THE REGIONAL DISPARITY OF CONGENITAL ANOMALIES IN SASKATCHEWAN AND ITS IMPACT ON THE UTILIZATION OF HEALTH SERVICES

A Thesis Submitted to the College of Graduate Studies and Research in Partial Fulfillment of the Requirements for the Degree of Masters of Science in the Department of Community Health and Epidemiology

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ABSTRACT

Congenital anomalies (CAs) are the leading cause of infant mortality and one of the leading causes of death for young children in developed countries. As significant improvements have been seen world-wide in controlling childhood infectious disease and issues related to poor nutrition, CAs are now making a proportionally bigger impact on the health of the world's children. In addition to the impact of CA status on the individual child and one's family, prevalence of CAs has a significant impact on the population, as children with birth defects can cost the system a great deal of money in the provision of specialized health and education services.

When conducting surveillance of five selected CAs between 1990 and 1999, Saskatchewan Health found significant regional differences in the prevalence of these CAs. The purpose of this study is to ascertain whether or not there is a regional difference in all types of CAs, to assess whether or not any regional disparities also exist in the use of health care services by children with and without CAs and to determine what factors influence children's use of health care services in the study population.

This study follows a birth cohort of 17,414 children (9169 cases and 8245 controls) born between January 1, 1994 to December 31, 1998 until their 5^{th} birthday, death or emigration out of Saskatchewan. Through graphical analysis, it was revealed that while an overall regional difference does not exist in the prevalence of CAs in Saskatchewan, there are regional differences in the prevalence of 13 of the 22 specific categories of conditions studied. One-way ANOVAs showed that children with CAs have higher numbers of physician visits ($p<0.001$) and hospitalizations ($p<0.001$), and longer lengths of stay in hospital ($p<0.001$) than children without CAs. Regional

differences were found for all outcome variables for the total population, and for children with and without CAs. The outcome with the most substantial differences between children with and without CAs was length of stay, which may indicate differential access to outpatient services throughout the province. Finally, using Anderson's theoretical framework of factors that influence the use of health care services (need characteristics, predisposing characteristics and enabling characteristics) three negative binomial models were built to examine children's use of health care services using variables from each category.

This study found significant regional differences for all outcome measures studied, and found that region of residence was a significant predictor of children's use of health care services even after accounting for a variety of other maternal and child factors.

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CHAPTER ONE: INTRODUCTION

Congenital anomalies (CAs) are any abnormalities that are present at birth, even if they are not detected until much later (1, 2). In developed nations, CAs are the leading cause of infant mortality, and one of the leading causes of death for young children (3, 4). While the risk of dying as a result of a CA (or multiple CAs) has decreased between 1950 and 2000, the rate of decline has slowed in recent years; infant mortality (deaths to live born children in the first year of life), however, remains a significant issue in our society today (3, 5). As significant improvements have been seen world-wide in controlling childhood infectious diseases and issues related to poor nutrition, CAs now have a proportionally bigger impact on the health of the world's children (6, 7). In addition to the impact of CA status on the individual child and family, prevalence of CAs has a significant impact on the population, as children with birth defects can cost the system a great deal of money in the provision of specialized health and education services (7). CA status is likely to be a major predictor of children's use of health care services as these children may need to use a higher level of services to treat and/or manage their birth defect and they may be more susceptible to other comorbidities due to the presence of a CA than unaffected children.

1.1 Study Rationale

In 2000, Saskatchewan Health released a report entitled 'The Epidemiology of Infant Mortality in Saskatchewan 1982-1996'. To date this is the only study of its kind in

Saskatchewan. This report showed that while the absolute number of infant deaths due to congenital anomalies decreased by 33% from 246 in the first five year period (1982-1986) that the study considered to 165 in the second five year period (1992-1996), the proportion of deaths due to CAs remained stable at approximately 28% throughout the entire study (5). This indicates that the importance of CA status as it relates to infant mortality has not lessened over time. A nation-wide study found similar results (8). In Canada, the rate of infant mortality due to lethal congenital anomalies decreased from 3.11 per 1000 live births in 1981 to 1.89 per 1000 live births in 1995, this represents 30% and 34% of infant mortality respectively (8). This same study examined provincial differences in the rates of infant mortality due to lethal CAs and found that the province of Saskatchewan had a significantly higher overall rate of infant deaths due to CAs than the province of Quebec which served as the reference group (2.48 deaths per 1000 live births versus 1.91 deaths per 1000 live births) (8).

These findings, along with more current unpublished data collected by the Population Health Branch at Saskatchewan Health, shows that both rates of infant mortality and CAs are not consistent across all health regions (5, 9). Figure 1.1 shows regional differences for the combined prevalence of several selected CAs: neural tube defects (NTDs), limb reduction deficits, Down syndrome, cleft lip/cleft palate and congenital heart defects. The prevalence of these conditions ranges from a low of 54 per 1000 live births in the Cypress Health Region to a high of 163 per 1000 live births in Northern Saskatchewan (this includes the Keewatin Yatthé Health Authority, the Mamawetan Churchill River Health Authority, and the Athabasca Health Authority).

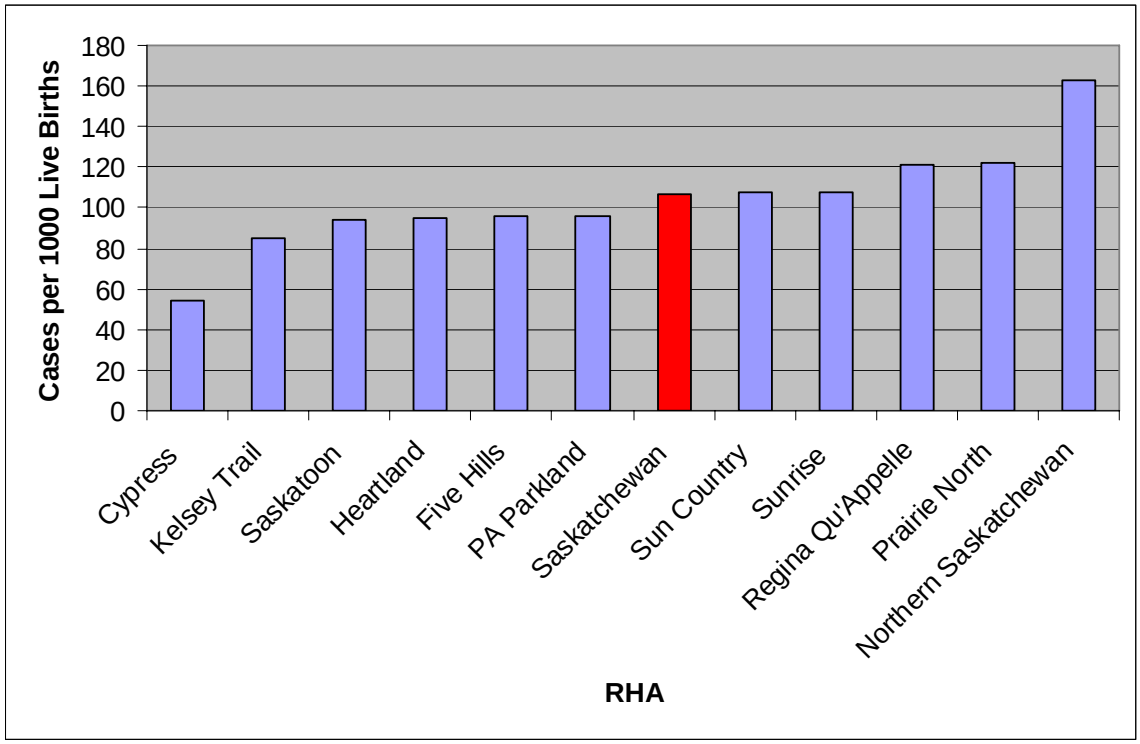


Figure 1.1: Combined prevalence of five selected CAs (neural tube defects, limb reduction deficits, Down syndrome, cleft lip/cleft palate and congenital heart defects) by regional health authority (1990-1999) (9)

While it is known that there is a regional disparity in the rates and types of CAs, it is not known whether this same disparity extends to the health outcomes of children born with CAs in their first five years of life. By further analyzing the regional differences in CAs and the use of health care services by region for children with CAs as compared to those without CAs in the first five years of life, a better understanding of CAs and the subsequent health care burden in Saskatchewan can be achieved. By examining regional differences with regards to various aspects of population demographics that have been shown to have an effect on healthy child development, the determinants of regional disparities will be revealed along with information on how to allocate resources to better manage the care of vulnerable children in Saskatchewan (10).

1.2 Objectives and Research Questions

The purpose of this study is to determine whether or not any regional disparities exist in the use of health care services for children with and without CAs, and to understand what factors influence children's use of health care services in the study population.

This thesis will address three principal questions:

- Question One: Is the level of health care used by children with CAs significantly different from the level of health care used by children without CAs?
- Question Two: Is there a regional difference in the level of health care used by children in their first five years of life? Does this relationship hold for children with and without CAs?
- Question Three: What factors influence the level of health care utilization in the first five years of life for children in Saskatchewan?

It is hypothesized that children with CAs will utilize significantly more health care services than children without CAs in their first five years of life. Furthermore, it is believed that this relationship will be significantly affected by a variety of factors related to one's illness level (need), factors that make certain individuals more inclined to access health care services such as one's values, socio-economic status and gender (predisposing characteristics) and factors that permit someone to access services such as the availability of nearby health services (enabling characteristics) (11).

This type of research is important because if a regional difference is found in the use of health care services (especially for children with congenital anomalies, an already vulnerable population), it provides strong evidence to the regional health authorities and the provincial ministry of health that more needs to be done to "equalize" the differential health care utilization patterns across regions.

CHAPTER TWO: LITERATURE REVIEW

This chapter provides an overview of the literature around the major themes of this study: congenital anomalies, health disparities and health care utilization. While some studies exist that tie two of these three themes together, no studies could be found that link all three themes. The chapter begins by describing congenital anomalies (definitions, causes, types and prevention), next is a discussion on health disparities and how geographical health disparities relate to healthy child development, and finally a discourse on the factors that contribute to one's use of health care services. These sections are followed by a discussion on the provision of health care services in Saskatchewan and finally the use, validity and reliability of administrative databases in health research.

2.1 Congenital Anomalies

2.1.1 What is a Congenital Anomaly?

The term congenital anomaly (also known as [a.k.a.] birth defect, congenital malformation, congenital abnormality) encompasses any abnormality that is present at birth, even if it is not detected until much later (1, 2). Various sources estimate the prevalence of CAs to be in the range of 1-3% of all live born infants (and considerably higher for infants that are stillborn or spontaneously aborted) (2, 12, 13). This rate increases to 5-6% when the ascertainment period is extended to the age of five or six years (2, 12, 13). CAs can be subdivided into major and minor anomalies related to

their clinical significance (1). In addition to these types of CAs, there are normal variations of development that are seen in all individuals (1).

There are four clinically relevant types of CAs: malformations, disruptions, deformations and dysplasia (1). A malformation is a "morphological defect of an organ, part of an organ, or larger region of the body that results from an intrinsically abnormal developmental process" (1). A disruption is a "morphological defect of an organ, part of an organ, or larger region of the body that results from the extrinsic breakdown of, or an interference with, an originally normal developmental process" (1). A deformation is "an abnormal form, shape or position of a part of the body that results from mechanical forces" (1). Dysplasia is "an abnormal organization of cells into tissue(s) and its morphological result(s), … [it is] causally nonspecific and often affects several organs because of the nature of the underlying cellular disturbances" (1).

Physical defects develop during the period of organ formation called organogenesis (weeks 3-11 of pregnancy), while most CAs that cause developmental delay occur later in pregnancy when the brain is maturing (1). Figure 2.1 illustrates the sensitive stages of development for the various organ systems.

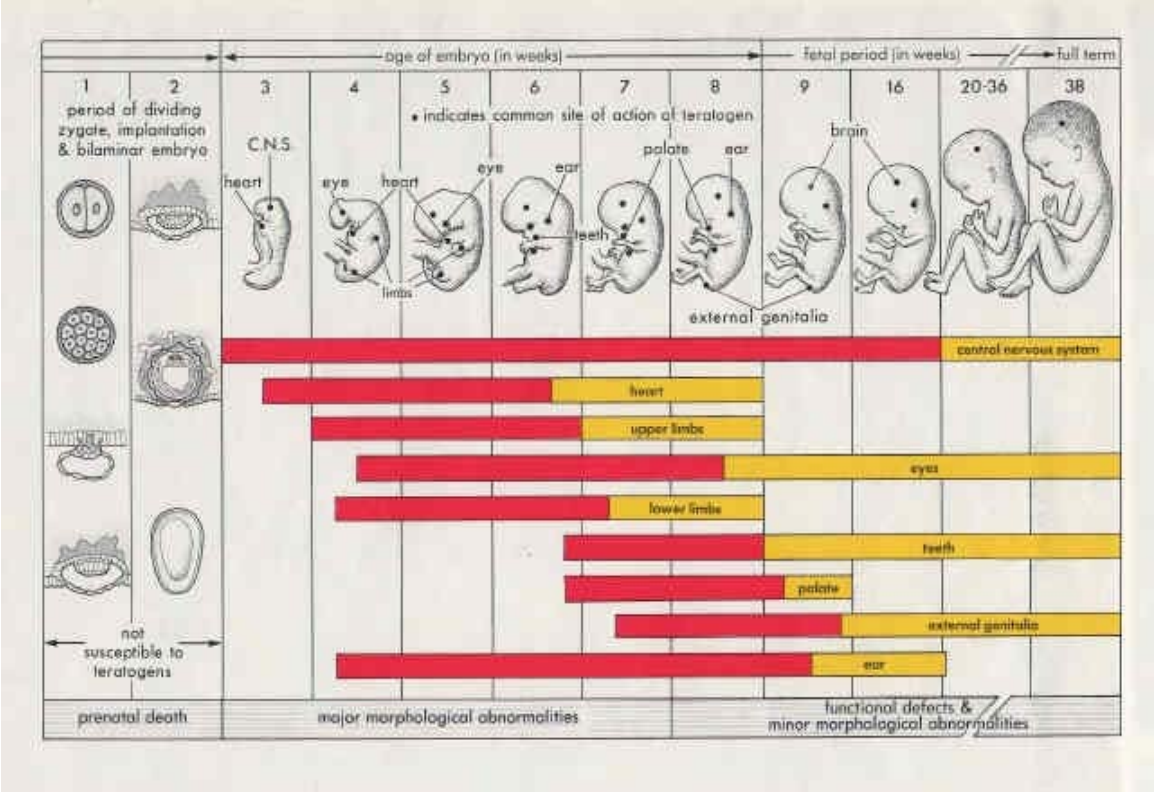


Figure 2.1: Sensitive stages of development (1)

2.1.2 Causes of Congenital Anomalies

As seen in Figure 2.2, the majority of CAs are of unknown origin, which makes prevention problematic. Generally CAs, of known origin, are due to one of three principal causes: genetic factors, environmental factors, or a combination of genetic and environmental factors (multifactorial inheritance) (2).

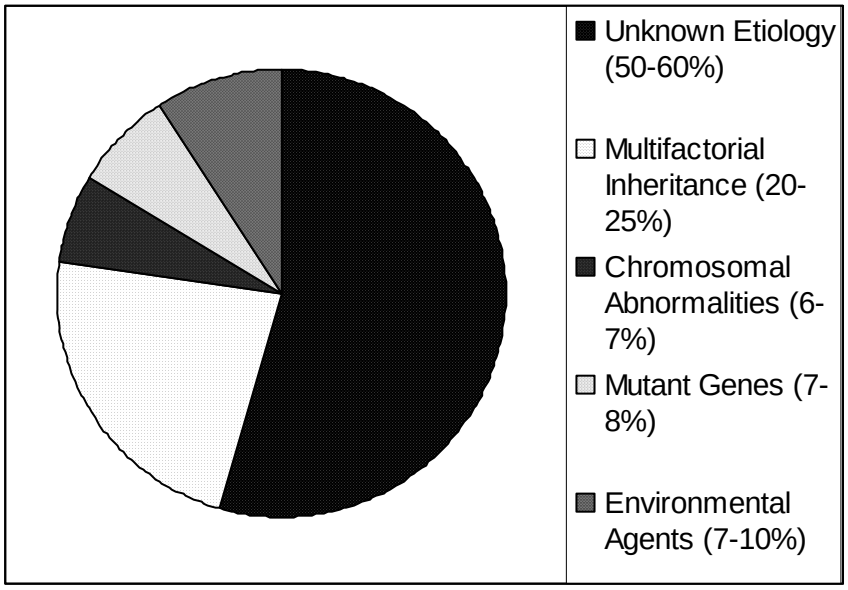


Figure 2.2: Causes of congenital anomalies (2)

After the thalidomide tragedy in the 1950s, a great deal of emphasis was placed on the potentially harmful role that drugs can play in the development of CAs. While Thalidomide is an extreme example of the potential teratogenicity of a pharmaceutical product, only 1% of CAs with a known cause are attributed to drug therapy (13). Furthermore, there are only approximately 25 drugs that are currently in use that are known to have a teratogenic effect (13).

In addition to pharmaceuticals, other environmental agents that have been shown to cause CAs include: maternal behaviours such as smoking, alcohol use, and poor nutritional status; infectious agents such as rubella, syphilis, and herpes simplex virus; high-dose ionizing radiation; and environmental contaminants such as herbicides, pesticides, and methyl mercury (14). When examining the potential teratogenicity of an environmental agent, one must keep in mind that for an agent to act as a teratogen, the fetus must have been exposed to at least the threshold dose, during the sensitive period of development for which that particular substance is known to have an effect (see

Figure 2.1) (14). Only a small percentage of CAs are caused by things in one's environment. The largest known cause of CAs is genetics (14). Genetic causes of birth defects can be either autosomal or sex-linked in nature, recessive or dominant traits, single-gene or multiple-gene disorders, chromosomal defects, or be related to new mutations in the fetus (14).

2.1.3 Types of Congenital Anomalies

CAs – regardless of their cause – can affect any organ or system in the body, yet some types of CAs are more common then others (1). Most CAs can be classified under the general categories of musculoskeletal defects, congenital heart defects, digestive system defects, circulatory system defects, central nervous system defects, urinary system defects and genital organ defects. Figure 2.3 illustrates the prevalence of the most common groups of CAs in Canada in 1995 (15).

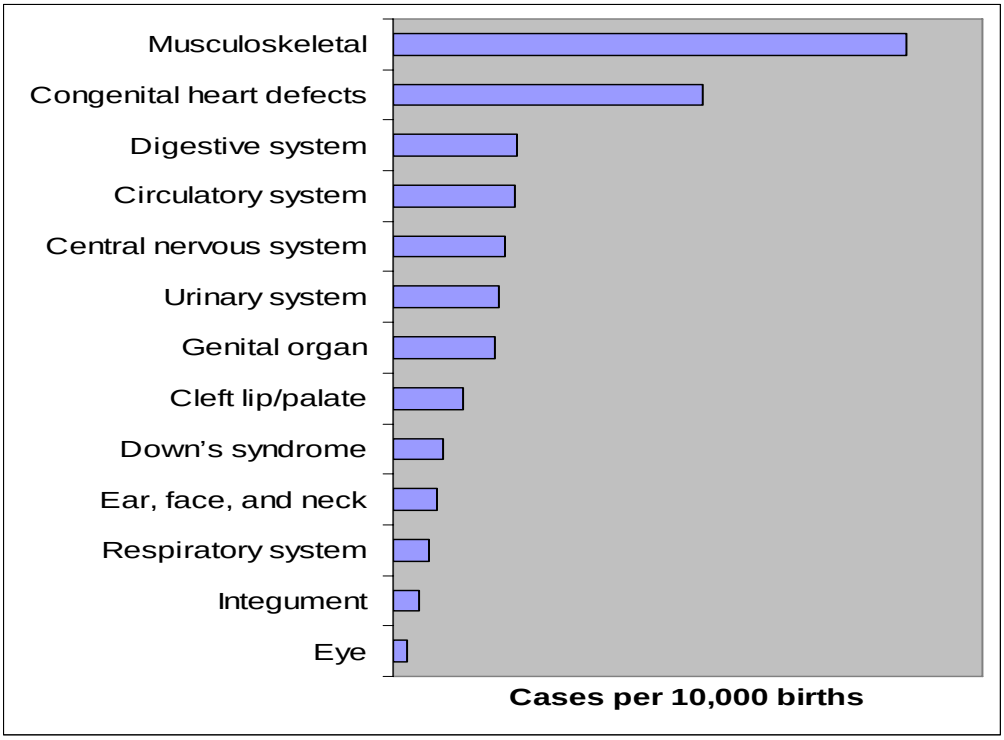


Figure 2.3: Prevalence of the most common types of CAs in Canada (*excluding Nova Scotia and Quebec) per 10,000 births (15)

A study conducted in Glasgow, UK examining the prevalence of selected CAs from 1980 to 1997 found that during this time period in Glasgow, the prevalence of most CAs declined (6). Overall, statistically significant decreases in prevalence were seen for CAs of the ear (88% decrease), CAs of the heart (69% decrease), CAs of the integument (67% decrease), CAs of the nervous system (61% decrease), CAs of limbs (54% decrease), and CAs of the urogenital system (including the renal system) (31% decrease) (6). In this same time period, an increase was seen in chromosomal abnormalities (50% increase) (6). Despite an overall decrease in the prevalence of CAs from 382 per 10,000 births in 1980 to 238 per 10,000 births in 1997, the proportion of affected children remained stable around 2.5% (6).

2.1.4 Prevention of Congenital Anomalies

The prevention of birth defects is an important public health issue as birth defects tend to reoccur in families due to the shared genetic and environmental factors (16). Additionally, a longitudinal, population-based study conducted in Norway examining the survival of females with birth defects found that only 80% of those with birth defects survived until their 15th birthday compared to 98% of subjects without birth defects (i.e. children with CAs were more likely to die before their 15th birthday than children without CAs) (16). This study went on to examine the likelihood of females with birth defects to have children by the age of 30 compared to their non-affected peers, and found that women with birth defects were one third less likely to give birth in this time period (16). Additionally, the children of women who had a birth defect were more likely to have a birth defect themselves than the children of women without birth defects; however, this increased risk was only for the condition that affected the mother,

not birth defects in general (i.e. women with cleft palate had a higher risk of having a child with cleft palate, but not with a congenital heart defect) (16). The increased relative risk of birth defects in the offspring of women with birth defects ranged from 5.5 to 82 depending on the defect (16).

When discussing the "prevention" of CAs, quite frequently prevention is used as a pseudonym for early termination. While some large-scale prevention practices have been implemented (such as the fortification of foods with folic acid to prevent neural tube defects) and some educational programs have shown some degree of success in encouraging pregnant women to adapt healthier lifestyles, many CAs cannot be prevented.

