de l'unité de soins intensifs*/intensive care unit code
5	5- *Nombre de jours aux soins intensifs*/days in the intensive care

Appendix 3 ICD-9-Quebec and ICD-10-Standard for CMC (adapted from Cohen et al)[17]

CCC category	ICD-10	ICD-9-Quebec
NI	G11x, G12x, G20x, G23x, G310, G318, G319, G32x, G40x, G41xG241, G242, G248, G250, G251, G252, G253, G254, G255, G25x, G71x, G72x, G80x, G81x, G82x, G83x, G901, G903, G904, G94x, G95x, G99x, G91x, G9388, G939, G10x, F700, F701, F708, F709, F71x, F72x, F73x, F78x, F79x, F842, Q00x, Q01x, Q02x, Q03x, Q04x, Q05x, Q06x, Q068, Q07x	318x, 319x, 343x, 344x, 345x, 3488, 3489, 3590 à 3593, 740x, 741x, 742x
Cardio	I420, I421, I422, I423, I424, I425, I427, I428, I429, I44x, I45x, I47x, I48x, I49x, I515, Q20x, Q21x, Q22x, Q23x, Q24x, Q25x, Q26x	4250 à 4254, 4259, 4291, 426x, 4270 à 4274, 4276, 4278, 4279 745x, 746x, 7470 à 7474
Resp	E84x, P27x, Q30x, Q31x, Q32x, Q33x, Q34x	2770, 748x, 770x
Renal	Q60x, Q61x, Q62x, Q63x, Q64x, N18x	585x, 753x
GI	Q431, Q437, Q39x, Q41x, Q42x, Q44x, Q45x, K50x, K51x, , K73x, K74x, K754, K758 , K760	5714 à 5719, 5739, 555x, 556x 7503, 751x
Heme and Immuno	B20x, B21x, B22x, B23x, B24x, D570, D571, D572, D578, D55x, D561, D562, D564, D568, D569, D58x, D80x, D81x, D82x, D83x, D84x, D898, D899	042x, 043x, 044x 279x, 2820 à 2824, 2826, 2881 à 2882
Metabolic	E70x, E710, E711, E712, E713, E72x, E730, E74x, E75x, E76x, E77x, E78x, E79x, E803, E804, E806, E805, E807, E881, E882, E83x, E85x, E888, E889	2750 à 2753 270x, 271x, 272x, 2773, 2775 2772 à 2776, 2778, 2779

Appendix 3 (…continued)

Congenital or genetic defect	E343, K44x, Q75x, Q76x, Q77x, Q78x, Q790, Q791, Q792, Q793, Q794, Q795, Q87x, Q897, Q898, Q899, Q90x, Q91x, Q92x, Q93x, Q952, Q953, Q958, Q96x, Q97x, Q98x, Q992, Q998, Q999, M41x	2594, 5513, 5523, 5533, 7373, 7560 à 7565, 7834, 7566, 7567, 758x, 7597 à 7599
Malignancy	C00, C01, C02, C03, C04, C05, C06, C07, C08, C09, C10, C11, C12, C13, C14, C15, C16, C17, C18, C19, C20, C21, C22, C23, C24, C25, C26, C27, C28, C29, C30, C31, C32, C33, C34, C35, C36, C38, C39, C40, C41, C43, C44, C45, C46, C47, C48, C49, C50, C51,C52, C53, C54, C55, C56, C57, C58, C60, C61, C62, C63, C64, C65, C66, C67, C68, C69, C70, C71, C72, C73, C74, C75, C76,C77, C78, C79, C80, C81, C82, C83, C84, C85, C86, C87, C88, C89, C90, C91, C92, C93, C94, C95, C96, C97, D00, D01, D02, D03, D04, D05, D06, D07, D08, D09, D10, D11, D12, D13, D14, D15, D16, D17, D18, D19, D20, D21, D22, D23, D24, D25, D26, D27, D28, D29, D30, D31, D32, D33, D34, D35, D36, D37, D38, D39, D40, D41, D42, D43, D44, D45, D46, D47, D48	140x à 239x

Appendix 4 ICD-9-Quebec, ICD-10-Standard, CCA-Quebecand CCI for technology assistance (adapted from Cohen et al)[17]