Screening healthy women for disease and their unborn baby's risk of disease has become part of the routine practice of prenatal care, as advances in medical diagnostic technology has allowed these tests to be administered more easily, safely and cheaper than ever before (17, 18). This practice of routine screening (especially when women are considered "high-risk" due to having had a previous child with a congenital anomaly, is of advanced maternal age, or have certain pre-existing conditions) can have many benefits – it may help provide peace of mind and reduce stress to know that one's child is unlikely to have a certain condition, or if it is revealed that the child has a CA, it provides time for families to decide how they would like to proceed (19). That being said, no test is perfect, and false-positive results can be extremely distressing and sometimes can result in the termination of an unaffected fetus (19, 20). Just as distressing, can be the psychological impact of a false-negative result when parents were advised that their child was not going to have a CA, only to find out once the child is

delivered that s/he has a potentially serious disability (20). In addition to the psychological burden that can be associated with the routine screening for certain CAs in pregnancy, there is a minefield of ethical issues surrounding this practice that involves society's acceptance of disabled persons, what kind of life is worth living and who is able to make that decision for others, and the "eugenic thrust in the practice of selectively aborting fetuses with disabilities" (18). This is not to imply that a woman who chose to abort a fetus with a CA is practicing eugenics, merely that as a whole, society needs to be more accepting of individuals with disabilities.

While the actual impact of the routine testing for CAs in the antenatal period is unknown, it is suspected that there is a strong correlation between the decrease in the prevalence of specific CAs (such as anencephaly and spina bifida) and the increase in screening for specific CAs (21). Many CA surveillance systems are not able to capture the true incidence of CAs as they tend to only record CAs for live born infants, stillbirths when the cause is known, or fetuses who are carried beyond a certain gestational age. Therefore it is impossible to ascertain whether there has really been a decrease in rates of specific CAs in recent years or if there has simply been an increase in prenatal diagnosis of these CAs and a subsequent increase in early terminations of these pregnancies (21). A Canadian study by researchers for the Fetal and Infant Health Study Group of the Canadian Perinatal Surveillance System found that between 1991 and 1997 fetal deaths from pregnancy terminations increased by 578%, or almost 6-fold, with the most significant increase occurring in 1995 (22). The researchers also found that while infant mortality rates due to congenital anomalies had remained stable from 1991 to 1995, there was a 21% decrease between 1995 and 1996, and that infant

mortality rates due to CAs had remained low in 1997 (22). During this same time period, both the rate of prenatal testing for CAs and the selective termination of affected pregnancies were increasing, both of which are related to the overall decrease in Canada's infant mortality rate (22).

2.2 Health Disparities

2.2.1 What is a Health Disparity?

A health disparity (a.k.a. inequality) is a difference between two or more population groups on the basis of a specific criterion related to one's health status (23, 24). Some definitions are more specific as they define a health disparity as a difference in health status that is unnecessary, avoidable, unfair and unjust (known as health inequities as opposed to inequalities) (23). Many disparities are caused by inequities.

Disparities have been noted for various population groups for all of Health Canada's determinants of health (income and social status, physical environments, social environments, personal health practices and coping skills, social support networks, biology and genetic endowment, culture, gender, health services, healthy child development, education, employment and working conditions) (23, 25). This project will examine only two of these determinants – healthy child development and health services.

2.2.2 Geographic Health Disparities and Healthy Child Development

Healthy child development has one of the most far-reaching effects of all the health determinants, as it affects the way a child's brain develops, which in turn reflects his/her success in school (which will have an impact on the amount of education a child receives, the type of job s/he gets, how much money an individual will make and what

sort of physical environment one will live in) (23). Furthermore, healthy child development helps children develop their social skills, which in turn impacts their coping abilities later in life and their personal health practices.

While several studies have shown a disparity in children's health across communities with regard to socioeconomic status (SES), the availability of health services, and various other demographic factors, no published work appears to exist that can explain why there is such a regional disparity with regard to CA rates in Saskatchewan or what impact this disparity has on the overall health of these children (10).

While individual factors are known to have an impact on health, the social environment in which one lives also has an effect over and above individual characteristics (26). While in Canada it is known that health outcomes differ at the regional level, it is still unknown to what extent this regional disparity is due to the composition of the population in each area and the social context in a region (26). Generally it is believed that individuals who live in the same health region tend to be more alike than individuals living in a different health region as they share similar experiences related to things such as the environment, health care services, culture and health behaviour (26). These conclusions are questionable in large regions with diverse populations that encompass both inner-city and rural areas as is seen in Saskatchewan; however, may be accurate for more homogeneous areas. Tremblay and Berthelot concluded that regional differences with regard to the availability of health care services are not a factor in the disparities of individual health status that exist between regions (26).

It has been shown that individuals who live in neighbourhoods with low SES during their pregnancy are more likely to have an adverse birth event (i.e. having a child with a CA or having a low birth weight baby) than individuals who live in a neighbourhood with a higher level of SES (10). Researchers found that low SES residents who lived in low-SES neighbourhoods and low SES residents who did not live in low-SES neighbourhoods both had an increased risk of having a child with a neural tube defect (27).

A study conducted in Ireland in the early 1990s determined that children living in poor areas were approximately nine times more likely to be hospitalized for any reason than children who did not live in poor areas (28). A more recent Canadian study indicated that in their first year of life, children in low SES families use more treatment related health services and less preventative health services than children in higher SES families; and that parental education plays a bigger role in determining the use of health services than parental income (29).

While this information on the impact of SES (measured by parental income and education level) on the risk of having a child with a CA and the child's use of health care services is interesting, SES alone cannot explain the regional difference in CA rates; nor does it provide enough evidence to accurately predict whether or not children with CAs in a particular health region will use a significantly different amount of health services compared to children with CAs in another health region. This study will be able to begin to answer these questions.

In addition to health disparities as they relate to socioeconomic status, in Saskatchewan there is a need to examine disparities as they relate to access to health

services (most especially in remote communities in the northern part of the province) and the gross health disparities that exist between Aboriginals and non-Aboriginals (30). Studies have shown that health care is less accessible for rural residents than urban residents and that this problem is further magnified for remote communities (31).

It is well known that Aboriginal people in Saskatchewan experience many health disadvantages. Multiple studies have shown that people of Aboriginal ancestry in Canada, and elsewhere throughout the developed world, suffer from more health problems than the general population (32-35). These health disparities are not limited to Aboriginal people living in urban environments but also those living on reserves or in isolated communities. Geographic isolation has been shown to negatively impact health status as access to health professionals and services, in particular for prevention, and secondary treatment is often challenging for residents in remote or isolated locations (34, 36). Finally, many Aboriginal people are living in poverty which further impacts their health status (33, 35).

2.3 Health Services Utilization

In 1968, the federal government approved the Medical Care (Medicare) Act, which granted medical insurance to all Canadian citizens free of charge by removing payments from the point of service (37). Almost twenty years later, the Canada Health Act was passed to ensure that all of the Canadian provinces and territories upheld the principles of accessibility, comprehensiveness, portability, public administration and universality in order to continue receiving federal transfers for health care (38). These two pieces of legislation act as the backbone on which the Canadian health care system today is based on. They ensure that all Canadians, regardless of what province they live in, or if they

live in a rural area or an urban centre have access to medically necessary services without financial impediments (31, 39, 40).

Use of health care services is commonly believed to be a type of individual behaviour, with the volume of services used determined by the predisposition of an individual to use health services, the person's ability to access services and how sick an individual is (11). Figure 2.4 outlines Anderson and Newman's model of the individual determinants of health services utilization. Predisposing determinants are factors that are present before the illness begins and they explain in part why some people use services more than others; enabling determinants are characteristics that represent how people use health services; and need, or illness level, represents a person's current health status (11, 41). Poor health is the most immediate predictor for health care utilization (11, 41).

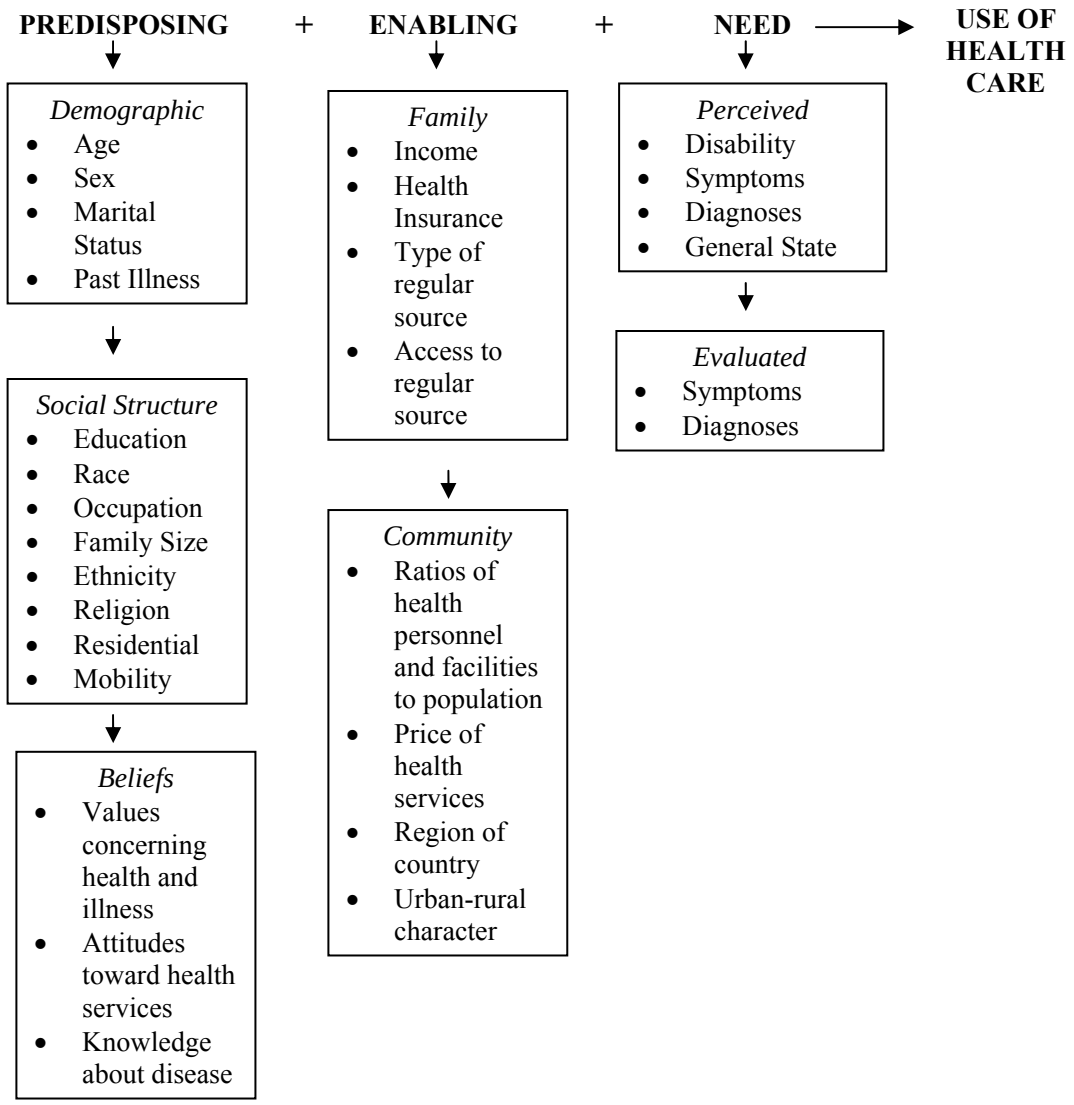


Figure 2.4: Individual determinants of health services utilization (11).

A meta-analysis conducted by a nurse-researcher at the University of Alberta examined the barriers and facilitators in the health care relationship that either prevented or encouraged Canadians with chronic diseases to access health services (please see Table 2.1 for a summary of the findings) (39). It is interesting to note that despite the broad inclusion criteria for this meta-analysis, and the twelve-year study period (1990-2002), the researcher found that there is a significant lack of research on the

geographical barriers to access, especially in remote areas such as the Northwest Territories and Nunavut (39).

Table 2.1: Barriers and facilitators to accessing health services (39)

Barriers	Facilitators
Poor relationship with service provider as characterized by provider disbelief of family or client perceptions, undervaluing/devaluing of client or family knowledge, inappropriate use of power, provision of inadequate information to clients	Open/trusting relationship between service provider and patient
Previous negative experiences of clients with service providers and fear of privacy violations	Personal follow-up contact by service provider (i.e. appointment reminders, etc)
Gender or sexual identity differences between patients and service providers	Same gender/sexual identity between service providers and clients
Language or cultural differences between clients and service providers	Service provider displays sensitivity and understanding of client culture; advice by service provider fits with the cultural beliefs of the client
Differences in beliefs between marginalized groups and service providers, along with fear of discrimination, stigma or humiliation	Client knowing someone who works in the system who can advocate on his/her behalf
Differences in generational values	Personal/social connection between patient and service provider
Uncertainty or fear about the outcome of the encounter	

While Anderson and Newman's model and information on barriers and facilitators to access are widely used to understand adult's use of health care services, it is unknown how adaptable either of these models are to children, where typically predisposing characteristics of the parents, and things that inhibit or encourage parental access to care are more likely to influence the child's use of health care services than the child's predisposing characteristics. There is a documented association between maternal use of selected health care services and children's use of the same level of health care services for: any doctor visits, six or more doctor visits (in a one year

period), any emergency room visits, any hospitalizations, and any mental health visits (42). Another study found that, in Sweden, geographic location (i.e. urban versus rural) did have an impact on adult's use of health care services, although it did not impact children's use of health care services in the first seven years of life (43). This same study found a correlation between high-consulting children and sicker parents, indicating that disease is in large part a family matter and was not unique to one individual within the family (43).

Even though it is not uncommon to see small-area variations in the use of health care services, one must be cautious when interpreting these differences (44). Differences in the use of health care services between areas could arise due to any number of 'systems related' factors such as the population's need for services, the availability of health care services, people's ability to pay (although this is not common in Canada where the individual patient does not pay directly at the point of service for most health care services), variations in local medical cultures, or clinical uncertainty (44). One must also consider that the small-area variations may be seen merely due to chance, bias and/or unaccounted for confounding (44).

Despite the fact that financial barriers that prohibit access to medical care are believed to have been removed in Canada, one cannot forget other barriers to care brought about by low income. A study in the United States found that the parents of low-income children as compared to the parents of middle-to-high income children were more likely to report having difficulty getting a referral to specialists (2.4% vs. 1.0%), and that their health care provider never or only sometimes listened carefully to them (10.0% vs. 5.1%), explained things clearly to them (9.6% vs. 3.4%), or showed respect

for what they had to say (9.2% vs. 4.2%) (45). This same study found that middle-to-high income children were less likely than low-income children to visit an emergency room (11.5% vs. 14.6%) (45). Additionally, low-income children were more likely to be admitted to the hospital for ambulatory case sensitive (ACS) conditions (conditions that are amenable to primary care intervention and if treated appropriately should not lead to subsequent secondary/tertiary care) (45, 46). This is indicative of the middle-to-high income children receiving more appropriate or timely care from primary health practitioners, which generally results in higher income children being able to avoid more costly tertiary (in hospital) care (45). Similar findings have also been reported in Canada (41).

A study conducted in Nova Scotia examined the differences in the receipt of obstetric services by socio-economic status and found that affluent women were equally likely to have a cesarean section or labour induction than less affluent women (30). However, some important differences were noted; the researchers found that a larger proportion of pregnant women over 35 years of age came from more affluent households, and that this phenomenon was even more pronounced when it came to nulliparous women (30). This finding has direct implications on this study as advanced maternal age is associated with a variety of CAs – most notably Down Syndrome (47). Other notable findings from the study by Joseph et al. include: women in a lower socioeconomic group were less likely to attend prenatal classes or be married, and were more likely to smoke, weigh 75kg or more, and live in rural areas (30). These differences are important because they encompass a variety of lifestyle choices that can have negative impacts on child development. Maternal smoking is a significant public

health concern as most women who smoke during their pregnancy will continue to smoke after giving birth, meaning that their children have not only been exposed to nicotine and other harmful chemicals in utero, but also to environmental tobacco smoke (ETS) in the home (48). Both of these situations have been shown to negatively impact child development as maternal smoking has been linked to low birth weight (<2500g), very pre-term birth (<32 weeks gestation), perinatal death and lower rates of breastfeeding initiation and duration; furthermore, ETS has been shown to be associated with lower respiratory infections, Sudden Infant Death Syndrome (SIDS), middle ear disease and asthma in children (49, 50).

When examining the use of health care services in Canada it is important to acknowledge the difficulties faced by rural and remote communities. Access to health professionals and services in rural areas, and even more so in the North, is a problem – physicians are often few and far between and typically do not stay in these communities on a permanent basis (36). In addition to challenges caused by the lack of health professionals, harsh weather conditions in the winter often impedes travel to other communities and to health care facilities further south (36).

There is also a need for culturally appropriate care to address the unique health care needs of Saskatchewan's Aboriginal population. First Nations' health clinics exist on reserves to provide culturally appropriate, interdisciplinary care to those who live on the reserves (33, 51). These same benefits are not available for Aboriginals who live in urban communities (33, 51).

2.4 Saskatchewan Context

It has long been a source of pride for Saskatchewan residents that Saskatchewan has been an innovator in health care for the past fifty years (52, 53). In 1947, Saskatchewan was the first Canadian province to provide universal hospital care insurance to its citizens, and again in 1962, Saskatchewan was the first province to provide comprehensive medical insurance to all of its citizens (52).

The years after World War II were marked by tremendous growth in the health care industry in Canada, but most particularly in Saskatchewan. Thanks to increased federal funding transfers, provinces were able to construct new hospitals using 50¢ dollars, as every dollar that the provinces spent constructing hospitals was matched by the federal government (53). In this era, the government of Saskatchewan decided that a series of small, rural hospitals was the best way to provide health care to its population as a large proportion of the population lived in rural communities, and travel between communities was difficult due to harsh winters and poor road conditions (53). And while this decision made sense at the time, it didn't take long before this delivery system for health services became obsolete (53). As early as 1961, reports commissioned by the provincial government argued that Saskatchewan no longer required its small rural hospitals (53). While eight (out of a recommended 38) hospitals were closed in 1967, and an additional three were closed in the early 1970s (53). No more hospitals were closed until the 1990s due to fear of political retribution (53). In fact, in the late 1980s and the early 1990s a great deal of money was spent building additional rural hospitals and renovating existing ones (53). In 1992 there were 134 hospitals in Saskatchewan serving approximately one million people (52). In comparison, Quebec, whose

population is approximately seven times the population of Saskatchewan, had far fewer hospitals (52).

In the 1980s and early 1990s two things were happening world-wide that would necessitate changes in the way health care was delivered in Saskatchewan (53). There was an economic recession and a shift in thinking for academics and politicians was occurring as new research on the social determinants of health emerged (53). In order to address both of these phenomena, the government of Saskatchewan decided that they would move towards a 'Wellness' model of care that would emphasize health promotion and disease prevention along with reducing family violence, stress, unemployment, drug abuse and poverty (53). Focusing on prevention of disease instead of just treating disease involved the process of regionalization, whereby the 400 plus boards that represented hospitals, home care, long term care and ambulances were mandated by the provincial government to amalgamate by August 17, 1993 (53). Municipalities were left to their own devices to determine how they wanted to consolidate, which resulted in the formation of 32 health districts and the Athabasca Health Authority (please see Figure 2.5) (53). To further complicate this situation, the provincial government also announced that as of October 1, 1993 they were converting 52 small rural hospitals into either Wellness Centres or long-term care facilities (53).

Citizens were extremely distressed and angry at the prospect of losing their small town hospitals; they felt that the provincial government was abandoning rural residents and they were concerned about how these 'conversions' would impact their health (52, 53). Interestingly, in a study conducted in 2001 that examined mortality rates in communities that had lost their hospital, still had their hospital and had never had a

hospital, researchers found that those who lived in a community that had never had a small hospital had the lowest mortality rates of all the groups (52). Furthermore, they found that the communities in which the hospitals had been closed had lower mortality rates than in communities that kept their hospitals (52). The researchers hypothesized that this surprising trend was because the presence of small hospitals unintentionally created patterns of care and dependencies that resulted in poorer outcomes (52).

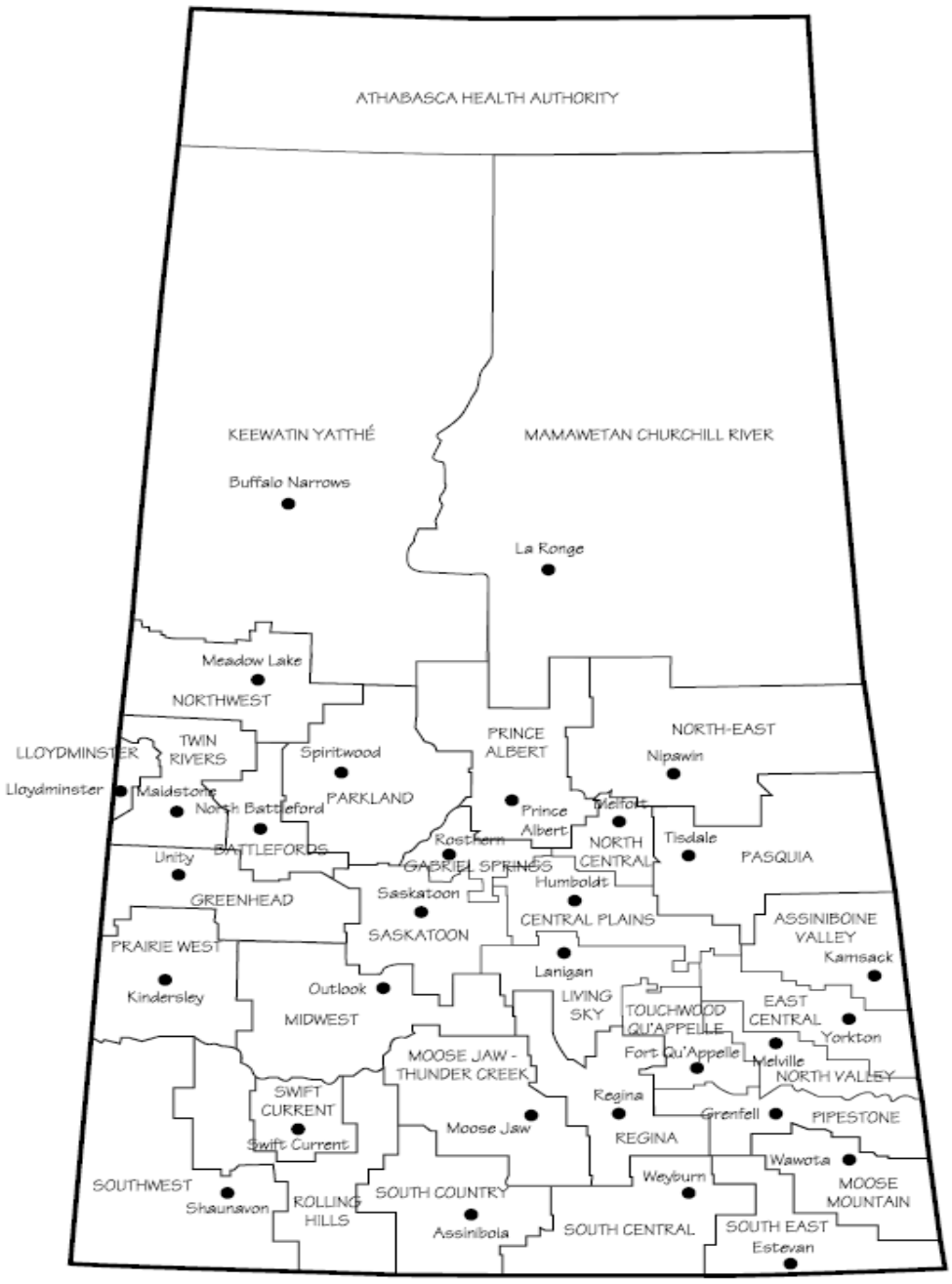


Figure 2.5: Map of Saskatchewan health districts (54)

It wasn't long before the provincial government realized that the health districts were too small to provide the economies of scale that would be required to reduce costs or to offer the types of services that are required for a Wellness Model. Consequently,

in 2002 the health districts were again amalgamated – this time into 13 Regional Health Authorities (RHAs) (please see Table 2.2 and Figure 2.6).