ICD-10-Standard	**ICD-9-Quebec**
K9140, K9141, K9142, , K9143, K9144, K9145, K9146, K9149, J9500, J9501, J9502, J9503, J9508, J9509, Z430, Z431, Z432, Z433, Z435, Z436, Z451, Z452, Z458, Z459, Z465, Z469, Z490, Z491, Z492, Z930, Z931, Z932, Z933, Z935, Z936, Z950, Z960, Z961, Z962, Z963, Z964, Z965, Z9660, Z9661, Z9668, Z9669, Z967, Z968, Z969, Z992, T823, T824	5696, 5190, 5519, 5529, 5839, 1036, 5652, 1151, 1152, 4319, 4329, 1123, 1139, 4339, 1531, 1532, 1533, 1534, 1539, 1541, 1542, 1543, 1694, 1698, 1699,1661, 1669, 1690, 1693, 1697, 5811, 5823, 5127, 1052, 5195, 6698, 6693, 1053, 7199, 5142, 1161, 4971, 49743, 4983, 4986, 4987, 9805, 9961 V539, V550, V551, V552, V553, V555, V556, V560, V568, V451, V535, V450, V435, V431, V438, V436, V440, V441, V442, V443, V445, V446, V450, V451, V530, V535, V550, V551, V552, V553, V555, V556, V560, V568
CCI	**CCA-Quebec**
1NF53HATS, 1NF53LAQB, 1NF53LATS, 1NF53BTQB, 1NF53BTTS, 1NF53DAQB, 1NF53DATS, 1NK53BTTS, 1NK53DATS, 1NK53HATS, 1NK53LAQB, 1NK53LATS, 1NK53TGTS, 1NK77EM, 1NK77RQ, 1OW12ZZ, 1OW35CAD1, 1OW35CAD2, 1OW35CAD3, 1OW35HAD1, 1OW35HAD2, 1OW35HAD3, 1NF54HAFA, 1NF54HAQB, 1NF54HATS, 1NF80DA, 1NF55HATS, 1NF55JATS 1NK54HAQB, 1NK54HATS 1NK55BATS, 1NK55CATS 1NK55DATS, 1GJ77LALG, 1GJ77LA, 1GJ77QB, 1GJ77HA, 1GJ54CANR, 1GJ54JATS, 1GJ54JANG, 1GJ77HA, 1GJ77LA, 1GJ77LALG, 1GJ77QB, 1GJ55BAEB, 1GJ55BANR 1GJ55CAEB, 1GJ55CANG, 1GJ55CANR, 1GJ55CATS, 1GJ55JAEB, 1GJ55LAEB, 1GJ55LANR, 1GJ55LAPM, 1AP52MJSJ, 1AC52MFSJ, 1AP52MFSJ, 1AB52GISJ, 1AB52MFSJ, 1AC52GISJ, 1AC52MQSJ, 1AP52MQSJ, 1AB52GJSJ, 1AB52MQSJ, 1AC52GJSJ, 1AC52MESJ, 1AP52MESJ, 1AC52SESJ, 1AB52GNSJ, 1AB52MESJ, 1AC52GNSJ, 1AC52MPSJ, 1AC52GKSJ, 1AC54HATS, 1AC54MESJ, 1AP54MQSJ, 1AP54MJSJ, 1AP54MFSJ, 1AP54MESJ, 1AC54MQSJ, 1AC54MPSJ, 1AC54MJSJ, 1AC54MFSJ, 1AB54HATS, 1AB54MESJ, 1AB54MFSJ, 1AB54MQSJ, 1AA55SETS, 1AC55SZSJ, 1AC55DANR, 1AC55SENR, 1AA55SZSJ, 1AB55SETS, 1AB55SZSJ, 1AX55LADV, 1AX55LASJ, 1AX55LAQK, 1AX55LAFT, 1AX52MESJ, 1AX52MBSJ, 1AX52MQSJ,	5529, 5519, 5839, 1036, 5652, 1151, 1152, 4319, 4329, 1123, 4329, 4319, 1139, 4339, 1531, 1532, 1533, 1534, 1539, 1541, 1542, 1543, 1694, 1698, 1699, 1661, 1669, 1690, 1693, 1697, 5811, 5823, 5127, 1052, 5195, 6698, 6693, 1053, 7199, 5142, 1161, 49721, 49731, 4971, 49743, 4983, 4986, 4987, 9805

Appendix 4 (…continued)

1AX53DAFT, 1AX53LAFT, 1AX53HHFT, 1AX53DADV, 1AX53LADV, 1AX54HASJ, 1NM77EP, 1NM77RS, 1NM77RSXXG, 1NK77EN, 1NK84RRXXG, 1NK77RR, 1NK77RRXXG, 1KY76LA, 1KY76LAXXN, 1KY76LAXXL, KY76LAXXA, 1KY76LASJ, 1PE54JATS, 1PZ21HQBR, 1PZ21HPD4, 1OT53DATS, 1OT53HATS, 1OT53LATS, 1PE54BANR, 1PE54DANR, 1PE54LANR, 1PV50BABJ, 1PV50BABM, 1PV50BABP, 1PV57BAAM, 1PV57BAGX, 1PV57LAGX, 1PV59BAAG, 1PV59BAAS, 1PV59BAAT, 1PV59BAAZ, 1PV59BAGX, 1PV59BAX7, 1PV59LAGX, 1PZ94BA, 1PZ94DA, 1PZ94HA, 1PZ94LA, 1KY80LA, 1KY80LAXXA, 1KY80LAXXK, 1KY80LAXXN, 1PE55CATS, 1PE55JATS, 1HB53LAJA, 1HD53GRJA, 1HZ53QANM, 1HZ53QANL, 1HZ53QANK, 1HZ53LANN, 1HZ53LANM, 1HZ53LANL, 1HZ53LANK, 1HZ53GRNN, 1HZ53GRNM, 1HZ53GRNL, 1HZ53GRNK, 1HZ53GRFR, 1HZ53LAFR, 1HZ53SYFR, 1HZ53GRFS, 1HZ53LAFS, 1HZ53HAFS, 1HZ53SYFS, 1HZ55QANM, 1HZ55QANL, 1HZ55QANK, 1HZ55QAFS, 1HZ55LANM, 1HZ55LANL, 1HZ55LANK, 1HZ55LAKP, 1HZ55LAFS, 1HZ55GPNM, 1HZ55GPNL, 1HZ55GPNK, 1HZ55GPFS, 1HB55LAJA, 1HB55LAJB, 1HD55GPJB, 1HD55GRJA, 1HZ38GRNN, 1YY55LANJ, 1AX53LAQK

Appendix 5 RAMQ physician billing codes

RAMQ billing codes used by general practitioners during any encounter—primary and emergency care		
CHSGS (outpatient clinic/emergency) Codes	**Office, CLSC, UMF-CH Codes**	**Details**
00005	08870	Registered patient ORDINARY EXAM <60 years
00056	08871	Registered patient COMPLETE EXAM <60 years
00097	08872	Registered patient MAJOR COMPLETE EXAM <60 years
00006, 00098, 00057	00058	EXAM with URGENCY/TRAVEL
00012, 00002	---	Home visit <70 years
00061		MINOR CONSULTATION <70 years
00060		ORDINARY CONSULTATION <70 years
00062		MAJOR CONSULTATION <70 years

Centre local de services communautaires (CLSC)/Local community services centre, *Unité de médicine familial-centre hospitalier* (UMF-CH)/Family medicine unit-hospital centre, *Centre hospitalier services géreraux ou specialises*(CHSGS)/General and specialized hospital centre

Appendix 5 (...continued)

Billing codes used by pediatricians	
Codes	**Details**
OFFICE	
09194	General exam in office by pediatrician
09127	Main (non-consultative) visit in office by pediatrician
09129	Follow-up visit by pediatrician
09165	Consultation by pediatrician
15538	Consultation by pediatrician for complex pathology
15164	Multidisciplinary or parent meeting in regards to a complex pathology
16099	First evaluation by pediatrician after referral from a health professional other than a doctor
OUTPATIENT HOSPITAL CLINIC	
09162	Main (non-consultative) visit by pediatrician
09170	Consultation by pediatrician
09164	Follow-up visit by pediatrician
15547	Consultation by pediatrician for complex pathology
15166	Multidisciplinary or parent meeting in regards to a complex pathology
16100	First evaluation by pediatrician after referral from a health professional other than a doctor
15186	Main responsibility of care taken by an pediatrician in a day hospital setting
15550	Main visit by pediatrician for a patient under chemotherapy or immunosuppressed
15551	Main responsibility of care taken by an pediatrician for palliative care
HOME VISIT	
09171	Main visit by pediatrician
09172	Follow-up visit by pediatrician
09164	Palliative care visit by pediatrician