Table 2.2: Amalgamation of health districts into regional health authorities

Health Districts	Regional Health Authorities
Athabasca Health Authority	Athabasca Health Authority
Keewatin Yatthé	Keewatin Yatthé
Mamawetan Churchill River	Mamawetan Churchill River
Northwest, Lloydminster, Twin Rivers, Battlefords	Prairie North
Parkland, Prince Albert	Prince Albert Parkland
North-East, North Central, Pasquia	Kelsey Trail
Greenhead, Prairie West, Midwest	Heartland
Saskatoon, Gabriel Springs, Central Plains, Living Sky	Saskatoon
Assiniboine Valley, East Central, North Valley	Sunrise
Southwest, Swift Current, Rolling Hills	Cypress
South Country, Moose Jaw-Thunder Creek	Five Hills
Regina, Touchwood Qu'Appelle, Pipestone	Regina Qu'Appelle
South Central, South East, Moose Mountain	Sun Country

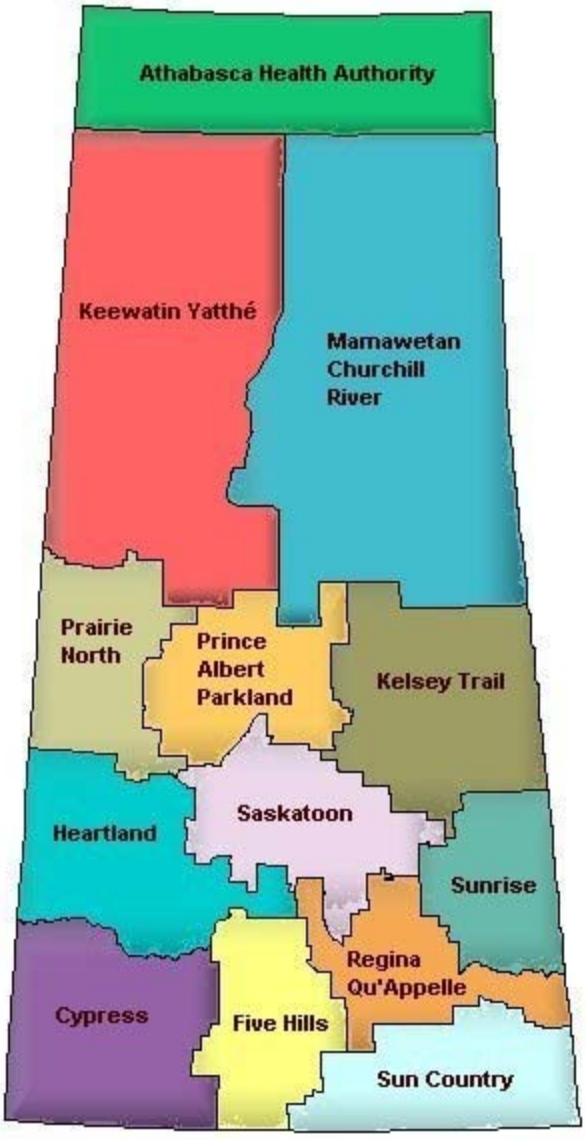


Figure 2.6: Map of Saskatchewan regional health authorities (55)

2.5 Utility, Validity and Reliability of Administrative Databases

Administrative data (data collected for some administrative purpose, not primarily for research or surveillance) and health care claims data (hospital and physician billings) are commonly used to assess the population impacts of health issues (44, 56, 57). By linking the information found in several databases, researchers are able to analyze a single, comprehensive data source that includes information on patient diagnoses, provider services, utilization of resources and socio-demographic characteristics (56).

There are many advantages to using administrative data and/or claims data as opposed to specially-collected data: cost (most of the time, it is considerably less expensive to use pre-existing data than to collect new data), large sample size (traditionally this type of data will cover an entire population), and relative freedom from bias (this type of data was collected blindly without any specific hypotheses in mind) (44, 56).

Use of administrative and/or claims data is not without its limitations. Primarily, this data was not collected with research in mind, consequently many variables that are important to researchers are not included as they were not important for the administrators that originally collected the data (44). Additionally, due to privacy concerns, researchers are often limited in their ability to access this type of data and with what they are able to do with this type of data (44). Furthermore, there may be valid concerns regarding the accuracy of this type of data. A study conducted in the United States found that of 348 physician visits, physicians made an incorrect primary diagnosis 15% of the time, forms where the data was recorded were missing 8% of the time, and data was entered incorrectly 22% of the time (57). It is important to note that of the three sites where this study occurred, only one site used a partial fee-for-service payment mechanism. It is possible that when physicians' income depends on the quality of the data they submit they may have more incentive to accurately record what was done and what the diagnosis was (57). Furthermore, this particular study only allowed the physicians one visit to make an accurate diagnosis (57). It is possible that if a physician saw a patient on a regular basis s/he would have the opportunity to review previous diagnoses, re-assess the patient's condition, and make the correct diagnosis on a subsequent visit (57).

While limitations of this type of data source need to be kept in mind when utilizing this type of data, researchers can take steps to minimize the potential for error in the data they receive by linking databases, examining comorbidities and prior use of health care services to partially account for severity of illness, and use of statistical methods such as imputation to account for missing data (56). Studies have been done specifically examining the reliability of the data contained in the Saskatchewan Health data banks. One such study examined the reliability of the recording of hysterectomies in the hospitalization data files and in the clinical charts (58). This study concluded that the health care utilization files maintained by Saskatchewan Health act as a valid source of data for both research and evaluation studies (58).

CHAPTER THREE: METHODOLOGY

This chapter will describe the materials and methods used in this study. It will begin by describing the study population, the study design, and the study variables before continuing on to the analysis plan.

3.1 Study Population

This study follows 17,414 children (9169 cases and 8245 controls) born between January 1, 1994 to December 31, 1998 from birth until their fifth birthday (1827 days). Children remained in the study until their fifth birthday, death, or emigration out of Saskatchewan, whichever event occurred first. All live born children in Saskatchewan with a congenital anomaly in the aforementioned time period were included in the case group. A CA was defined as the presence of an International Classification of Disease, Version 9 (ICD-9) code between 740 and 759, a Medical Services Branch (MSB) code of 60 or 61, or an International Classification of Disease, Version 10 (ICD-10) code between Q00 and Q99 (please see Appendix A for a list of included codes) that was diagnosed at anytime prior to the child's fifth birthday. Four hundred sixty-nine (469) children eligible for inclusion as controls in the birth cohort had CAs that were diagnosed after their fifth birthday and were not included in the study as either cases or controls. During the follow-up period, use of physician services and hospital services were tracked through routinely collected health administrative data and physician billing

data. The length of stay in hospital was derived by subtracting the date of admission from the date of discharge.

3.2 Study Design

This is a retrospective cohort study that involves the use of individual level variables. In a cohort study, the researcher follows a group of exposed individuals (i.e. those with congenital anomalies) and unexposed individuals (i.e. those without congenital anomalies) and follows both groups for a period of time to compare the incidence of disease (or in this particular case, utilization of health care services) in both groups (59). This particular study is more complicated than a typical cohort study would be because of the number of outcome variables. In addition to examining the differential utilization of health care services between children with and without congenital anomalies, this research is also attempting to examine regional differences in health care utilization for children with and without congenital anomalies.

This study uses data from administrative health databases maintained by Saskatchewan Health. A matching with replacement selection process was used to match controls to cases based on Regional Health Authority of residence at birth, gender, Registered Indian status and year of birth. Matching is the process of equalizing the distribution of selected factors within study populations (i.e. the case population and the control population), and is used frequently to control for selection bias in both cohort and case-control studies (60). In cohort studies, matching has two main functions: first it can be used to control confounding, and second it can be used to increase the efficiency of the study (60). While there can be many drawbacks associated with matching in case-control studies, there are fewer drawbacks in cohort studies because

the matching process is independent of the outcome under study (i.e. use of health care services) (60). In this particular study, one-to-one matching with replacement was used, meaning that for each case, one matched control was selected from all live births between January 1, 1994 to December 31, 1998. Matching with replacement means that once a control was selected for a particular case, it was put back into the pool of eligible controls and therefore was available to be randomly selected as a control for another case. Because of this matching process, 9169 controls are used in the analysis instead of the 8245 individual controls as some controls were matched to more than one case.

3.3 Data Sources

Data was obtained from the following Saskatchewan Health databases: Health Insurance Registration File (a.k.a. Population Registry, Person Registry System), Vital Statistics, Hospital Separations, and Physician Claims.

Through the process of administering a publicly funded health care system Saskatchewan Health has accumulated a large amount of administrative health care data that has been used for over one hundred studies (61). The Population Registry contains information on all individuals who are eligible for Saskatchewan Health benefits (the covered population) and is updated daily for name and address changes, births, deaths, receipt of social assistance, new residents and departing residents from the province (61).

Vital statistics data includes information on all births, deaths, stillbirths and marriages for Saskatchewan Health beneficiaries (61). Live birth registration is the responsibility of the family and includes both obstetrical and infant information (61).

As all Saskatchewan Health beneficiaries are eligible to receive medically necessary hospital services without charge, Saskatchewan Health is able to collect hospital services data from all hospitals in Saskatchewan; this includes all acute care in-patient hospital separations, in-patient psychiatric separations and day surgeries (61). Information on out-of-province hospital separations is also captured for individuals with Saskatchewan HSNs; however, the level of detail may not be the same as for in-province hospital separations (61).

Since 1979, medical services diagnoses have been reported according to the International Classification of Diseases, Ninth Revision (ICD-9) coding scheme and procedures were coded according to the Canadian Classification of Diagnostic, Therapeutic and Surgical Procedures (CCP) guidelines (61). As of April 2002, diagnoses are coded according to the International Statistical Classification of Diseases and Related Health Problems, Tenth Revision, Canada (ICD-10-CA) guidelines, and procedures are recorded using the Canadian Classification of Health Interventions (CCI) coding scheme (61).

In addition to hospital benefits, members of the covered population are also eligible to receive benefits for insured medical services without charge (61). These benefits include: anesthesia, diagnostic services, obstetrical services and surgical services; however, there are some medical services (e.g. cosmetic surgery, examinations for insurance and/or employment purposes) that are not insured (61). Medical services data is based on physicians' claims for payment under a fee-for-service payment plan (61). While there are a number of physicians in Saskatchewan who fall under an alternative payment plan (i.e. salary, contract), unless they choose to shadow or dummy

bill Saskatchewan Health for the services they provide, this information will not be captured in this database (61). Additionally, medical services from non-physicians (i.e. nurse practitioners) will not be captured in this database.

Data was abstracted from the abovementioned databases by Saskatchewan Health personnel, linked by health services number and de-identified prior to its release.

3.4 Study Variables

Table 3.1 describes the variables used in this particular study. Cut points for categorical variables were selected based on their clinical significance, definitions found in the literature and data availability. Predictor variables are grouped by characteristics related to need, predisposing factors and enabling factors as per Anderson's model for the determinants of health care utilization (11).

Table 3.1: Description of study variables

Predictor Variables	Description	Variable Coding
Need Characteristics		
Congenital Anomaly Status	This is a dichotomous variable, with children who do not have any congenital anomalies being coded as '0' and children who have at least one congenital anomaly being coded as '1'.	No (ref) Yes
Multiple Congenital Anomaly Status	This is a derived dichotomous variable, with children who have at least two congenital anomalies being coded as '1' and all other children being coded as '0'.	No (ref) Yes
Type of Congenital Anomaly	This is a series of 22 dichotomous variables (ICD-9 740-759, MSB 60-61) which indicate the specific congenital anomaly the child has. Children who do not have that particular congenital anomaly are coded as '0', and children who do have that particular congenital anomaly are coded as '1'.	No (ref) Yes

Birth Weight	This is a categorical variable with 6 values: '1' for a birth weight of less than 1000g, '2' for a birth weight of 1000-1499g, '3' for a birth weight of 1500-1999g, '4' for a birth weight of 2000-2499g, '5' for a birth weight of 2500-3999g, and '6' for a birth weight of 4000g or higher.	<1000g (ref) 1000g-1499g 1500g-1999g 2000g-2499g 2500g-3999g ≥4000g
Gestational Age	This is a categorical variable with 4 levels: '1' for a gestational age of less than 28 weeks (very pre-term), '2' for a gestational age of 28-36 weeks (pre-term), '3' for a gestational age of 37-41 weeks (term), and '4' for a gestational age of 42 weeks or more (post-term).	< 28 weeks (ref) 28-36 weeks 37-41 weeks ≥42 weeks
Predisposing Characteristics		
Baby's Sex	This is a dichotomous variable, with males coded as '1' and females coded as '2'.	Male (ref) Female
Registered Indian Status	This is a dichotomous variable, with Registered Indians being coded as '1', and the general population being coded as '0'. Please note that this is not an accurate reflection of Aboriginal status as many individuals who would self-identify as Aboriginal (i.e. Métis) do not qualify for Registered Indian status (32).	No (ref) Yes
Mother's Age Group	This is a categorical variable with 3 levels that indicates the mother's age group at the time of the child's birth. Mothers who were less than 20 years old at the time of the baby's birth were coded as '1', mothers who were between the ages of 20 and 34 when their babies were born were coded as '2', and mothers who were 35 years of age or older when their babies were born were coded as '3'.	< 20 (ref) 20-34 ≥35
Mother's Marital Status	This is a categorical variable with 3 levels that indicates the mother's marital status at the time of the child's birth. Mothers who were single were coded as '1', mothers who were married or in a common law relationship were coded as '2'. All mothers for whom this information was not available were coded as '3' for other.	Single (ref) Married/Common Law Other

Parity	This is a categorical variable with 5 levels that measures the combined number of live births and stillbirths (please note that the birth of this child is included in this number, i.e. if this is a first child, the parity would equal 1). First time mothers were coded as '1', mothers who had given birth twice were coded as '2', mothers who had given birth three times were coded as '3', mothers who had given birth four times were coded as '4', and mothers who had given birth five or more times were coded as 5. Please note that this variable is not able to distinguish between live births and stillbirths.	1 (ref) 2 3 4 ≥5
Number of Times Mother is Enrolled in the Study	This is a continuous variable ranging from 1-4 that measures the number of times the mother is enrolled in the study.	1 (ref) 2 3 4
Enabling Characteristics		
RHA of Residence	This is a series of 5 categorical variables that indicate in which RHA the child lived as of December 31st of each year the child was enrolled in the study. Individuals living in Saskatoon were coded as '1', individuals living in Five Hills were coded as '2', individuals living in Cypress were coded as '3', individuals living in Regina Qu'Appelle were coded as '4', individuals living in Sunrise were coded as '5', individuals living in Heartland were coded as '7', individuals living in Kelsey Trail were coded as '8', individuals living in Prince Albert Parkland were coded as '9', individuals living in Prairie North were coded as '10', individuals living in Sun Country were coded as '11' and individuals living in Northern Saskatchewan (Mamawetan Churchill River, Keewatin Yatthé, and Athabasca) were coded as '99'.	Saskatoon (ref) Sun Country Five Hills Cypress Regina Qu'Appelle Sunrise Heartland Kelsey Trail PA Parkland Prairie North Northern Saskatchewan

Family Received Income Assistance	This is a series of 6 dichotomous variables that indicate whether or not the child's family was receiving some form of government income assistance as of December 31st. This variable is measured separately for each of the five years of follow-up and a derived variable was created to measure whether or not a family had ever received government income assistance. Families that had received income assistance were coded as '1' and families that had not received income assistance were coded as '0'. Please note that this variable does not capture families that had received income assistance during the year, but were not currently receiving income assistance as of December 31st.	No (ref) Yes
Travel for Physician Visits	This is a series of 8 dichotomous derived variables that measure whether or not a child traveled outside of his/her home RHA for a physician visit in another RHA during the first 7 days, the first 28 days, the first year, the second year, the third year, the fourth year, the fifth year, or at any point in time during the study period. Children who did not travel outside of their home RHA for a physician visit were coded as '0', and children who did travel outside of their home RHA for a physician visit were coded as '1'.	No (ref) Yes
Travel for Hospitalization	This is a series of 8 dichotomous derived variables that measure whether or not a child was admitted to a hospital outside of his/her home RHA during the first 7 days, the first 28 days, the first year, the second year, the third year, the fourth year, the fifth year, or at any point in time during the study period. Children who were not hospitalized outside of their home RHA were coded as '0', and children who were hospitalized outside of their home RHA were coded as '1'.	No (ref) Yes

Child Moved to Another RHA	This is a derived dichotomous variable that indicates whether or not a child moved to at least one other RHA during their enrollment in the study (i.e. as of December 31st they were living in a different RHA than at December 31st of the previous year). Children who had not moved were coded as '0' and children that had moved at least once were coded as '1'. Please note, this variable does not account for multiple moves to other RHAs within the course of one calendar year, or moves within RHAs.	No (ref) Yes
Follow-Up Time Period	This is a continuous variable, ranging from 0 days to 1827 days, that measures the number of days the child was enrolled in the study.	Continuous Variable
Reason for Study Exit	This is a dichotomous variable with a value of '0' if the child remained in the study until the study was finished and a value of '1' if the child exited the study due to any form of health coverage termination (i.e. emigrated out of province or death).	Study End (ref) Health Coverage Terminated
Outcome Variables	**Description**	
Hospital Admissions	This is a series of 8 count variables, ranging from 0-67, that indicates the number of hospital admissions in the first 7 days, the first 28 days, the first year, the second year, the third year, the fourth year, the fifth year, or at any point in time during the study period.	Count Variable
Physician Visits	This is a series of 8 count variables, ranging from 0-713, that indicates the number of physician visits in the first 7 days, the first 28 days, the first year, the second year, the third year, the fourth year, the fifth year, or at any point in time during the study period.	Count Variable
Length of Stay	This is a count variable that ranges from 0-779, that indicates the total length of stay in hospital during the study period.	Count Variable

3.5 Ethics Approval and Confidentiality

This study was approved by the University of Saskatchewan Behavioural Ethics Review Committee (BEH #05-159, 2005). De-identified individual record level data and aggregate data were forwarded to the researcher from Saskatchewan Health following approval from Saskatchewan Health's Data Access Review Committee (DARC). All results are be reported at the RHA or provincial level, and a minimum cell size of five (5) was used for all analyses.

3.6 Software Used

A variety of software packages were used during the completion of this study:

- Microsoft Excel (2000) and Microsoft Access (2000) were used for data cleaning
- SPSS (version 13.0) and STATA (version 9.0) were used for data analysis
    - STATA was used for model building, while SPSS was used for all other analyses
- Excel and SPSS were used to create tables and figures
- EndNote (version 8.0.2) was used for reference management

3.7 Data Analysis

3.7.1 Characteristics of the Study Population

Frequencies are presented for all predictor variables seen in Table 3.1. Bivariate analyses were used to compare the distribution of predictor variable for children with and without congenital anomalies. One way ANOVAs were used to compare continuous variables and chi squares were used to compare categorical variables

3.7.2 Regional Differences in the Prevalence of Congenital Anomalies

Prevalence of CAs is defined as the proportion of children with a birth defect in Saskatchewan that were born between January 1, 1994 to December 31, 1998 (62). As

many birth defects lead to early embryonic death, which, if even recognized, is often classified as a miscarriage instead of a birth (either live or still), the number of children with CAs at birth is only a small proportion of all affected embryos (62). Hence, when studying CAs, prevalence is used as the standard measure of disease frequency instead of incidence (62).

When calculating the prevalence of CAs, children with multiple CAs were counted as one, but when calculating the prevalence of specific CAs, children with multiple CAs were counted for each type of CA they are affected by (63). Graphical analysis was used to assess regional differences in the overall prevalence of CAs and regional differences in specific types of CAs. The Saskatoon Health Region was selected as the reference category for all regional comparisons for a number of reasons. Foremost, is that the Saskatoon Health Region has the most tertiary care centres in Saskatchewan. Additionally, it has the largest population of all of the regional health authorities, and due to more resources this population is the most studied. Finally, the Saskatoon Health Region is used as a reference category in many Saskatchewan Health reports.

3.7.3 Study Question 1: Is the Level of Health Care Used by Children With Congenital Anomalies Significantly Different from the Level of Health Care Used by Children Without Congenital Anomalies?

One way ANOVAs were used to compare each of the three outcome variables for children with and without CAs, and for children with specific types of CAs versus children without CAs.

3.7.4 Study Question 2: Is There a Regional Difference in the Level of Health Care Used by Children in their First Five Years of Life? And Does This Relationship Hold For Children With and Without CAs?

One way ANOVAs and Bonferroni post-hoc tests were used to determine whether or not there is a crude regional difference in each of the three outcome variables for all children in the study, for children with congenital anomalies and for children without congenital anomalies. Additionally, as travel outside of one's home RHA for health care is likely to be a significant confounder of use of health care services, crude regional differences in travel for physician visits and for hospitalizations were also assessed using one way ANOVAs and Bonferroni post-hoc tests. For all regional comparisons, the Saskatoon Health Region was used as the reference category.

3.7.5 Study Question 3: What Factors Influence the Level of Health Care Utilization?

Health care utilization is a broad term, and in this particular study encompasses three distinct outcome variables: total number of physician visits in the study period, total number of hospital admissions in the study period and total length of stay (in days) in the study period (see Table 3.1 for a description of these variables). Since the factors that influence the use of these services may differ, each outcome variable was assessed separately. As the outcome variables were based on count data with substantial over-dispersion, a negative binomial distribution was used to model each of the outcome variables (60, 64, 65).

All variables were initially included in the model, and variables that were statistically significant ($p<0.05$) were retained as main effects. As can be seen in Table 3.1, all variables used in the model were categorical in nature and the first category was

left as the default reference category. If any one of the categories achieved statistical significance the entire variable was retained in the model.

Once main effects were established for each of the three models, the interaction terms seen in Table 3.2 were tested for statistical significance provided that all the constituent parts of the interaction terms had already achieved statistical significance. Interaction terms were added simultaneously and were retained in the final model if they were statistically significant at the alpha is less than 0.05 level. Interaction terms were also treated as categorical variables. Interaction terms were selected as the literature shows that these variables have the biggest known impact on health care utilization.

Table 3.2: Interaction terms tested in all predictive models for health care utilization

Interaction Term	Description
RHA*TRAVEL	*RHA of Residence in Birth Year x Child Ever Traveled Outside of Home RHA for Health Care†*
RI*CA	*Congenital Anomaly x Registered Indian Status*
AGE*CA	*Congenital Anomaly x Mother's Age Group*
SEX*CA	*Congenital Anomaly x Baby's Sex*
MARITAL*CA	*Congenital Anomaly x Mother's Marital Status*
MARITAL*AGE	*Mother's Marital Status x Mother's Age Group*

† For physician visits model only travel for physician visits was included and for hospitalization model and LOS model only travel for hospitalization was included

After all main effects and interaction terms had been tested for statistical significance, a final model was run that only included terms that were statistically significant.