Billing codes used by pediatric specialists	
Code	**Details**
09127	Main (non-consultative) visit in office by specialist in office
09129	Follow-up visit by specialist in office
09162	Main (non-consultative) visit by specialist in outpatient clinic
09164	Follow-up visit by specialist in outpatient clinic
15363	Supplement for main visit by specialist in office for patient under 10 years
15368	Supplement for main visit by specialist in hospital outpatient clinic for patient under 10 years
09165	Consultation by specialist in office
09170	Consultation by specialist in outpatient clinic
15148	Pediatric subspecialty outpatient clinic consultation (intensive care, neonatology, infectious disease, emergency medicine, adolescent medicine)
15549	Pediatric subspecialty outpatient clinic consultation (intensive care, neonatology, infectious disease, emergency medicine, adolescent medicine) for complex pathology

Appendix 6 Algorithm to identify "usual provider of primary care

Algorithm to identify "usual provider of primary care
STEP 1 First, use codes for "enrollment" under a family physician. If subject has one of the following codes, then "usual provider of primary care" if a family doctor: 08875, 08877, 15144, 15145, 00059, 15159, 15148, 15169, 15170, 15171, 15158, 19952, 19951, 19954, 19955, 99800, 99500 to 99515
STEP 2 If subjects do not have a codes to identify a family physician as "usual provider of primary care" (STEP 1), search for enrollment by a pediatrician using the 09129 code. This code is not specific to "enrollment" of patients under a pediatrician but it is used by pediatricians for follow-up or growth and development millstones.
STEP 3 If a subject does not have a code to identify a family physician or pediatrician as "usual provider of care" (STEPS 1 and 2), calculate the number of visits by a family physician (09092, 08870 (00005), 08871 (00056), 08872 (00097), 08901 (08807), 08902 (08904), 15161, 15230, 00474 -- brackets mean these are billed by CHSGS/CLSC outpatient clinic) and for each visits by a pediatrician (09194, 09127). Only one act per day per doctor can be included when calculating number of visits. Only physicians with at least 2 visits can be considered for STEP 3. If more than 2 physicians has more than 2 visits, the n the one with the most visits is selected as the "usual provider primary care".
STEP 4 If no "usual provider of primary care" is identified STEPS 1 through 3, then the subject does not have a "usual provider of primary care"

Centre local de services communautaires (CLSC)/Local community services centre, Family medicine unit-hospital centre, *Centre hospitalier services géreraux ou specialisés* (CHSGS)/General and specialized hospital centre

Appendix 7 Full final interaction hazard model

Table 5.4 Interaction terms for adjusted hazard ratios of readmission within 30 days following index hospitalization according to driving distance to a pediatric tertiary care centre and level of medical complexity	
Variables	**HR (95% CI)**
<80km driving distance*Single CCC without TA	**Reference**
≥80km driving distance*Single CCC with TA	0.39(0.11-1.42)
≥80km driving distance*Multiple CCC without TA	0.76(0.42-1.37)
≥80km driving distance*Multiple CCC with TA	0.75(0.32-1.79)
≥80km driving distance*NI CCC without TA	0.79(0.49-1.28)
≥80km driving distance*NI CCC with TA	0.95(0.38-2.35)
Distance (kilometres) <80	Reference
≥80	1.27(0.91-1.78)
Level of Medical Complexity Single without TA	Reference
Single with TA	2.21(1.29-3.77)
Multi without TA	1.97(1.47-2.64)
Multi with TA	3.37(2.17-5.21)
NI without TA	1.42(1.10-1.83)
NI with TA	1.79(1.14-2.79)
Age (years) 2-4	Reference
5-9	1.27(0.99-1.61)
10-13	1.11(0.85-1.45)
14-16	1.15(0.89-1.48)
Gender Female	Reference
Male	1.05(0.88-1.25)
Neighbourhood SES[#] Q1(highest)	Reference
Q2	1.30(0.96-1.6)
Q3	1.29(0.96-1.75)
Q4	1.52(1.13-2.05)
Q5(lowest)	1.52(1.13-2.05)
Residence(urban/rural)[#] Urban	Reference
Strong MIZ	0.91(0.65-1.26)
Moderate MIZ	0.99(0.69-1.43)
Weak MIZ	1.11(0.82-1.5)
Rural	0.86(1.16-1.37)
PCP[#] Yes	Reference
No	0.93(0.77-1.12)

[#] See **Section 4.1.4.3** in text for definitions of neighbourhood SES, residence and primary care provider
Hazard ratio (HR), confidence interval (CI), complex chronic condition (CCC), technology assistance (TA), neurologic impairment (NI), socio-economical status (SES), quintiles (Q1-5), metropolitan influenced zones (MIZ), primary care provider(PCP), kilometers (km)

Appendix 8 R statistical software output for logrank test the probability of 30-day readmission free time according to driving distance to the closest pediatric tertiary care centre

```
Call:
survdiff(formula = surv.obj ~ tdata$distance)

n=6724, 20972 observations deleted due to missingness.

                          N Observed Expected (O-E)^2/E (O-E)^2/V
tdata$distance=less80 5054      379      387     0.151      0.61
tdata$distance=more80 1670      135      127     0.458      0.61

 Chisq= 0.6  on 1 degrees of freedom, p= 0.435
```

Appendix 9 R statistical software output for final hazard model#1

```
Call:
coxph(formula = surv.obj ~ tdata$distance + tdata$CCCTA_cat +
    tdata$age_cat1 + tdata$sexe + tdata$SGC_cat + tdata$pcp +
    tdata$ses_q)
  n= 6563, number of events= 502
   (21133 observations deleted due to missingness)


---Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1

                          exp(coef) exp(-coef) lower .95 upper .95
tdata$distancemore80         1.0747     0.9305    0.8637     1.337
tdata$CCCTA_catSingleTA      1.7705     0.5648    1.0896     2.877
tdata$CCCTA_catMulti_noTA    1.8377     0.5442    1.4250     2.370
tdata$CCCTA_catMultiTA       3.1220     0.3203    2.1406     4.553
tdata$CCCTA_catNI_noTA       1.3335     0.7499    1.0731     1.657
tdata$CCCTA_catNITA          1.7498     0.5715    1.1867     2.580
tdata$age_cat15-9            1.2658     0.7900    0.9970     1.607
tdata$age_cat110-13          1.1089     0.9018    0.8484     1.449
tdata$age_cat114-16          1.1508     0.8689    0.8922     1.484
tdata$sexeM                  1.0497     0.9526    0.8804     1.252
tdata$SGC_catStrong MIZ      0.9096     1.0994    0.6546     1.264
tdata$SGC_catMod MIZ         0.9942     1.0058    0.6915     1.429
tdata$SGC_catWeak MIZ        1.1131     0.8984    0.8248     1.502
tdata$SGC_catRural           0.8539     1.1711    0.5372     1.357
tdata$pcpyes                 0.9332     1.0716    0.7757     1.123
tdata$ses_qQ2                1.2994     0.7696    0.9623     1.755
tdata$ses_qQ3                1.2919     0.7740    0.9560     1.746
tdata$ses_qQ4                1.5201     0.6579    1.1289     2.047
tdata$ses_qQ5                1.2179     0.8211    0.8944     1.659

Concordance= 0.592  (se = 0.013 )
Rsquare= 0.009   (max possible= 0.738 )
Likelihood ratio test= 61.41  on 19 df,   p=2.312e-06
Wald test            = 65.77  on 19 df,   p=4.58e-07
Score (logrank) test = 68.5  on 19 df,   p=1.629e-07
```