The odds ratios presented for RHA in the models for health care utilization are adjusted for all other factors present in the model.

CHAPTER FOUR: RESULTS

This chapter begins by describing the characteristics of the total study population, and then separately describes the population of children with congenital anomalies and children without congenital anomalies. It goes on to describe the regional differences in birth defects in Saskatchewan prior to presenting the results of the study questions.

4.1 Description of the Study Population

As the covariates in this study fall into two general categories – those that do not vary over time and those that do vary over time – the results of these types of variables are presented separately in Tables 4.1 and 4.2 respectively and in Figures 4.1 through 4.3. Table 4.1 presents the descriptive results for all of the covariates that do not vary over time for the study population. For categorical variables, the frequency and the percentage are presented for each of the categories, while the mean, median, mode and range are presented for the continuous variables.

Table 4.1: Non-time-varying covariates for the study population (N=18338)

Predictor Variables	Category	Frequency (%)
Need Characteristics		
Congenital Anomaly	Yes	9169 (50.0%)
	No	9169 (50.0%)
Multiple Congenital Anomalies	Yes	2082 (11.4%)
	No	16256 (88.6%)
Birth Weight	<1000g	160 (0.9%)
	1000g-1499g	198 (1.1%)
	1500g-1999g	315 (1.7%)
	2000g-2499g	752 (4.1%)
	2500g-3999g	14425 (78.7%)
	≥4000g	2488 (13.6%)

Gestational Age	< 28 weeks	165 (0.9%)
	28-36 weeks	1492 (8.1%)
	37-41 weeks	16181 (88.2%)
	≥42 weeks	500 (2.7%)
Predisposing Characteristics		
Baby's Sex	Male	10026 (54.7%)
	Female	8312 (45.3%)
Registered Indian Status	Yes	2980 (16.3%)
	No	15358 (83.7%)
Mother's Age Group	< 20	1915 (10.4%)
	20-34	14669 (80.0%)
	≥35	1754 (9.6%)
Mother's Marital Status	Single	4922 (26.8%)
	Married/Common Law	12238 (66.7%)
	Other	1177 (6.4%)
Parity	1	6377 (34.8%)
	2	6162 (33.6%)
	3	3346 (18.2%)
	4	1383 (7.5%)
	≥5	1070 (5.8%)
Number of Times Mother is Enrolled in the Study	1	15496 (84.5%)
	2	2695 (14.7%)
	3	143 (0.8%)
	4	4 (0.0%)
Enabling Characteristics		
Child Moved to Another RHA	Yes	1202 (6.6%)
	No	17136 (93.4%)
Follow-Up Time Period (Days)	Mean	1707.12
	Median	1826.00
	Mode	1826
	Range	0-1827
Reason for Study Exit	Study End	17157 (93.6%)
	Health Coverage Terminated	1181 (6.4%)

Table 4.1 indicates that there are more males than females in the study population (54.7% vs. 45.3%), and that there are substantially fewer Registered Indians than members of the general population (16.3% vs. 83.7%) in the sample. The percentage of Registered Indians in the current study is reflective of the percentage of Registered Indians in the total Saskatchewan population. The majority of infants were born at term (37-41 weeks gestational age) (88.2%) and were of a normal birth weight (2500-3999g)

for term infants (78.7%). However, 2.7% of the children enrolled in the study were born post-term (gestational age ≥42 weeks), while 8.1% were pre-term (gestational age between 28 and 36 weeks) and 0.9% were born very pre-term (< 28 weeks gestational age). As mentioned previously, most of the infants had a normal birth weight for term babies; however, 13.6% had a high birth weight (≥4000g for term infants), and 3.7% had a very low birth weight (<2000g for term infants). The remaining 4.1% of infants had a low birth weight (2000g-2499g for term infants. Additionally, over 90% of the children were enrolled in the study until the study was finished, with only 6.4% of children exiting the study prematurely, either due to death or emigration. The mean number of days of follow-up was 1707.12 days.

As depicted in Table 4.1, 80.0% of mothers were between the ages of 20 and 34 at the time of their child's birth, while 10.4% were teen mothers, and 9.6% were 35 years of age or older. At the time of their child's birth, 66.7% of mothers were married or in a common-law relationship, 26.8% were single, and 6.4% were defined by Saskatchewan Health as 'other'. Eighty-four point five percent (84.5%) of the mothers were enrolled in the study once, while 14.7% were included in the study twice as they had two children included in the study; the remaining 0.8% were included in the study three times. In this study, the term parity refers to the combined total of all live births and stillbirths; however, due to the manner in which data was released for the study it is not possible to differentiate between the two. In this study, 34.8% of mothers were first-time mothers, while 33.6% had had one previous birth prior to the child that was included in the study. An additional 18.2% of mothers had a parity of three, 7.5% had a parity of four and the remaining 5.8% had a parity of five or more.

Table 4.2 and Figures 4.1 to 4.3 describe the covariates that do vary over time for the study population. As the majority of children were enrolled in the study for the entire duration of the study, and only 6.6% of children moved to another RHA during the study period (see table 4.1) it is not surprising that there is little variability in RHA of residence for the duration of the study. The largest change is in the Regina Qu'Appelle health region which saw a decrease of 3.0% over the entire five year time period. Slightly more variability over time is seen with regards to whether or not children's families are receiving some form of governmental income assistance. This changes from a low of 16.8% of all children in the first year of follow-up to a high of 22.5% in the fifth year of follow-up (resulting in a net increase of 5.7%). Each year there was a decrease in the number of children who traveled outside of their home RHA to obtain physician care or for hospitalizations.

Table 4.2: RHA of residence for the study population by year (N=18338)

		Frequency (%)					
Predictor Variables	**Category**	**1st Year**	**2nd Year**	**3rd Year**	**4th Year**	**5th Year**	**Ever**
Enabling Characteristics							
RHA of Residence	Sun Country	892 (4.9%)	859 (4.7%)	853 (4.7%)	846 (4.6%)	840 (4.6%)	N/A
	Five Hills	953 (5.2%)	921 (5.0%)	897 (4.9%)	878 (4.8%)	850 (4.6%)	N/A
	Cypress	468 (2.6%)	455 (2.5%)	444 (2.4%)	444 (2.4%)	430 (2.3%)	N/A
	Regina Qu'Appelle	5244 (28.6%)	5057 (27.6%)	4926 (26.9%)	4799 (26.2%)	4700 (25.6%)	N/A
	Sunrise	946 (5.2%)	902 (4.9%)	881 (4.8%)	866 (4.7%)	865 (4.7%)	N/A
	Saskatoon	5039 (27.5%)	4881 (26.6%)	4791 (26.1%)	4642 (25.3%)	4568 (24.9%)	N/A
	Heartland	669 (3.6%)	647 (3.5%)	655 (3.6%)	651 (3.6%)	639 (3.5%)	N/A
	Kelsey Trail	621 (3.4%)	618 (3.4%)	599 (3.3%)	606 (3.3%)	616 (3.4%)	N/A

	PA Parkland	1369 (7.5%)	1331 (7.3%)	1308 (7.1%)	1294 (7.1%)	1265 (6.9%)	N/A
	Prairie North	1310 (7.1%)	1241 (6.8%)	1185 (6.5%)	1139 (6.2%)	1112 (6.1%)	N/A
	Northern Sask	827 (4.5%)	785 (4.3%)	759 (4.1%)	745 (4.1%)	732 (4.0%)	N/A
	Missing	0 (0.0%)	641 (3.5%)	1040 (5.7%)	1428 (7.8%)	1721 (9.4%)	N/A

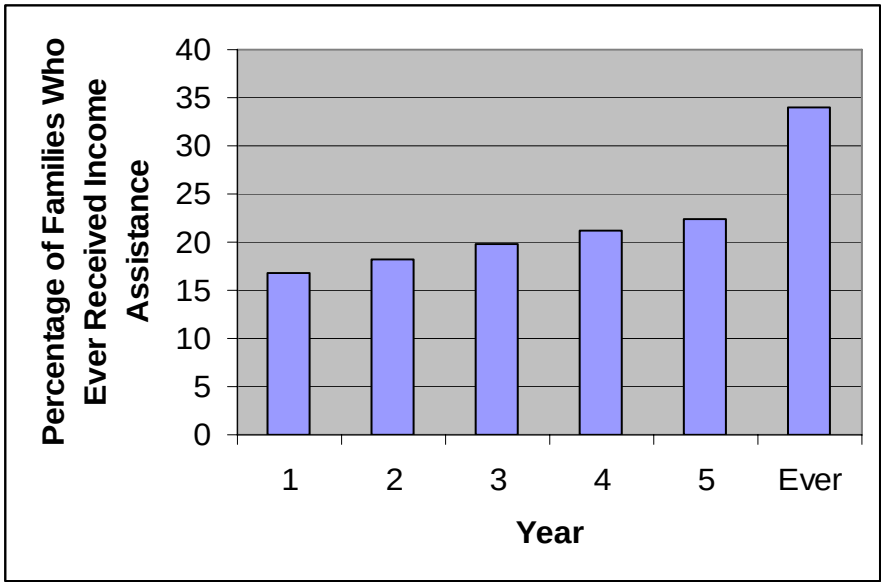


Figure 4.1: Percentage of families receiving income assistance by year in the study population

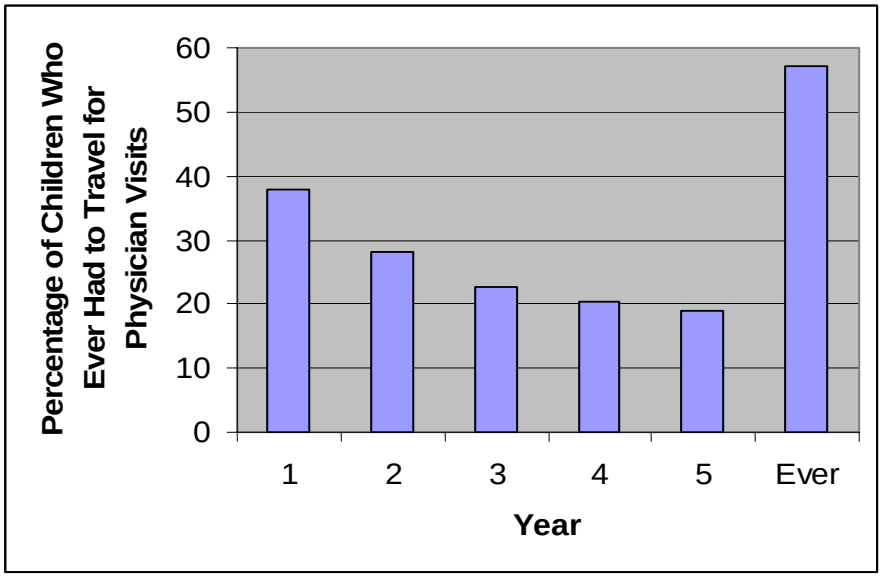


Figure 4.2: Percentage of children who traveled outside of their home RHA for a physician visit by year

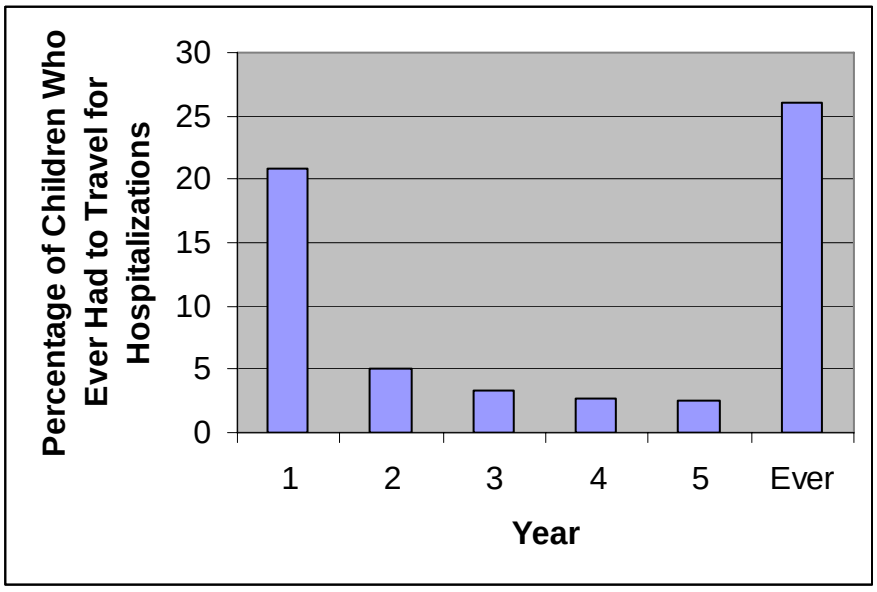


Figure 4.3: Percentage of children who traveled outside of their home RHA for a hospital admission by year

The prevalence per 1000 live births of different types of congenital anomalies in the study population can be seen in Figure 4.4 (see Appendix A for more detail regarding what specific CAs make up each larger category of CAs). Figure 4.5 illustrates the prevalence per 1000 live births of the broader categories of CAs seen in the study population.

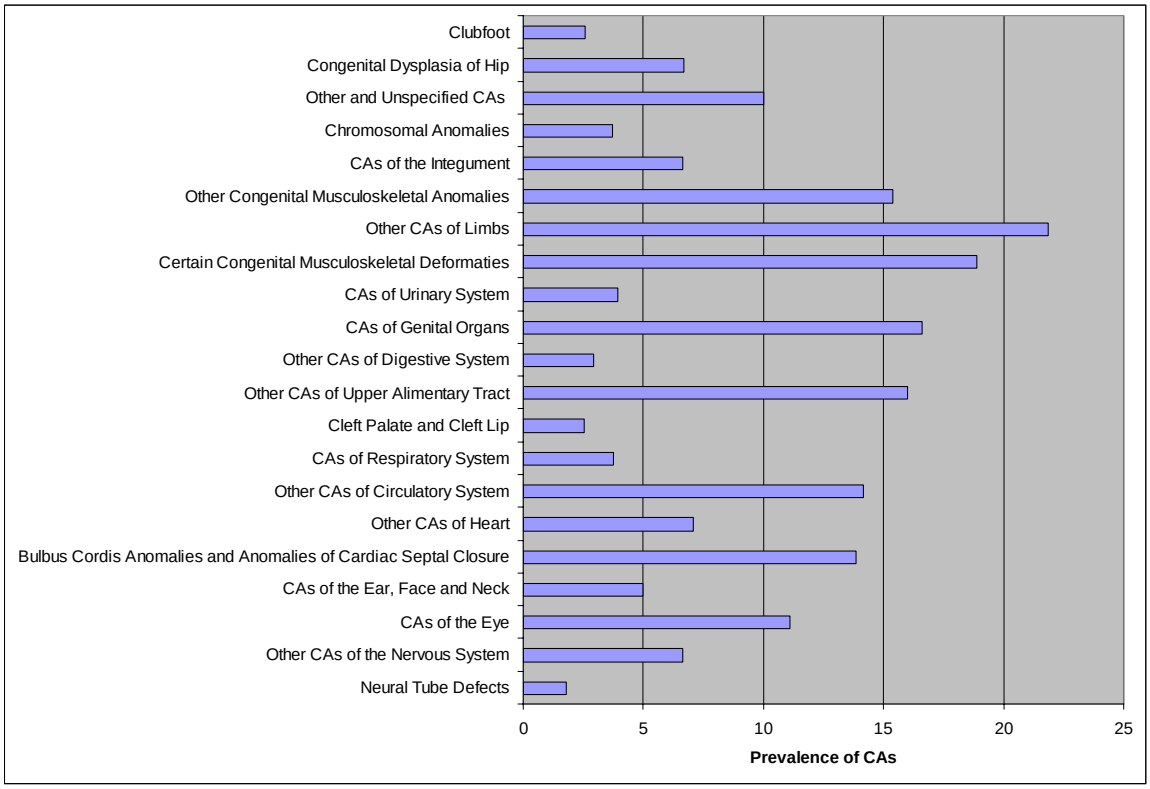


Figure 4.4: Prevalence of congenital anomalies in the study population per 1000 live births

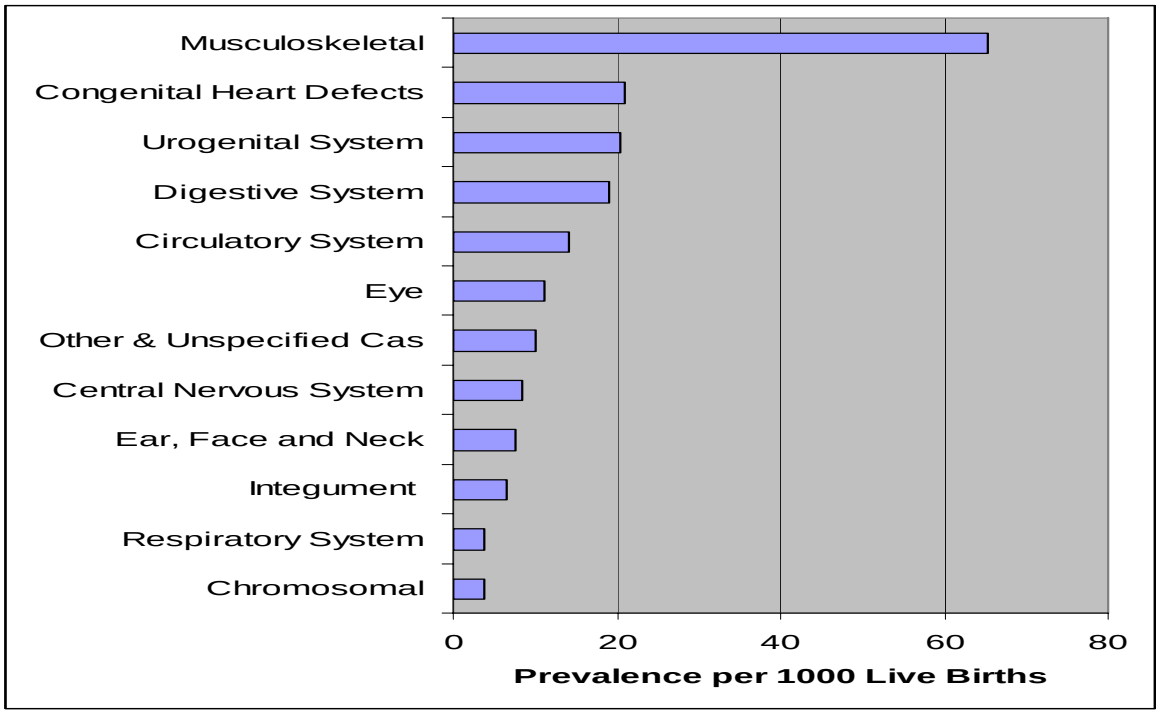


Figure 4.5: Prevalence of categories of CAs in the study population

4.2 Bivariate Analysis by Congenital Anomaly Status

As congenital anomaly status (the presence or absence of at least one congenital anomaly) is the principal covariate in this study, bivariate analyses were conducted to examine the differences in baseline characteristics between children with birth defects and children without birth defects. Please note that children with and without birth defects were matched on year of birth, sex, RHA of residence in their first year and Registered Indian status, so no variability will be present for these categories.

Table 4.3 illustrates the differences and similarities between children with and without birth defects for predictor variables that do not vary over time. Pearson χ^2 tests were used to assess differences between groups for categorical variables and one-way ANOVAs were used to assess differences between groups for continuous variables.

Table 4.3: Non-time-varying predictor variables for children with congenital anomalies (N=9169) and children without congenital anomalies (N=9169)

Predictor Variables	Category	Children with Congenital Anomalies Frequency (%)	Children without Congenital Anomalies Frequency (%)	Statistical Test (significance level)
Need Characteristics				
Birth Weight	<1000g	157 (1.7%)	3 (0.0%)	$\chi^2 = 503.916$
	1000g-1499g	177 (1.9%)	21 (0.2%)	**(<0.001)***
	1500g-1999g	248 (2.7%)	67 (0.7%)	
	2000g-2499g	509 (5.6%)	243 (2.7%)	
	2500g-3999g	6869 (74.9%)	7556 (82.4%)	
	≥4000g	1209 (13.2%)	1279 (13.9%)	
Gestational Age	< 28 weeks	160 (1.7%)	5 (0.1%)	$\chi^2 = 376.529$
	28-36 weeks	1020 (11.1%)	472 (5.1%)	**(<0.001)***
	37-41 weeks	7748 (84.5%)	8433 (92.0%)	
	≥42 weeks	241 (2.6%)	259 (2.8%)	
Predisposing Characteristics				
Baby's Sex	Male	5013 (54.7%)	5013 (54.7%)	Matched
	Female	4156 (45.3%)	4156 (45.3%)	Variable
Registered Indian Status	Yes	1490 (16.3%)	1490 (16.3%)	Matched
	No	7679 (83.7%)	7679 (83.7%)	Variable

Mother's Age Group	< 20 20-34 ≥35	958 (10.4%) 7298 (79.6%) 913 (10.0%)	957 (10.4%) 7371 (80.4%) 841 (9.2%)	$\chi^2 = 3.319$ (0.190)
Mother's Marital Status	Single Married/Common Law Other	2525 (27.5%) 6020 (65.7%) 623 (6.8%)	2397 (26.1%) 6218 (67.8%) 554 (6.0%)	$\chi^2 = 10.577$ **(0.005)***
Parity	1 2 3 4 ≥5	3385 (36.9%) 3050 (33.3%) 1523 (16.6%) 679 (7.4%) 532 (5.8%)	2992 (32.6%) 3112 (33.9%) 1823 (19.9%) 704 (7.7%) 538 (5.9%)	$\chi^2 = 52.227$ **(<0.001)***
Number of Times Mother is Enrolled in the Study	1 2 3 4	7657 (83.5%) 1426 (15.6%) 82 (0.9%) 4 (0.0%)	7839 (85.5%) 1269 (13.8%) 61 (0.7%) 0 (0.0%)	$\chi^2 = 18.368$ **(<0.001)***
Enabling Characteristics				
Child Moved to Another RHA	Yes No	633 (6.9%) 8536 (93.1%)	569 (6.2%) 8600 (93.8%)	$\chi^2 = 3.647$ (0.056)
Follow-Up Time Period (Days)	Mean Median Mode Range	1697.93 1826.00 1826 0-1827	1716.31 1826.00 1826 0-1827	F = 10.371 **(0.001)***
Reason for Study Exit	Study End Health Coverage Terminated	8060 (87.9%) 1109 (12.1%)	9097 (99.2%) 72 (0.8%)	$\chi^2 = 973.236$ **(<0.001)***

*** Statistically Significant**
† Matched Variables

For the majority of predictor variables there is a statistically significant difference between children with and without congenital anomalies, many of which can be explained by the presence or absence of the anomaly. The early detection of certain CAs may result in the attending physician inducing labour which can in turn result in a lower birth weight and gestational age. Also, significantly more children with birth defects left the study earlier than children without birth defects. While the specific reason why each individual child left the study is unknown, the difference could at least

in part be explained by the higher risk of early death for children with serious birth defects versus children without a birth defect, or the need to emigrate out of province to a larger centre where more specialized care can be offered. Finally, as children with at least one birth defect were more likely to leave the study prematurely than children without any birth defects, it is not surprising that children with birth defects have a shorter mean length of follow-up than children without birth defects. While there is also a statistically significant difference between children with and without birth defects for maternal characteristics, these differences cannot be easily explained due to the presence or absence of an anomaly.