Appendix 10 R statistical software output for Schoenfeld residuals for Cox Proportional Hazard Model#1

```
                               rho   chisq      p
tdata$distancemore80       0.01748  0.1508 0.6978
tdata$CCCTA_catSingleTA   -0.01168  0.0689 0.7930
tdata$CCCTA_catMulti_noTA -0.00792  0.0314 0.8594
tdata$CCCTA_catMultiTA     0.04067  0.8259 0.3635
tdata$CCCTA_catNI_noTA    -0.02520  0.3215 0.5707
tdata$CCCTA_catNITA        0.00759  0.0289 0.8651
tdata$age_cat15-9          0.02966  0.4489 0.5028
tdata$age_cat110-13       -0.02311  0.2689 0.6041
tdata$age_cat114-16       -0.03373  0.5746 0.4485
tdata$sexeM               -0.01410  0.1011 0.7505
tdata$ses_qQ2              0.04415  0.9856 0.3208
tdata$ses_qQ3              0.02345  0.2761 0.5993
tdata$ses_qQ4              0.05179  1.3197 0.2507
tdata$ses_qQ5              0.03427  0.6092 0.4351
tdata$SGC_catStrong MIZ   -0.06079  1.8636 0.1722
tdata$SGC_catMod MIZ      -0.03039  0.4485 0.5030
tdata$SGC_catWeak MIZ     -0.02610  0.3402 0.5597
tdata$SGC_catRural         0.02583  0.3331 0.5638
tdata$pcpyes              -0.07561  2.9000 0.0886
GLOBAL                         NA 11.6044 0.9018
```

Appendix 11 R statistical software output for Cox proportional hazard model#2 (including interaction terms)

```
Call:
coxph(formula = surv.obj ~ tdata$distance + tdata$CCCTA_cat +
    tdata$age_cat1 + tdata$ses_q + tdata$SGC_cat + tdata$pcp +
    tdata$sexe + tdata$CCCTA_cat * tdata$distance)
  n= 6563, number of events= 502
   (21133 observations deleted due to missingness)

---Signif. codes:  0 ‘***’ 0.001 ‘**’ 0.01 ‘*’ 0.05 ‘.’ 0.1 ‘ ’ 1
```

	exp(coef)	exp(-coef)	lower .95	upper .95
tdata$distancemore80	1.2747	0.7845	0.9153	1.775
tdata$CCCTA_catSingleTA	2.2056	0.4534	1.2906	3.769
tdata$CCCTA_catMulti_noTA	1.9726	0.5069	1.4726	2.642
tdata$CCCTA_catMultiTA	3.3651	0.2972	2.1739	5.209
tdata$CCCTA_catNI_noTA	1.4209	0.7038	1.1012	1.833
tdata$CCCTA_catNITA	1.7849	0.5602	1.1434	2.786
tdata$age_cat15-9	1.2645	0.7908	0.9958	1.606
tdata$age_cat110-13	1.1067	0.9036	0.8466	1.447
tdata$age_cat114-16	1.1497	0.8698	0.8913	1.483
tdata$ses_qQ2	1.3012	0.7685	0.9635	1.757
tdata$ses_qQ3	1.2932	0.7733	0.9570	1.747
tdata$ses_qQ4	1.5207	0.6576	1.1293	2.048
tdata$ses_qQ5	1.2204	0.8194	0.8961	1.662
tdata$SGC_catStrong MIZ	0.9072	1.1023	0.6527	1.261
tdata$SGC_catMod MIZ	0.9919	1.0081	0.6894	1.427
tdata$SGC_catWeak MIZ	1.1111	0.9000	0.8231	1.500
tdata$SGC_catRural	0.8616	1.1606	0.5415	1.371
tdata$pcpyes	0.9309	1.0742	0.7735	1.120
tdata$sexeM	1.0483	0.9539	0.8792	1.250
tdata$distancemore80:tdata$CCCTA_catSingleTA	0.3945	2.5347	0.1096	1.421
tdata$distancemore80:tdata$CCCTA_catMulti_noTA	0.7566	1.3217	0.4181	1.369
tdata$distancemore80:tdata$CCCTA_catMultiTA	0.7535	1.3272	0.3174	1.788
tdata$distancemore80:tdata$CCCTA_catNI_noTA	0.7904	1.2652	0.4866	1.284
tdata$distancemore80:tdata$CCCTA_catNITA	0.9469	1.0561	0.3813	2.351

```
Concordance= 0.595  (se = 0.013 )
Rsquare= 0.01   (max possible= 0.738 )
Likelihood ratio test= 64.61  on 24 df,   p=1.39e-05
Wald test            = 68.66  on 24 df,   p=3.484e-06
Score (logrank) test = 71.69  on 24 df,   p=1.209e-06
```