Table 4.4 shows the differences and similarities between children with and without birth defects for time-varying predictor variables. Pearson χ^2 tests were used to assess differences between groups for categorical variables.

Table 4.4: Comparison of time-varying predictor variables for children with congenital anomalies (N=9169) and children without congenital anomalies (N=9169)

			Frequency (%)					
Predictor Variables	**CA Status**	**Category**	**1st Year**	**2nd Year**	**3rd Year**	**4th Year**	**5th Year**	**Ever**
Enabling Characteristics								
RHA of Residence	Children With CAs	Sun Country	446 (4.9%)	432 (4.7%)	432 (4.7%)	425 (4.6%)	423 (4.6%)	N/A
		Five Hills	476 (5.2%)	464 (5.1%)	443 (4.8%)	429 (4.7%)	421 (4.6%)	N/A
		Cypress	234 (2.6%)	224 (2.4%)	218 (2.4%)	216 (2.4%)	207 (2.3%)	N/A
		Regina Qu'Appelle	2622 (28.6%)	2513 (27.4%)	2449 (26.7%)	2381 (26.0%)	2316 (25.3%)	N/A
		Sunrise	472 (5.1%)	448 (4.9%)	432 (4.7%)	427 (4.7%)	426 (4.6%)	N/A
		Saskatoon	2521 (27.5%)	2444 (26.7%)	2395 (26.1%)	2320 (25.3%)	2283 (24.9%)	N/A
		Heartland	335 (3.7%)	325 (3.5 %)	326 (3.6%)	326 (3.6%)	323 (3.5%)	N/A
		Kelsey Trail	310 (3.4%)	311 (3.4%)	299 (3.3%)	301 (3.3%)	308 (3.4%)	N/A
		PA Parkland	685 (7.5%)	658 (7.2%)	649 (7.1%)	638 (7.0%)	618 (6.7%)	N/A

		Prairie North	655 (7.1%)	612 (6.7%)	594 (6.5%)	562 (6.1%)	557 (6.1%)	N/A
		Northern Sask	413 (4.5%)	395 (4.3%)	373 (4.1%)	374 (4.1%)	360 (3.9%)	N/A
		Missing	0 (0.0%)	343 (3.7%)	559 (6.1%)	770 (8.4%)	927 (10.1%)	N/A
	Children Without CAs	Sun Country	446 (4.9%)	427 (4.7%)	421 (4.6%)	421 (4.6%)	417 (4.5%)	N/A
		Five Hills	477 (5.2%)	457 (5.0%)	454 (5.0%)	449 (4.9%)	429 (4.7%)	N/A
		Cypress	234 (2.6%)	231 (2.5%)	226 (2.5%)	228 (2.5%)	223 (2.4%)	N/A
		Regina Qu'Appelle	2622 (28.6%)	2544 (27.7%)	2477 (27.0%)	2418 (26.4%)	2384 (26.0%)	N/A
		Sunrise	474 (5.2%)	454 (5.0%)	449 (4.9%)	439 (4.8%)	439 (4.8%)	N/A
		Saskatoon	2518 (27.5%)	2437 (26.6%)	2396 (26.1%)	2322 (25.3%)	2285 (24.9%)	N/A
		Heartland	334 (3.6%)	322 (3.5 %)	329 (3.6%)	325 (3.5%)	316 (3.4%)	N/A
		Kelsey Trail	311 (3.4%)	307 (3.3%)	300 (3.3%)	305 (3.3%)	308 (3.4%)	N/A
		PA Parkland	684 (7.5%)	673 (7.3%)	659 (7.2%)	656 (7.2%)	647 (7.1%)	N/A
		Prairie North	655 (7.1%)	629 (6.9%)	591 (6.4%)	577 (6.3%)	555 (6.1%)	N/A
		Northern Sask	414 (4.5%)	390 (4.3%)	386 (4.2%)	371 (4.0%)	372 (4.1%)	N/A
		Missing	0 (0.0%)	298 (3.3%)	481 (5.2%)	658 (7.2%)	794 (8.7%)	N/A
	χ^2 (sig. level)		Matched Variable	0.789 (1.000)	0.879 (1.000)	0.997 (1.000)	1.771 (0.998)	
Family Received Income Assistance	Children With CAs	Yes	1614 (17.6%)	1788 (19.5%)	1931 (21.1%)	2002 (21.8%)	2130 (23.2%)	3237 (35.3 %)
		No	7555 (82.4%)	7038 (76.8%)	6679 (72.8%)	6397 (69.8%)	6112 (66.7%)	5932 (64.7 %)
		Missing	0 (0.0%)	343 (3.7%)	559 (6.1%)	770 (8.4%)	927 (10.1%)	0 (0.0 %)
	Children Without CAs	Yes	1464 (16.0%)	1572 (17.1%)	1719 (18.7%)	1881 (20.5%)	1993 (21.7%)	3016 (32.9 %)
		No	7705 (84.0%)	7299 (79.6%)	6969 (76.0%)	6630 (72.3%)	6382 (69.6%)	6153 (67.1 %)
		Missing	0 (0.0%)	298 (3.3%)	481 (5.2%)	658 (7.2%)	794 (8.7%)	0 (0.0 %)

	χ^2 (sig. level)		8.784 **(0.003)** *	18.523 **(<0.001)** *	18.124 **(<0.001)** *	7.196 **(0.007)** *	9.323 **(0.002)** *	11.852 **(0.001)** *
Travel for Physician Visits	Children With CAs	Yes	4213 (45.9%)	3049 (33.3%)	2480 (27.0%)	2199 (24.0%)	2031 (22.2%)	5909 (64.4%)
		No	4956 (54.1%)	6120 (33.7%)	6689 (73.0%)	6970 (76.0%)	7138 (77.8%)	3260 (35.6%)
	Children Without CAs	Yes	2718 (29.6%)	2097 (22.9%)	1681 (18.3%)	1524 (16.6%)	1435 (15.7%)	4547 (49.6%)
		No	6451 (70.4%)	7072 (77.1%)	7488 (81.7%)	7645 (83.4%)	7734 (84.3%)	4622 (50.4%)
	χ^2 (sig. level)		518.402 **(<0.001)** *	244.819 **(<0.001)** *	198.456 **(<0.001)** *	153.556 **(<0.001)** *	126.371 **(<0.001)** *	412.766 **(<0.001)***
Travel for Hospital Admission	Children With CAs	Yes	2391 (26.1%)	692 (7.5%)	443 (4.8%)	351 (3.8%)	330 (3.6%)	2982 (32.5%)
		No	6778 (73.9%)	8477 (92.5%)	8726 (95.2%)	8818 (96.2%)	8839 (96.4%)	6187 (67.5%)
	Children Without CAs	Yes	1449 (15.8%)	251 (2.7%)	157 (1.7%)	149 (1.6%)	131 (1.4%)	1796 (19.6%)
		No	7720 (84.2%)	8918 (97.3%)	9012 (98.3%)	9020 (98.4%)	9038 (98.6%)	7373 (80.4%)
	χ^2 (sig. level)		292.290 **(<0.001)** *	217.417 **(<0.001)** *	140.938 **(<0.001)** *	83.895 **(<0.001)** *	88.118 **(<0.001)** *	398.121 **(<0.001)***

*** Statistically Significant**

As seen in Table 4.4, there is not a statistical difference in RHA of residence between children with and without CAs for any of the five years of follow-up. However, there is a statistically significant difference for the other three time-varying predictors for each of the five years of follow-up. Table 4.4 shows that the families of children with birth defects are more likely to be receiving some form of governmental social assistance, and

that these children travel outside of their home RHA for physician visits and for hospitalizations more frequently than children without birth defects.

4.3 Regional Differences in the Prevalence of Congenital Anomalies

Figure 4.6 illustrates the difference in the prevalence of congenital anomalies per 1000 live births per region. While graphical analysis did not reveal an overall difference in congenital anomalies by RHA, Table 4.5 and Figures 4.7 through 4.19 illustrate the regional differences in specific CAs (see Appendix B for graphs of the mean numbers of CAs per RHA).

Table 4.5: Regional differences in the mean number of cases of CAs compared to the Saskatoon Health Region

ICD-9 Code	Variable Description	RHAs that are Significantly Different from Saskatoon	Figure Where Data is Presented
ICD-9 743	CAs of the eye	Regina Qu'Appelle PA Parkland	Figure 4.7
ICD-9 745	Bulbus cordis anomalies and anomalies of cardiac septal closure	Heartland Northern Saskatchewan	Figure 4.8
ICD-9 746	Other CAs of the heart	PA Parkland Prairie North Northern Saskatchewan	Figure 4.9
ICD-9 747	Other CAs of the circulatory system	Sun Country Cypress Regina Qu'Appelle Sunrise Heartland	Figure 4.10
ICD-9 749	Cleft lip and cleft palate	Northern Saskatchewan	Figure 4.11
ICD-9 750	CAs of the upper alimentary tract	Sun Country Five Hills Cypress Regina Qu'Appelle Kelsey Trail	Figure 4.12
ICD-9 754	Certain congenital musculoskeletal deformities	Regina Qu'Appelle	Figure 4.13

ICD-9 755	Other CAs of limbs	Sun Country Five Hills Cypress PA Parkland Prairie North	Figure 4.14
ICD-9 756	Other musculoskeletal anomalies	Regina Qu'Appelle	Figure 4.15
ICD-9 757	CAs of the integument	Sun Country Regina Qu'Appelle PA Parkland	Figure 4.16
ICD-9 758	Chromosomal anomalies	Regina Qu'Appelle Sunrise	Figure 4.17
ICD-9 759	Other and unspecified CAs	Sun Country Five Hills Cypress Regina Qu'Appelle Sunrise	Figure 4.18
MSB Z60	Congenital dysplasia of hip	Kelsey Trail Prairie North	Figure 4.19

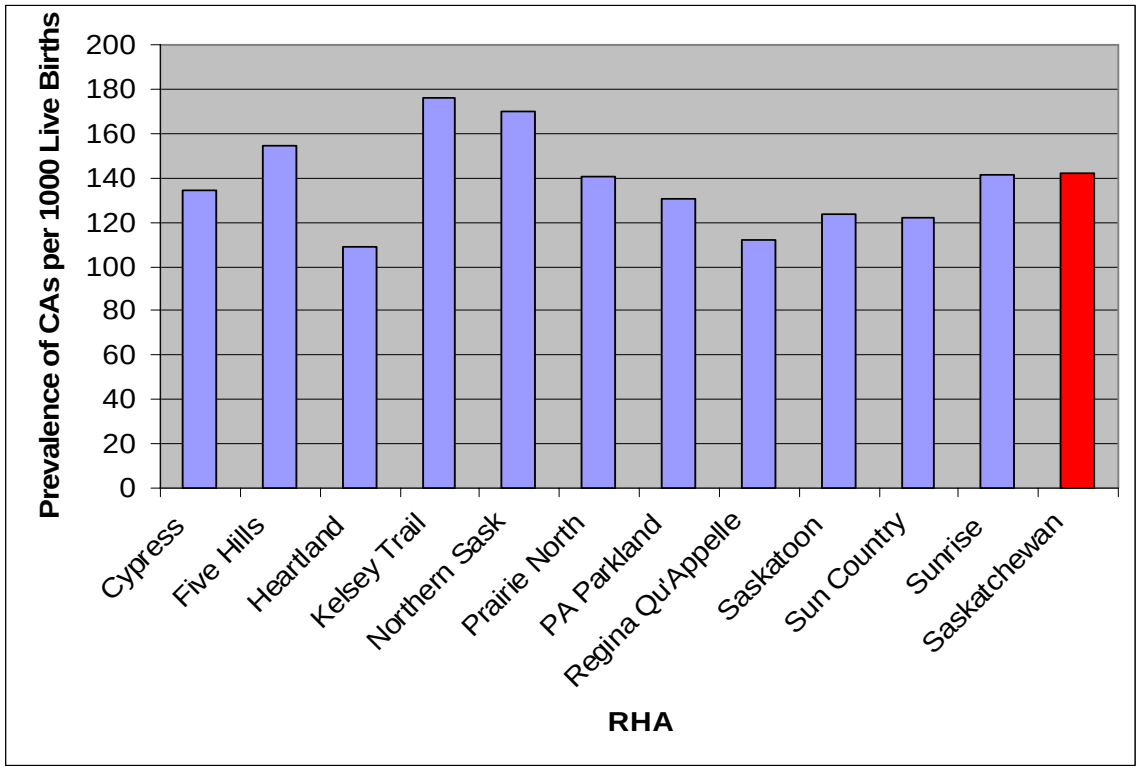
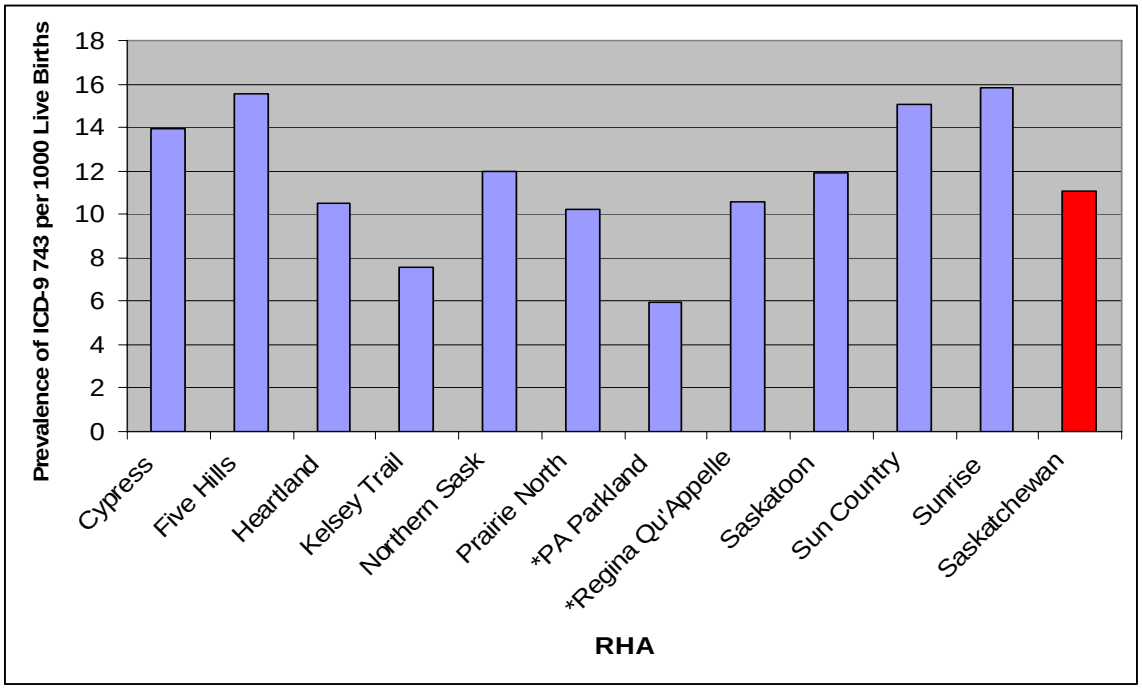
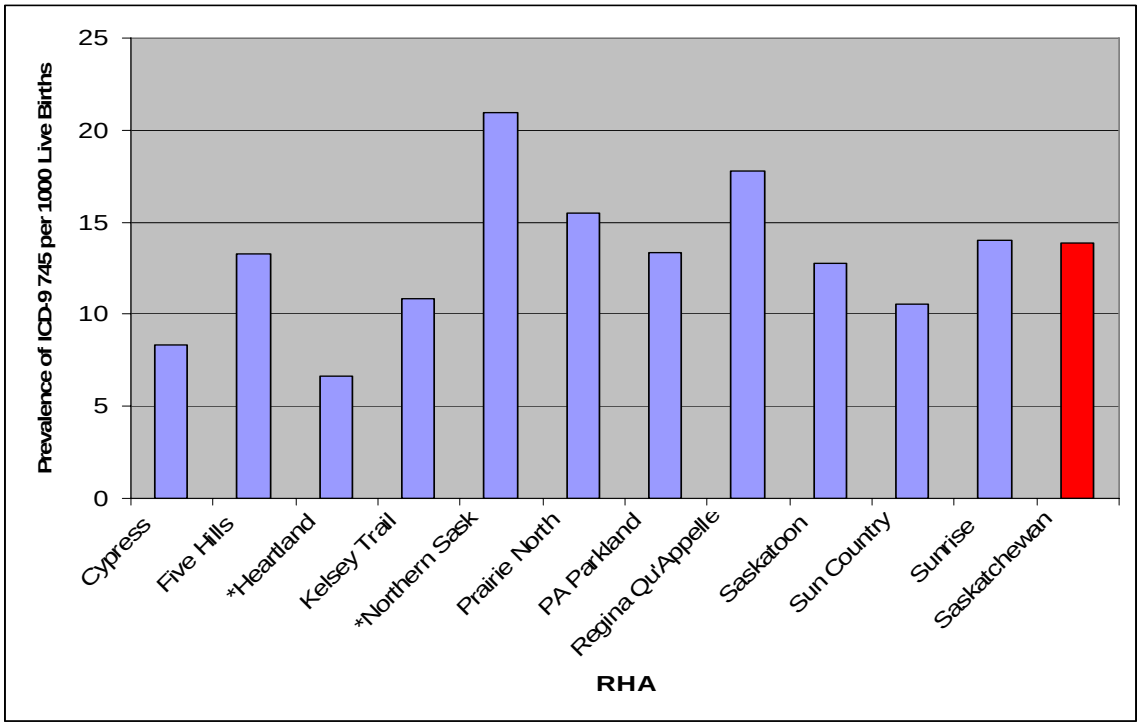


Figure 4.6: Prevalence of any CAs by RHA – No regional differences were found in the overall prevalence of birth defects compared to the Saskatoon Health Region (reference category).



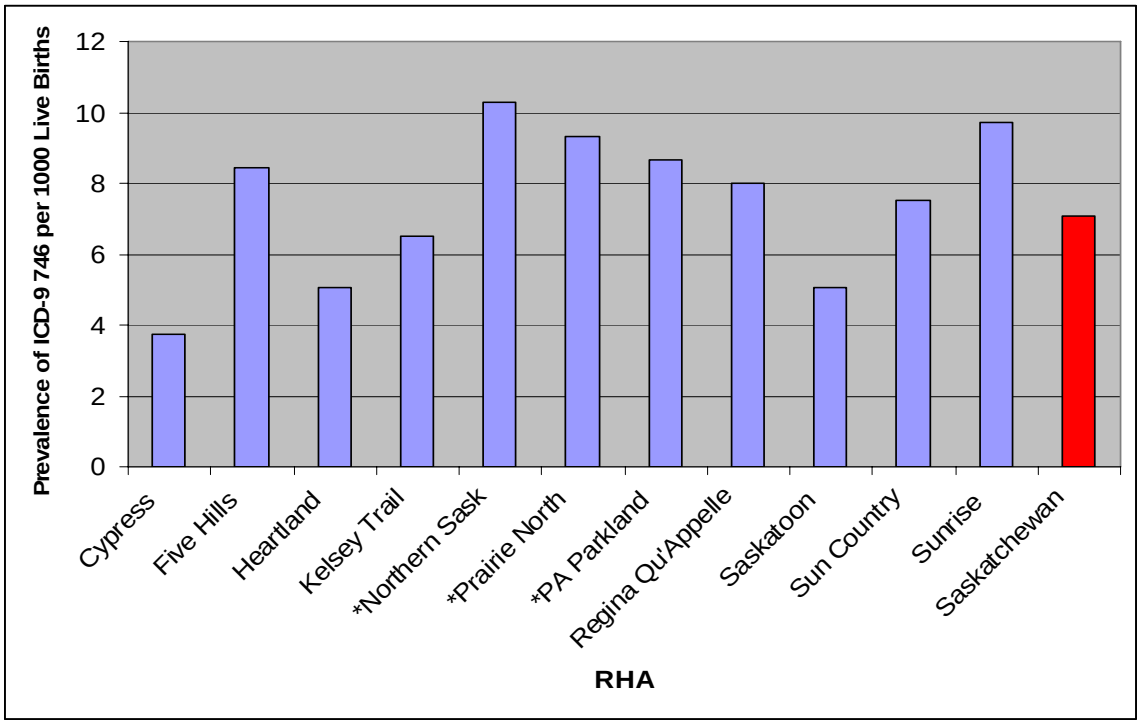
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.7: Prevalence of CAs of the eye (ICD-9 743) by RHA



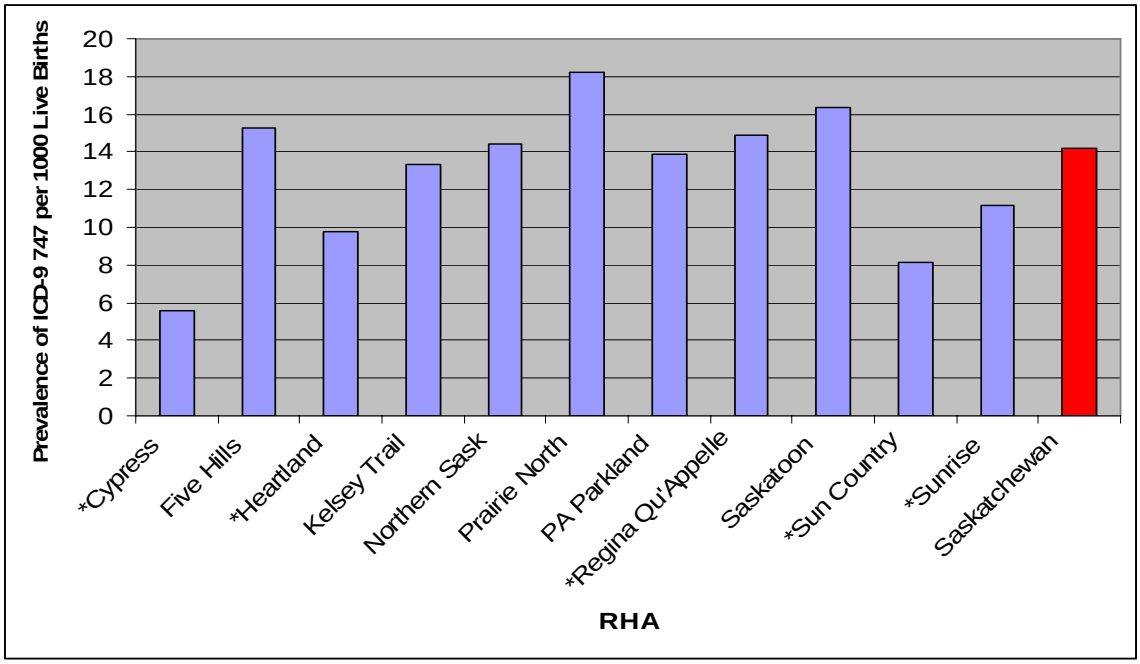
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.8: Prevalence of bulbus cordis anomalies and anomalies of cardiac septal closure (ICD-9 745) by RHA



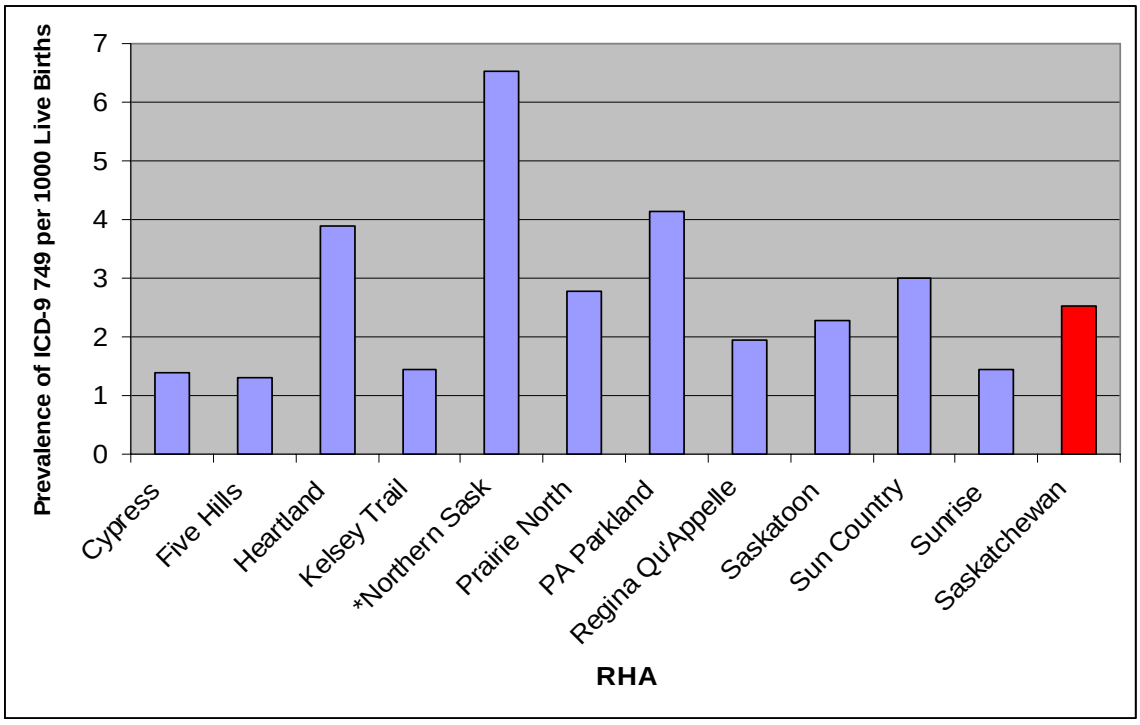
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.9: Prevalence of other CAs of the heart (ICD-9 746) by RHA



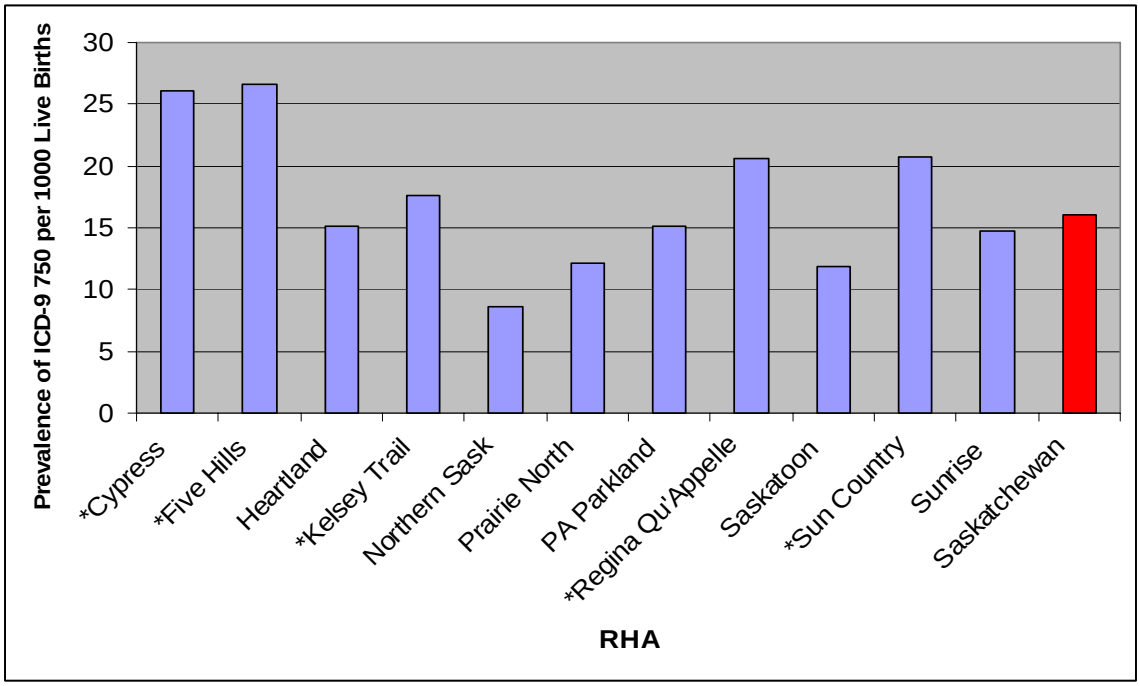
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.10: Prevalence of other CAs of the circulatory system (ICD-9 747) by RHA



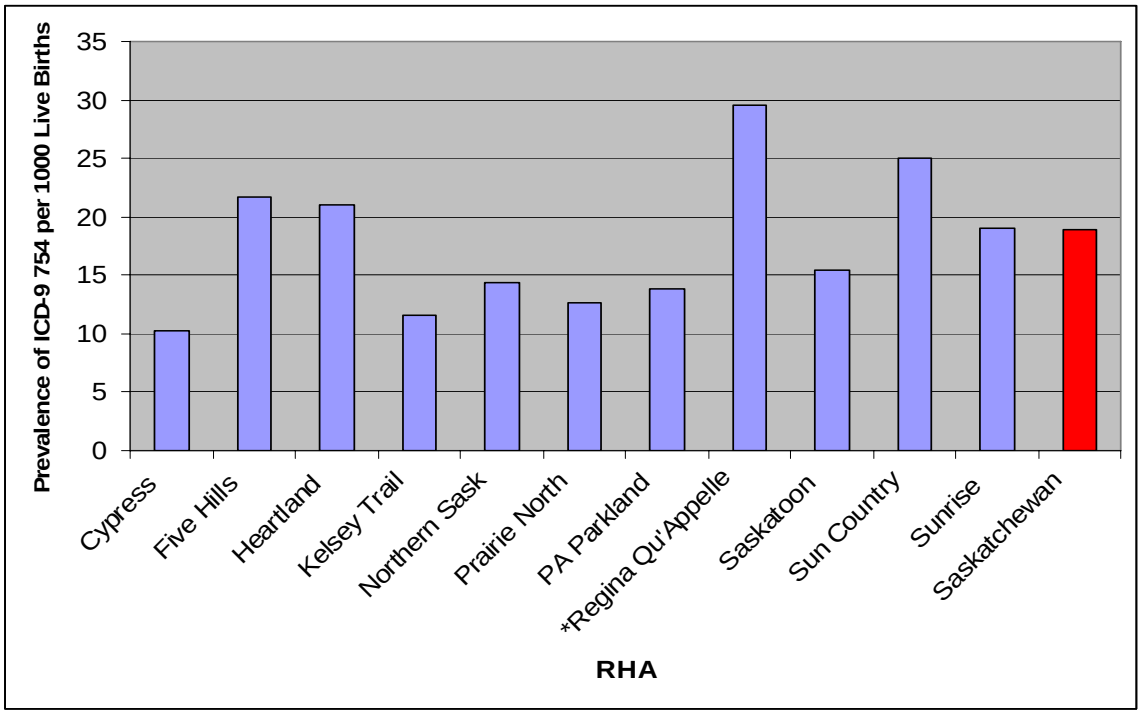
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.11: Prevalence of cleft lip and cleft palate (ICD-9 749) by RHA



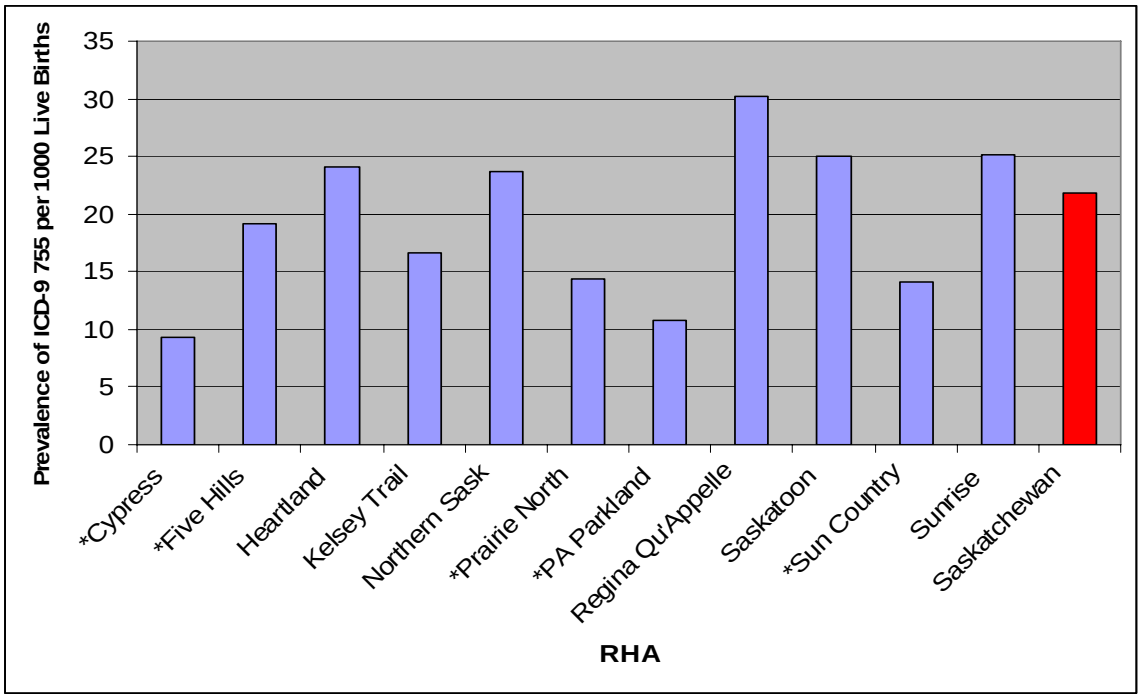
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.12: Prevalence of CAs of the upper alimentary tract (ICD-9 750) by RHA



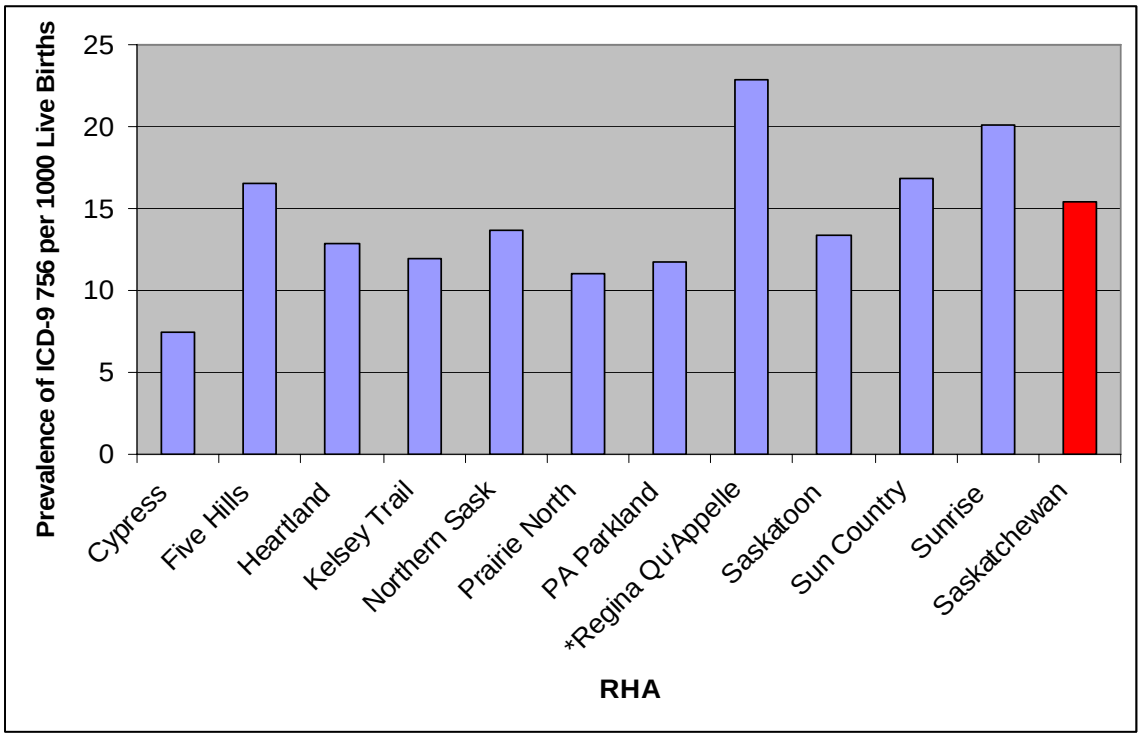
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.13: Prevalence of certain congenital musculoskeletal deformities (ICD-9 754) by RHA



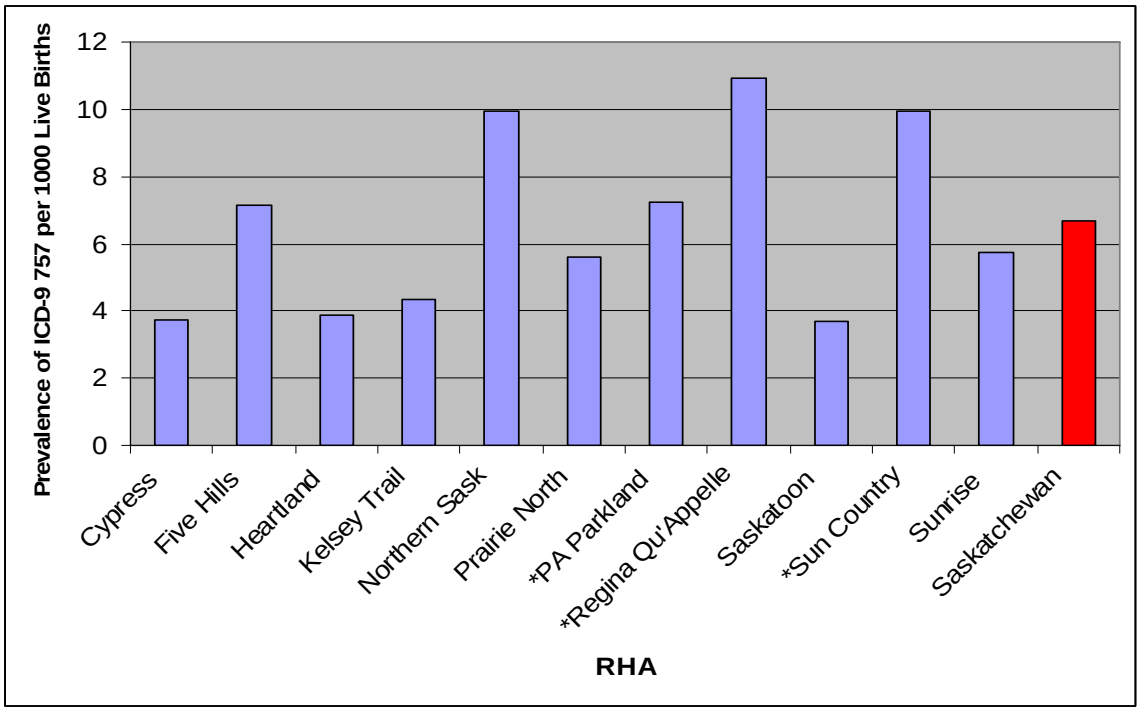
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.14: Prevalence of other CAs of limbs (ICD-9 755) by RHA



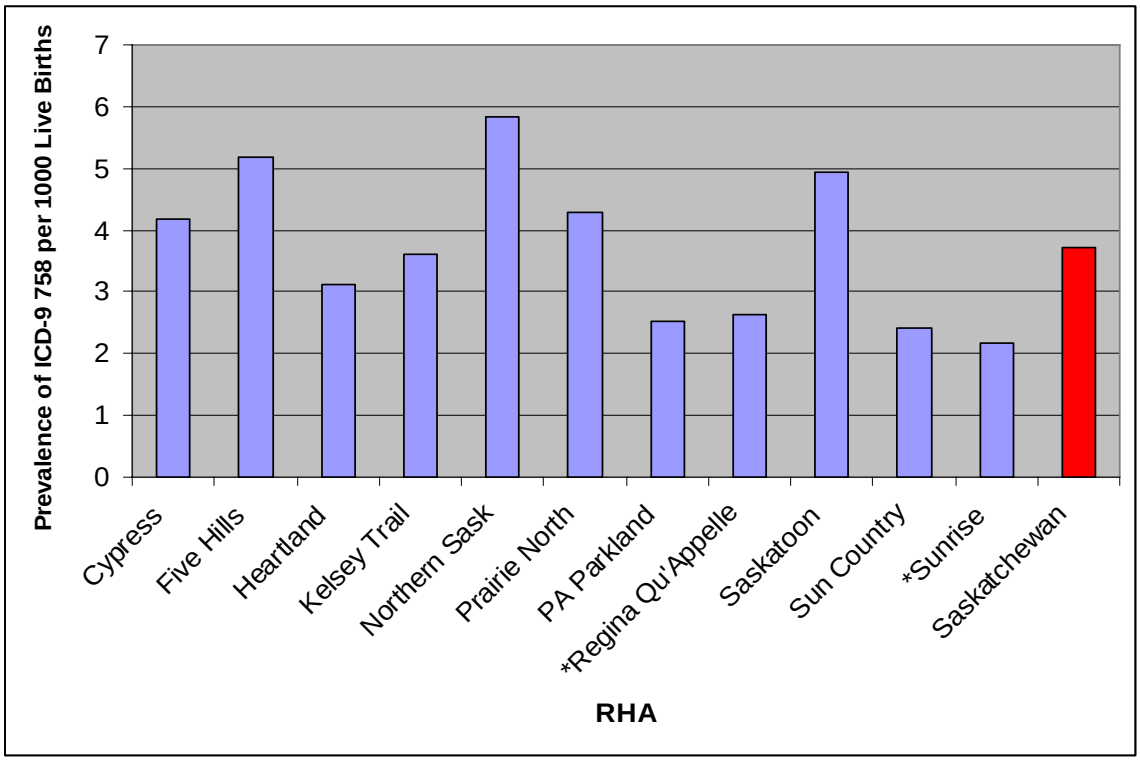
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.15: Prevalence of other musculoskeletal anomalies (ICD-9 756) by RHA



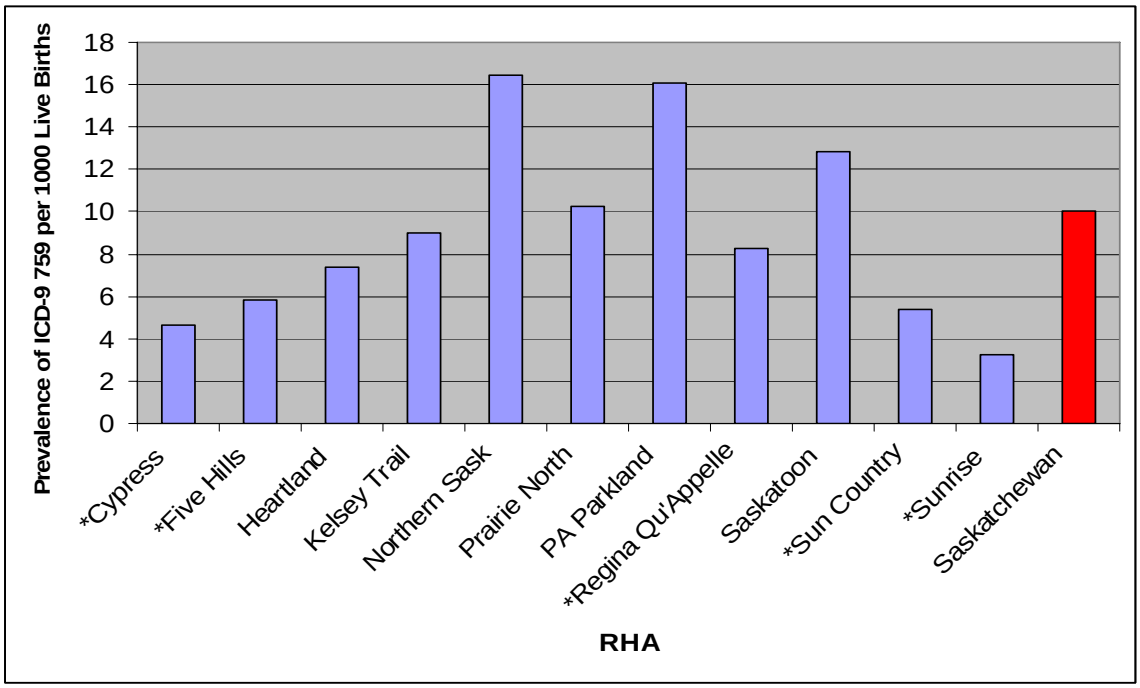
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.16: Prevalence of CAs of the integument (ICD-9 757) by RHA



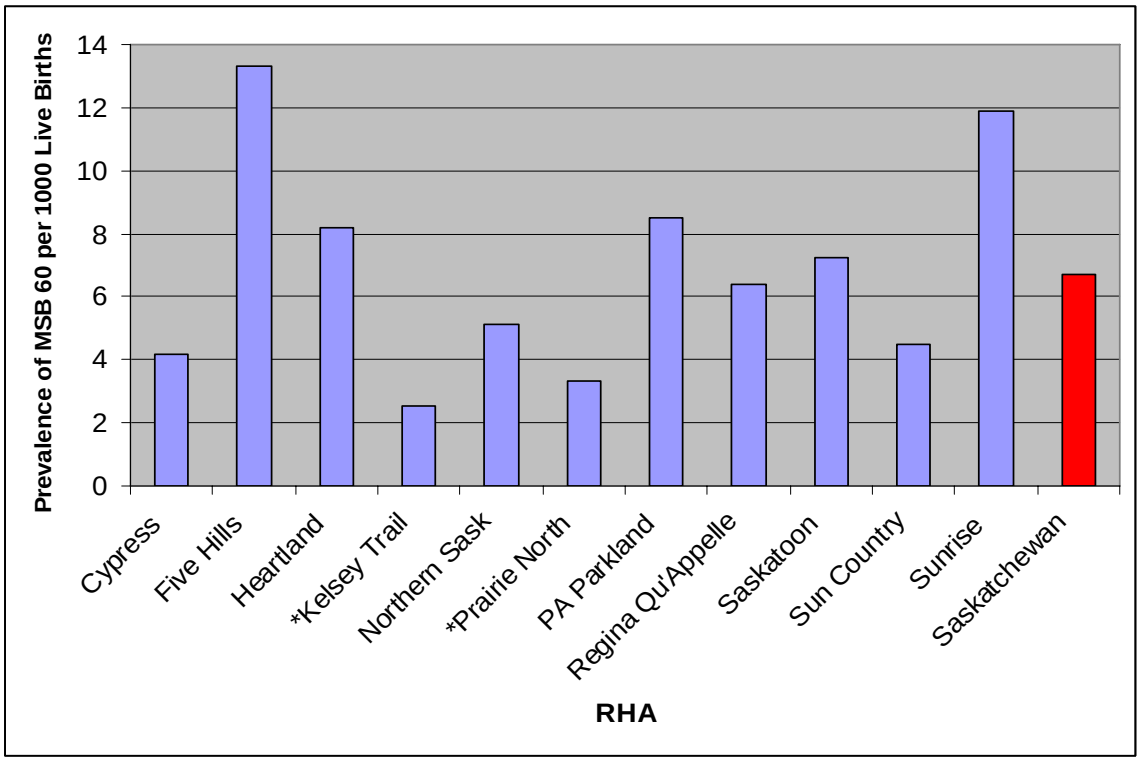
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.17: Prevalence of chromosomal anomalies (ICD-9 758) by RHA



*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.18: Prevalence of other and unspecified CAs (ICD-9 759) by RHA



*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.19: Prevalence of congenital dysplasia of hip (MSB Z60) by RHA

4.4 Study Question 1: Is the Level of Health Care Used by Children With Congenital Anomalies Significantly Different from the Level of Health Care Used by Children Without Congenital Anomalies?

As indicated in Table 4.6, one-way ANOVAs revealed that at the 5% level of significance, there is a statistically significant difference in the number of physician visits, the number of hospitalizations and total length of stay by CA status (i.e. child has at least one CA).

Table 4.6: Differences in use of health care for children with and without congenital anomalies

CA Status	Category	Total Number of Physician Visits in the first 5 Years	Total Number of Hospitalizations in the first 5 Years	Total Length of Stay (days) in the first 5 Years
Children With Congenital Anomalies	Mean	46.07	2.81	13.36
	Median	39.00	2.00	5.00
	Mode	31	1	3
	Range	0-713	0-67	0-779
Children Without Congenital Anomalies	Mean	34.07	1.83	5.31
	Median	30.00	1.00	3.00
	Mode	22	1	3
	Range	0-222	0-18	0-205
	F statistic (significance level)	**856.674 (< 0.001)***	**690.946 (< 0.001)***	**515.558 (< 0.001)***

*** Statistically Significant**

When the results of Table 4.6 are further broken down into differences between children with specific CAs and children without CAs, one way ANOVAs revealed that at the 5% level of significance, there is a statistically significant difference in the number of physician visits, the number of hospitalizations and length of stay for almost all conditions (see Table 4.7). The most notable exception here is for CAs of the integument (ICD-9 757) where children with conditions in this particular class use the same level of health care services as children without any CAs for all three measures of health care usage.

Table 4.7: Differential use of health care services for children with specific types of CAs versus children without that particular CA

Type of CA	**ICD-9 Code**	**Total Number of Physician Visits** F statistic (significance level)	**Total Number of Hospitalizations** F statistic (significance level)	**Total Length of Stay** F statistic (significance level)
Neural Tube Defects	ICD-9 740 & ICD-9 741	**156.779 (<0.001)***	**276.626 (<0.001)***	**129.194 (<0.001)***
Other congenital anomalies of nervous system	ICD-9 742	**403.064 (<0.001)***	**620.860 (<0.001)***	**592.405 (<0.001)***
Congenital anomalies of eye	ICD-9 743	**96.847 (<0.001)***	**60.384 (<0.001)***	**24.900 (<0.001)***
Congenital anomalies of ear, face, and neck	ICD-9 744	**23.402 (<0.001)***	**23.800 (<0.001)***	2.168 (0.141)
Bulbus cordis anomalies and anomalies of cardiac septal closure	ICD-9 745	**481.091 (<0.001)***	**463.171 (<0.001)***	**715.613 (<0.001)***
Other congenital anomalies of heart	ICD-9 746	**366.084 (<0.001)***	**469.535 (<0.001)***	**576.492 (<0.001)***
Other congenital anomalies of circulatory system	ICD-9 747	**691.150 (<0.001)***	**586.159 (<0.001)***	**2472.400 (<0.001)***
Congenital anomalies of respiratory system	ICD-9 748	**170.914 (<0.001)***	**402.527 (<0.001)***	**489.525 (<0.001)***
Cleft palate and cleft lip	ICD-9 749	**149.026 (<0.001)***	**292.704 (<0.001)***	**95.632 (<0.001)***
Other congenital anomalies of upper alimentary tract	ICD-9 750	**30.299 (<0.001)***	**51.254 (<0.001)***	2.086 (0.149)
Other congenital anomalies of digestive system	ICD-9 751	**194.283 (<0.001)***	**368.020 (<0.001)***	**401.636 (<0.001)***
Congenital anomalies of genital organs	ICD-9 752	**40.953 (<0.001)***	**26.832 (<0.001)***	0.005 (0.946)
Congenital anomalies of urinary system	ICD-9 753	**77.501 (<0.001)***	**129.859 (<0.001)***	**69.795 (<0.001)***

Certain congenital musculoskeletal deformities	ICD-9 754	**69.805 (<0.001)***	**30.484 (<0.001)***	0.102 (0.749)
Other congenital anomalies of limbs	ICD-9 755	**9.897 (0.002)***	0.170 (0.680)	**7.073 (0.008)***
Other congenital musculoskeletal anomalies	ICD-9 756	**149.397 (<0.001)***	**72.907 (<0.001)***	**87.293 (<0.001)***
Congenital anomalies of the integument	ICD-9 757	1.172 (0.279)	1.104 (0.294)	0.565 (0.452)
Chromosomal anomalies	ICD-9 758	**271.152 (<0.001)***	**275.831 (<0.001)***	**209.062 (<0.001)***
Other and unspecified congenital anomalies	ICD-9 759	**258.947 (<0.001)***	**254.123 (<0.001)***	**337.737 (<0.001)***
Congenital dysplasia of hip	MSB Z60	3.394 (0.065)	1.248 (0.264)	**6.050 (0.014)***
Clubfoot	MSB Z61	**58.120 (<0.001)***	**33.247 (<0.001)***	**14.060 (<0.001)***

*** Statistically Significant**

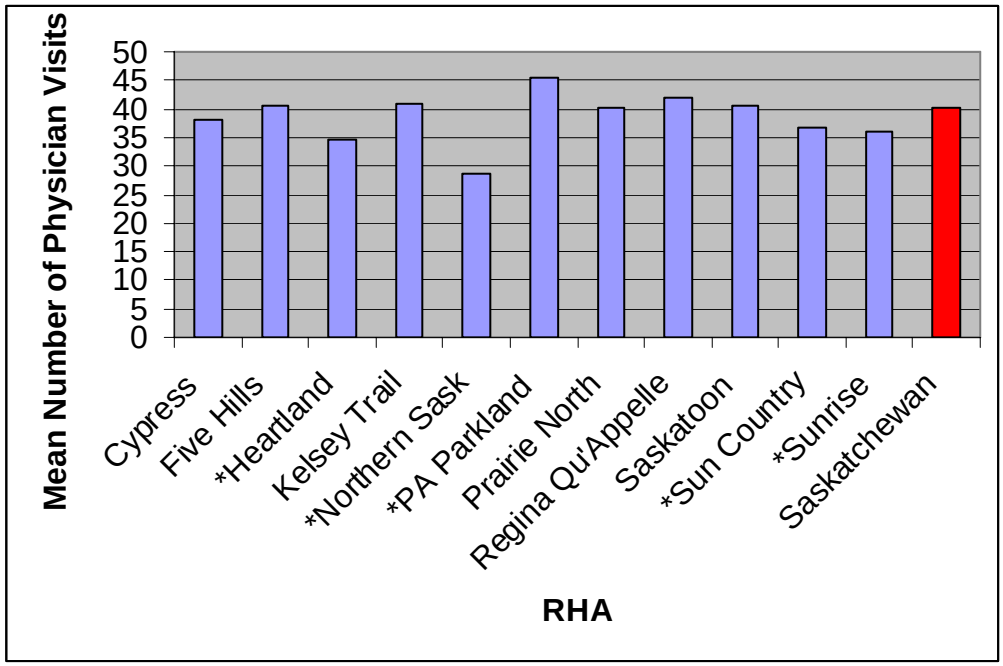
4.5 Study Question 2: Is There a Regional Difference in the Level of Health Care Used by Children in their First Five Years of Life? And Does This Relationship Hold For Children With and Without CAs?

4.5.1 Regional Differences in Use of Health Care Services for All Children

One-way ANOVAs and Bonferroni post-hoc tests revealed that at the 5% level of significance there is a crude regional difference in the total number of physician visits ($F=26.641$, $p<0.001$), hospitalizations ($F=34.181$, $p<0.001$) and length of stay ($F=9.905$, $p<0.001$). Table 4.8 and Figures 4.20-4.22 provide the results of the post-hoc tests and show the specific regional differences when the Saskatoon Health Region is used as a reference category.

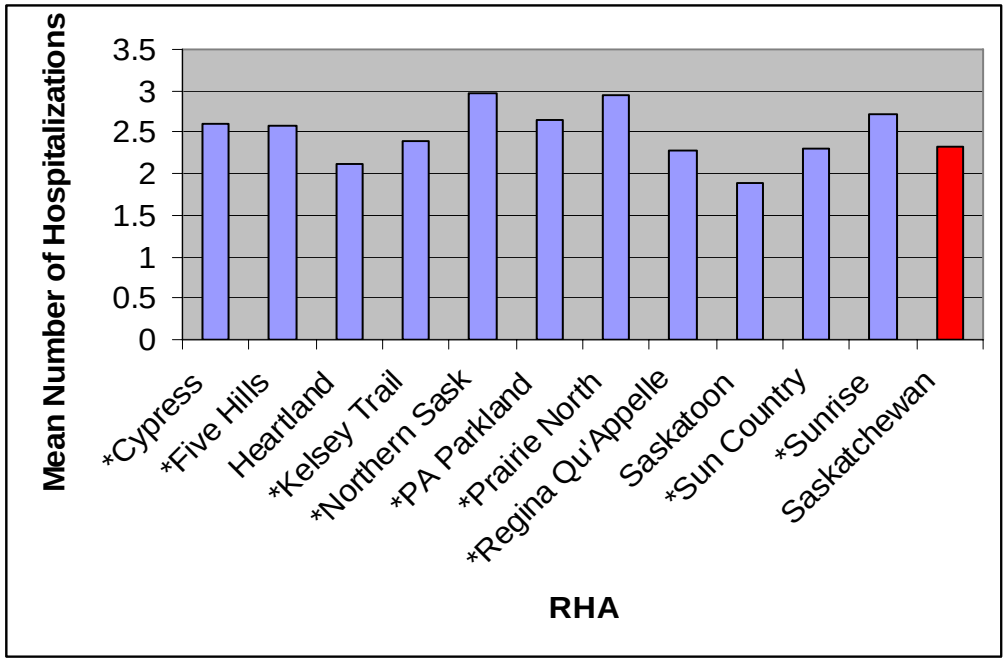
Table 4.8: Crude regional differences in use of health care services

Variable	RHAs that are Significantly Different from the Saskatoon Health Region (significance level)	Figure Where Data is Presented
Total Number of Physician Visits	Sun Country (0.008) Sunrise (<0.001) Heartland (<0.001) PA Parkland (<0.001) Northern Saskatchewan (<0.001)	Figure 4.20
Total Number of Hospitalizations	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Regina Qu'Appelle (<0.001) Sunrise (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)	Figure 4.21
Total Length of Stay	PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)	Figure 4.22



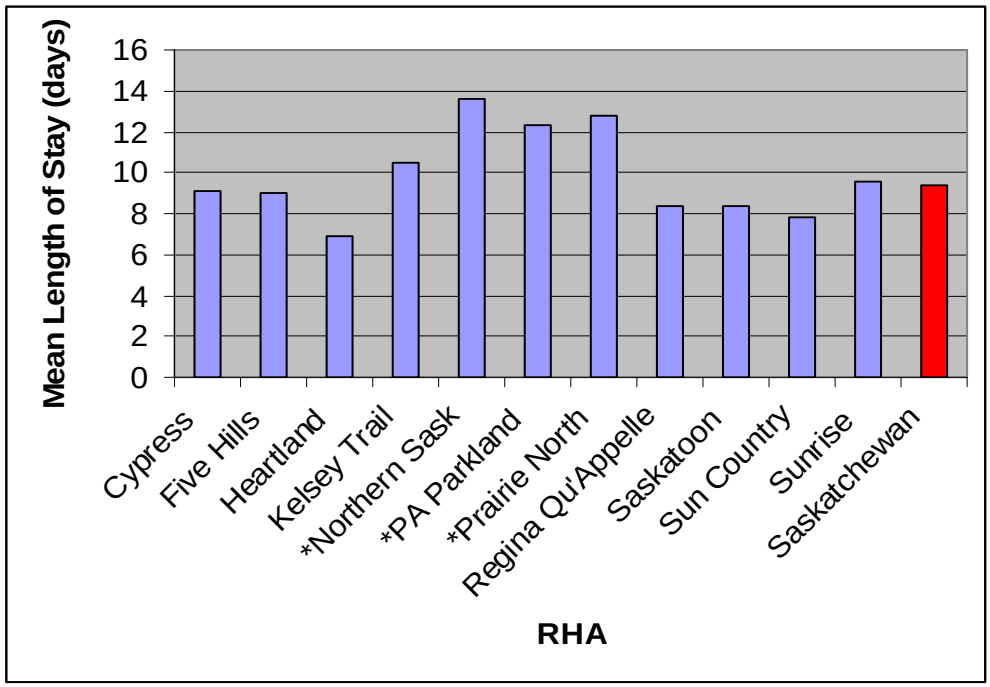
*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.20: Crude regional differences in the mean number of physician visits in the first five years of life compared to the Saskatoon Health Region



*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.21: Crude regional differences in the mean number of hospitalizations in the first five years of life compared to the Saskatoon Health Region



*** Significantly different from the Saskatoon Health Region (reference category)**

Figure 4.22: Crude regional difference in the mean length of stay (days) in the first five years of life compared to the Saskatoon Health Region

With regard to travel to access health care services, one-way ANOVAs indicate that at the 5% level of significance, there is a significant difference in number of residents ever having to travel outside of their home RHA for a physician visit for all regions when the Saskatoon Health Region is used as the reference group ($F=372.859$, $p<0.001$) (see Table 4.9). Additionally, as seen in Table 4.9, one-way ANOVAs show that at the 5% level of significance, there is a significant difference in the number of residents ever having to travel outside of their home RHA for a hospital admission for all regions except for Regina Qu'Appelle when the Saskatoon Health Region is used as the reference group ($F=660.888$, $p<0.001$).

Table 4.9: Crude regional differences in travel for health care services

Variable	**RHAs that are Significantly Different from the Saskatoon Health Region (significance level)**
Ever Traveled Outside of Home RHA for a Physician Visits	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Regina Qu'Appelle (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)
Ever Traveled Outside of Home RHA for a Hospital Admission	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)

4.5.2 Regional Differences in Use of Health Care Services for Children With and Without Congenital Anomalies

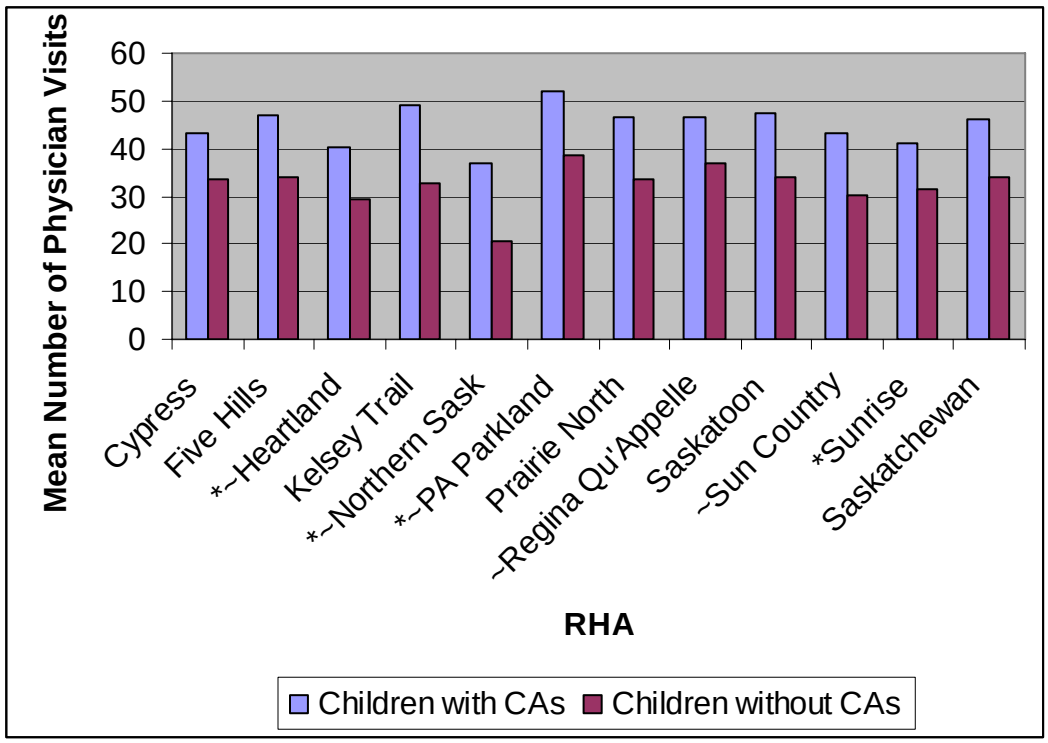
When use of health care services was considered for children with at least one congenital anomaly, one-way ANOVAs and Bonferroni post-hoc tests revealed that at

the 5% level of significance there is a regional difference in the total number of physician visits ($F=9.251$, $p<0.001$), hospitalizations ($F=18.538$, $p<0.001$) and length of stay ($F=8.047$, $p<0.001$). This is also the case when one exclusively examines the use of health care services for children without any congenital anomalies [physician visits ($F=29.945$, $p<0.001$), hospitalizations ($F=31.059$, $p<0.001$) and length of stay ($F=14.265$, $p<0.001$)]. While the same overall results were seen for children with and without congenital anomalies, different patterns of regional differences are revealed by the post-hoc tests when the Saskatoon Health Region is used as a reference group (see Table 4.10, Figures 4.23-4.25)

Table 4.10: Crude regional differences in health care utilization for children with and without congenital anomalies

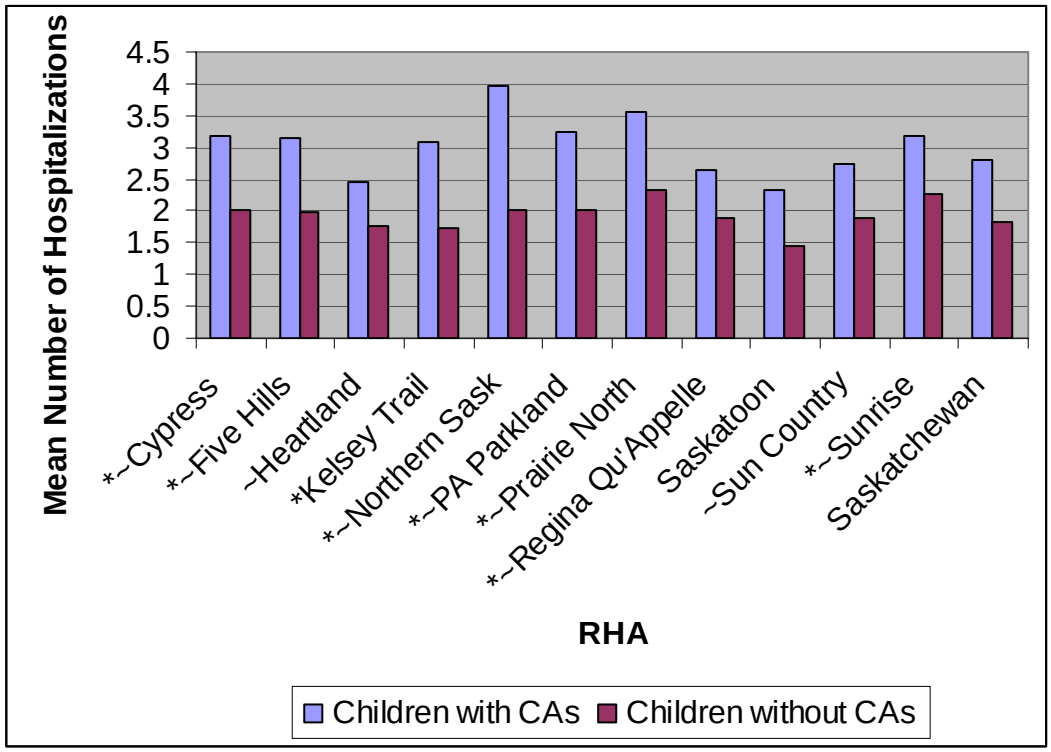
Variable	RHAs that are Significantly Different from Saskatoon for Children WITH CAs (significance level)	RHAs that are Significantly Different from Saskatoon for Children WITHOUT CAs (significance level)
Total Number of Physician Visits	Sunrise (0.006) Heartland (0.009) PA Parkland (0.028) Northern Saskatchewan (<0.001)	Sun Country (0.047) Regina Qu'Appelle (<0.001) Heartland (0.011) PA Parkland (<0.001) Northern Saskatchewan (<0.001)
	See Figure 4.23	
Total Number of Hospitalizations	Five Hills (<0.001) Cypress (0.007) Regina Qu'Appelle (0.020) Sunrise (<0.001) Kelsey Trail (0.006) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Regina Qu'Appelle (<0.001) Sunrise (<0.001) Heartland (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)
	See Figure 4.24	

Total Length of Stay	PA Parkland (0.006) Prairie North (0.001) Northern Saskatchewan (<0.001)	Five Hills (0.018) Cypress (0.028) Regina Qu'Appelle (<0.001) Sunrise (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)
	See Figure 4.25	



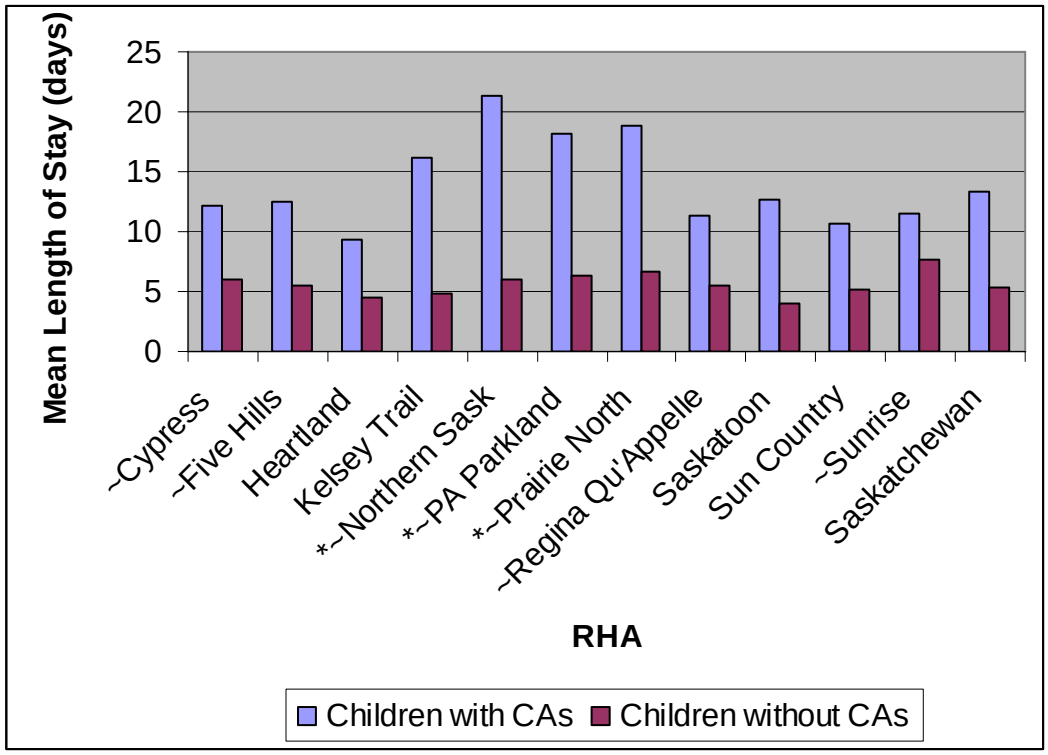
*** Significantly different from the Saskatoon Health Region (reference category) for children with CAs**
~ Significantly different from the Saskatoon Health Region (reference category) for children without CAs

Figure 4.23: Crude regional differences in the mean number of physician visits in the first five years of life compared to the Saskatoon Health Region for children with and without congenital anomalies



*** Significantly different from the Saskatoon Health Region (reference category) for children with CAs**
~ Significantly different from the Saskatoon Health Region (reference category) for children without CAs

Figure 4.24: Crude regional differences in the mean number of hospitalizations in the first five years of life compared to the Saskatoon Health Region for children with and without congenital anomalies



*** Significantly different from the Saskatoon Health Region (reference category) for children with CAs**
~ Significantly different from the Saskatoon Health Region (reference category) for children without CAs

Figure 4.25: Crude regional difference in the mean length of stay (days) in the first five years of life compared to the Saskatoon Health Region for children with and without congenital anomalies

As can be seen in Table 4.11, with regard to travel to access health care services for children with and without congenital anomalies, one way ANOVAs show that at the 5% level of significance, for children with congenital anomalies there is a significant difference in number of residents ever having to travel outside of their home RHA for a physician visit for all regions when the Saskatoon Health Region is used as the reference group ($F=252.396$, $p<0.001$) (see Table 4.11). For children without CAs there is a significant difference in number of residents ever having to travel outside of their home RHA for a physician visit for all regions except for Regina Qu'Appelle when the Saskatoon Health Region is used as the reference group ($F=152.977$, $p<0.001$).

Additionally, as seen in Table 4.11, one way ANOVAs show that at the 5% level of significance, there is a difference in number of residents ever having to travel outside of their home RHA for a hospital admission for all regions except for Regina Qu'Appelle when the Saskatoon Health Region is used as the reference group – this is the same for children with and without CAs (children with CAs: $F=432.929$, $p<0.001$, children without CAs: $F=289.237$, $p<0.001$).

Table 4.11: Crude regional differences in travel for health care utilization for children with and without congenital anomalies

Variable	RHAs that are Significantly Different from Saskatoon for Children WITH CAs (significance level)	RHAs that are Significantly Different from Saskatoon for Children WITHOUT CAs (significance level)
Ever Traveled Outside of Home RHA for a Physician Visits	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Regina Qu'Appelle (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)
Ever Traveled Outside of Home RHA for a Hospital Admission	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)	Sun Country (<0.001) Five Hills (<0.001) Cypress (<0.001) Sunrise (<0.001) Heartland (<0.001) Kelsey Trail (<0.001) PA Parkland (<0.001) Prairie North (<0.001) Northern Saskatchewan (<0.001)

4.6 Study Question 3: What Factors Influence the Level of Health Care Utilization?

As the literature indicates that different factors influence the use of different types of health care, separate models were constructed to predict the total number of physician visits in the study period, the total number of hospitalizations in the study period and the total length of stay (LOS) in hospital during the study period.

4.6.1 Model for Number of Physician Visits

Table 4.12 describes the statistically significant predictors of the total number of physician visits for children in Saskatchewan in their first five years of life.

Table 4.12: Statistically significant predictors of total number of physician visits in the first five years of life for children with CAs compared to children without CAs

Variable	Categories	Odds Ratio	95% Confidence Interval
Main Effects			
Need Characteristics			
Congenital Anomaly	No	(ref)	(ref)
	Yes	**1.19**	**1.16-1.21***
Multiple Congenital Anomalies	No	(ref)	(ref)
	Yes	**1.37**	**1.34-1.41***
Predisposing Characteristics			
Baby's Sex	Male	(ref)	(ref)
	Female	**0.92**	**0.90-0.93***
Registered Indian Status	No	(ref)	(ref)
	Yes	**1.35**	**1.31-1.38***
Parity	1	(ref)	(ref)
	2	**0.94**	**0.92-0.96***
	3	**0.89**	**0.87-0.91***
	4	**0.84**	**0.81-0.87***
	≥5	**0.82**	**0.79-0.86***
Enabling Characteristics			
RHA of Residence in Birth Year	Saskatoon	(ref)	(ref)
	Sun Country	**0.63**	**0.57-0.69***
	Five Hills	**0.84**	**0.79-0.90***
	Cypress	**0.84**	**0.76-0.92***
	Regina Qu'Appelle	1.01	0.98-1.04
	Sunrise	**0.74**	**0.69-0.80***
	Heartland	**0.51**	**0.43-0.61***
	Kelsey Trail	**0.69**	**0.61-0.78***
	PA Parkland	**0.90**	**0.84-0.96***
	Prairie North	**0.62**	**0.57-0.67***
	Northern Saskatchewan	**0.23**	**0.19-0.28***
Family Ever Received Income Assistance	No	(ref)	(ref)
	Yes	**1.12**	**1.10-1.14***
Child Ever Traveled Outside of Home RHA for a Physician Visit	No	(ref)	(ref)
	Yes	**1.97**	**1.65-2.37***

Interaction Terms			
RHA x Child Ever Traveled Outside of Home RHA for a Physician Visit	**Saskatoon x Yes Travel**	**0.60**	**0.50-0.72***
	Sun Country x Yes Travel	0.87	0.71-1.07
	Five Hills x Yes Travel	**0.73**	**0.60-0.89***
	Cypress x Yes Travel	**0.70**	**0.57-0.87***
	Regina Qu'Appelle x Yes Travel	**0.61**	**0.51-0.73***
	Sunrise x Yes Travel	**0.74**	**0.60-0.90***
	Heartland x Yes Travel	Dropped	Dropped~
	Kelsey Trail x Yes Travel	0.82	0.65-1.03
	PA Parkland x Yes Travel	**0.67**	**0.55-0.82***
	Prairie North x Yes Travel	0.88	0.72-1.08
	Northern Saskatchewan x Yes Travel	**1.49**	**1.14-1.94***

*** Statistically Significant**
~ Dropped from the model due to collinearity

As shown in Table 4.12, children who have multiple congenital anomalies visit the doctor more often than children who do not have any birth defects (OR=1.19) and children who have multiple congenital anomalies visit the doctor more often than other children (OR=1.37). Females have fewer physician visits than males (OR=0.92), while children whose families had ever received some form of governmental income assistance had more physician visits than children whose families did not receive income assistance (OR=1.12). Furthermore, Registered Indian children visited a physician more frequently than the general population of children (OR=1.35). A dose-response relationship was observed for parity, with each additional child a mother had was associated with decreasing numbers of physician visits for the child.

The presence of statistically significant interaction terms between RHA of residence and the need to travel outside of one's home RHA to see a physician implies that neither of the terms can be examined in isolation. This means that when one looks at the impact of home RHA on the number of physician visits in the first five years, it must be explained in the context of the need to travel outside of one's home RHA to see a physician (i.e. the relationship between home RHA and physician visits is different for

children who had to travel outside of their home RHA to see a physician in the first five years of life versus children who did not have to travel outside of their home RHA to see a physician in the first five years of life).

4.6.2 Model for Total Number of Hospitalizations

Table 4.13 shows the variables that are statistically significant predictors of the total number of hospitalizations for children in Saskatchewan in the first five years of life.

Table 4.13: Statistically significant predictors of total number of hospitalizations in the first five years of life for children with CAs compared to children without CAs

Variable	Categories	Odds Ratio	95% Confidence Interval
Main Effects			
Need Characteristics			
Congenital Anomaly	No	(ref)	(ref)
	Yes	**1.24**	**1.21-1.27***
Multiple Congenital Anomalies	No	(ref)	(ref)
	Yes	**1.50**	**1.45-1.54***
Birth Weight	<1000g	(ref)	(ref)
	1000g-1499g	0.99	0.86-1.12
	1500g-1999g	1.03	0.91-1.16
	2000g-2499g	0.94	0.84-1.05
	2500g-3999g	**0.86**	**0.78-0.95***
	≥4000g	**0.80**	**0.72-0.89***
Predisposing Characteristics			
Baby's Sex	Male	(ref)	(ref)
	Female	**0.86**	**0.85-0.88***
Registered Indian Status	No	(ref)	(ref)
	Yes	**1.48**	**1.43-1.54***
Mother's Age Group	< 20	(ref)	(ref)
	20-34	**0.91**	**0.87-0.94***
	≥35	**0.86**	**0.82-0.91***
Mother's Marital Status	Single	(ref)	(ref)
	Married/Common Law	**0.93**	**0.90-0.96***
	Other	0.99	0.94-1.04
Parity	1	(ref)	(ref)
	2	**1.05**	**1.02-1.08***
	3	**1.06**	**1.03-1.10***
	4	**1.08**	**1.03-1.13***
	≥5	**1.15**	**1.09-1.21***

Enabling Characteristics			
RHA of Residence in Birth Year	Saskatoon	(ref)	(ref)
	Sun Country	0.97	0.91-1.02
	Five Hills	**1.17**	**1.11-1.23***
	Cypress	**1.24**	**1.16-1.33***
	Regina Qu'Appelle	**1.17**	**1.13-1.21***
	Sunrise	**1.25**	**1.19-1.32***
	Heartland	**0.81**	**0.76-0.87***
	Kelsey Trail	0.95	0.89-1.02
	PA Parkland	0.98	0.93-1.03
	Prairie North	**1.07**	**1.02-1.12***
	Northern Saskatchewan	**0.78**	**0.74-0.83***
Family Ever Received Income Assistance	No	(ref)	(ref)
	Yes	**1.12**	**1.09-1.15***
Child Ever Traveled Outside of Home RHA for a Hospitalization	No	(ref)	(ref)
	Yes	**1.75**	**1.70-1.80***
Child Moved to Another RHA	No	(ref)	(ref)
	Yes	**0.95**	**0.91-0.99***

*** Statistically Significant**

As can be seen in Table 4.13, the adjusted odds ratio for RHA is statistically significant for seven of the ten regions included, meaning that children who lived in Heartland (OR=0.81) and Northern Saskatchewan (OR=0.78) at the end of their first year of life are hospitalized less often in the first five years than children in Saskatoon (the reference category), and children in Five Hills (OR=1.17), Cypress (OR=1.24), Regina Qu'Appelle (OR=1.17) and Sunrise (OR=1.25) at the end of their first year of life are hospitalized more often in the first five years than children in Saskatoon (the reference category). Not surprisingly, children with at least one congenital anomaly were more likely to be hospitalized than children without congenital anomalies (OR=1.24) and children with multiple congenital anomalies were also more likely to be hospitalized than other children (OR=1.50). Females were less likely than males to be hospitalized (OR=0.86), and Registered Indian children were more likely than the general population

of children to be hospitalized (OR=1.48). Children who had moved to another RHA during the study period were less likely to be hospitalized than children who did not move to another RHA (OR=0.95), and children who had to travel outside of their home RHA for a hospitalization were more likely to be hospitalized than children who did not have to travel outside of their home RHA for a hospitalization (OR=1.75). With regards to maternal factors that were predictive of a child's number of hospitalizations, being in married or in a common law relationship at the time of the child's birth is associated with fewer hospitalizations, compared to being single (OR=0.93). Additionally, children whose mothers were between the ages of 20 and 34 (OR=0.91) and 35 years or older (OR=0.86) at the time of their child's birth were less likely to be hospitalized than children whose mothers were teenagers when they were born. A dose-response relationship was observed for parity, with each additional child a mother had increasing the number of hospitalizations for the child included in this study as compared to first time mothers.

4.6.3 Model for Total Length of Stay

Table 4.14 shows the variables that are statistically significant predictors of the total length of stay in hospital for children in Saskatchewan in the first five years of life.

Table 4.14: Statistically significant predictors of total length of stay in hospital in the first five years of life for children with CAs compared to children without CAs

Variable	Categories	Odds Ratio	95% Confidence Interval
Main Effects			
Need Characteristics			
Congenital Anomaly	No **Yes**	(ref) **1.26**	(ref) **1.22-1.29***
Multiple Congenital Anomalies	No **Yes**	(ref) **2.09**	(ref) **2.01-2.17***

Birth Weight	<1000g	(ref)	(ref)
	1000g-1499g	0.94	0.77-1.14
	1500g-1999g	**0.54**	**0.43-0.66***
	2000g-2499g	**0.32**	**0.26-0.39***
	2500g-3999g	**0.21**	**0.17-0.26***
	≥4000g	**0.20**	**0.16-0.24***
Gestational Age	< 28 weeks	(ref)	(ref)
	28-36 weeks	0.92	0.76-1.12
	37-41 weeks	**0.55**	**0.45-0.67***
	≥42 weeks	**0.54**	**0.44-0.67***
Predisposing Characteristics			
Baby's Sex	Male	(ref)	(ref)
	Female	**0.88**	**0.86-0.91***
Registered Indian Status	No	(ref)	(ref)
	Yes	**1.79**	**1.70-1.89***
Mother's Age Group	< 20	(ref)	(ref)
	20-34	**0.88**	**0.84-0.91***
	≥35	**0.86**	**0.81-0.92***
Parity	1	(ref)	(ref)
	2	**0.94**	**0.91-0.97***
	3	0.96	0.93-1.00
	4	1.01	0.96-1.06
	≥5	**1.21**	**1.14-1.28***
Enabling Characteristics			
RHA of Residence in Birth Year	Saskatoon	(ref)	(ref)
	Sun Country	0.99	0.91-1.08
	Five Hills	**1.15**	**1.07-1.24***
	Cypress	**1.24**	**1.12-1.37***
	Regina Qu'Appelle	**1.13**	**1.09-1.17***
	Sunrise	**1.28**	**1.19-1.38***
	Heartland	0.99	0.86-1.14
	Kelsey Trail	0.94	0.85-1.04
	PA Parkland	**0.92**	**0.86-0.98***
	Prairie North	0.98	0.92-1.06
	Northern Saskatchewan	0.86	0.74-1.01
Family Ever Received Income Assistance	No	(ref)	(ref)
	Yes	**1.21**	**1.18-1.25***
Child Ever Traveled Outside of Home RHA for Hospitalization	No	(ref)	(ref)
	Yes	**1.58**	**1.35-1.86***
Child Moved to Another RHA	No	(ref)	(ref)
	Yes	**0.88**	**0.83-0.92***

Interaction Terms			
RHA x Child Ever Traveled Outside of Home RHA for a Hospital Admission	**Saskatoon x Yes Travel**	**1.40**	**1.17-1.68***
	Sun Country x Yes Travel	0.97	0.79-1.18
	Five Hills x Yes Travel	1.05	0.86-1.28
	Cypress x Yes Travel	1.09	0.87-1.37
	Regina Qu'Appelle x Yes Travel	**1.31**	**1.10-1.57***
	Sunrise x Yes Travel	1.09	0.90-1.33
	Heartland x Yes Travel	**0.78**	**0.63-0.98***
	Kelsey Trail x Yes Travel	1.14	0.92-1.41
	PA Parkland x Yes Travel	**1.28**	**1.06-1.54***
	Prairie North x Yes Travel	**1.30**	**1.08-1.56***
	Northern Saskatchewan x Yes Travel	dropped	Dropped~
Congenital Anomaly x Registered Indian	**CA Yes x RI Yes**	**1.14**	**1.07-1.22***

*** Statistically Significant**

~ Dropped from the model due to collinearity

As illustrated in Table 4.14, again, children with multiple congenital anomalies (OR=2.09) spend more days in a hospital than other children and females have a shorter length of stay than males (OR=0.88). An inverse dose response relationship was observed between birth weight and length of stay; as birth weight increases, length of stay decreases; however, this relationship did not achieve statistical significance for babies who weighed between 1000g and 1499g at birth. Term and post-term infants have a shorter length of stay than very preterm infants (OR=0.55 and OR=0.54 respectively); however, there is no difference in length of stay for preterm and very preterm infants. A child moving to another RHA was protective in terms of length of stay (OR=0.88). Children whose mothers were between the ages of 20 and 34 at the time of their birth had a shorter length of stay than children who were born to teen mothers (OR=0.88), and children whose mothers were over 35 at the time of their birth also had a shorter LOS compared to children who were born to teen moms (OR=0.86). An overall dose-response relationship was observed between children's length of stay and mother's parity; however, only categories parity=2 (OR=0.94) and parity=5 or more

(OR=1.21) achieved statistical significance. Finally, children whose families ever received some sort of government income assistance had a longer length of stay in hospital than children whose families did not receive any income assistance (OR=1.21).

The presence of statistically significant interaction terms between RHA and the need to travel outside of one's home RHA for a hospital admission implies that neither of the terms can be examined in isolation. This means that when one looks at the impact of home RHA on the total length of stay in the first five years, it must be explained in the context of the need to travel outside of one's home RHA for a hospitalization (i.e. the relationship between home RHA and length of stay is different for children who had to travel outside of their home RHA to be hospitalized in the first five years of life versus children who did not have to travel outside of their home RHA to be hospitalized in the first five years of life). The same can also been said for the Registered Indian children with and without congenital anomalies – the relationship between Registered Indian status and length of stay is different for children with and without CAs.

CHAPTER FIVE: DISCUSSION AND CONCLUSIONS

This chapter begins by interpreting the results of the present study and discussing the practical implications of these results. This is followed by descriptions of the study strengths and limitations, and finally a discussion of future research that could be undertaken in this area.

5.1 Interpretation of Results

It was hypothesized that children with CAs would have a higher level of health care utilization than children without CAs in their first five years of life; and that this relationship would be significantly affected by a variety of factors related to need, predisposing characteristics and enabling characteristics of the children and necessarily of their caregivers. Furthermore, it was also hypothesized that a regional disparity in the level of health service utilization would be found.

Not surprisingly, the results indicate that with the exception of children who have CAs of the integument (ICD-9 757), children with congenital anomalies have a higher level of health care utilization than children without congenital anomalies. While CAs of the integument (i.e. the skin) can sometimes be serious it is unlikely that a CA in this category would constitute a major disability or require a great deal of close monitoring by a physician.

When one examines the predictive models for each type of health care utilization studied, one does indeed find that factors related to need (e.g. CA status, gestational age,

birth weight), as well as predisposing characteristics (e.g. sex, mother's age group, Registered Indian status, etc) and enabling characteristics (e.g. home RHA, travel outside of home RHA for services, etc) influence a child's use of health care services throughout their first five years of life.

While regional differences were found in the use of health care services for children with and without congenital anomalies, a regional difference in the overall prevalence of congenital anomalies was not found. Additionally, due to concerns regarding sample size it was not possible to do a regional comparison of use of health care services for children with specific congenital anomalies. The combination of these factors makes it impossible to assess the accuracy of the second hypothesis in this study.

5.1.1 Regional Differences in the Prevalence of Congenital Anomalies

Regional differences were seen in the prevalence of most specific congenital anomalies. This could be due to a variety of factors, such as:

- Differences due to maternal risk factors (e.g. rates of smoking during pregnancy, proportion of women taking preconceptional folic acid, maternal age) (6, 63).
- Differences in environmental exposures (6, 7, 63).
- Differences in the ethnic make-up of the health regions (i.e. Aboriginal people make up a larger percentage of the total population in Northern Saskatchewan than they do in the rest of the province) (7, 63).
- Differences in access to care and prenatal diagnostic services (63).
- Variation due to chance (63).

It is unlikely that all of the regional differences seen in the prevalence of CAs can be attributed to chance due to the large number of differences seen.

5.1.2 Study Question 1: Is the Level of Health Care Used by Children With Congenital Anomalies Significantly Different from the Level of Health Care Used by Children Without Congenital Anomalies?

When examining the broad groupings of children who have at least one congenital anomaly compared to children who do not have any congenital anomalies, children with CAs had a higher level of health care use for all three outcome variables. This is expected as need for care is the greatest predictor of use of health care services and children with CAs have a greater need for care than children without CAs (11).

This question was further broken down to determine if children with specific types of congenital anomalies had a higher level of health care utilization than children without that particular CA. With few exceptions, children with specific types of CAs also had a higher level of health care use. The one notable exception is for children affected by CAs of the integument (ICD-9 757) for whom there was no significant difference between them and children without CAs of the integument for all outcome variables. This is likely due to the fact that CAs of the integument (i.e. the skin) while sometimes serious in nature tend to not be life-threatening or cause major disabilities.

5.1.3 Study Question 2: Is There a Regional Difference in the Level of Health Care Used by Children in their First Five Years of Life? And Does This Relationship Hold For Children With and Without CAs?

Statistically significant regional differences were found for the three health care use outcome variables in this study population. Regional differences were found for all three outcome variables for children with and without CAs, but different patterns were seen based on CA status. More regional differences for all outcome variables were seen for children without CAs than for children with CAs. This may indicate that while children with the greatest need (i.e. those with CAs) are able to access appropriate care, those without CAs may have more difficulty accessing care.

The greatest differences in level of care between children with and without congenital anomalies is for length of stay. For children with CAs, on average, those in the northern parts of the province (Northern RHAs, Prairie North Regional Health Authority and PA Parkland Regional Health Authority) have statistically significant longer lengths of stay compared to those in the Saskatoon Health Region. Length of stay is also higher in the Kelsey Trail Regional Health Authority although this did not reach statistical significance. There are a few potential explanations for this such as differential access to outpatient services throughout the province, different hospital policies on length of stay and the severity and complexity of the condition for which care is sought. This study is not able to conclusively determine which explanation or which combination of explanations clarifies this difference. Individuals in these regions may have longer lengths of stay as they do not have as ready access to out-patient care in their region, these children may be sicker and require more time in hospital, and/or there might be different hospital policies on length of stay in these regions than their southern counterparts. As this same pattern is not seen for children without congenital anomalies, this difference is not likely due to hospital policies. While less access to outpatient care may necessitate the longer length of stay that is seen for children in the far north, the same cannot be said for children in the Prince Albert Parkland Regional Health Authority as there are a number of pediatricians practicing in Prince Albert.

As travel variables had been derived for use in the predictive models, this study was also able to examine regional differences in the need to travel outside of one's home RHA for hospital admissions or physician visits. With the exception of residents of the Regina Qu'Appelle health region for some of the specific CAs investigated, residents of

all other regions were significantly more likely to travel outside of their home RHA for physician visits and hospitalizations compared to children in the Saskatoon Health Region. The need to travel outside of one's home RHA for health care was significantly higher for children of almost all RHAs compared to children in the Saskatoon Health Region. This should not indicate a degree of acceptance of the current need to travel to obtain care by policy makers and the public. Health care and support services that are easily accessible allow people with disabilities to lead more independent and healthier lifestyles, and as this population of children grows up to be a population of adults with disabilities access becomes increasingly important (4).

The crude regional differences in use of health care services indicates that even with universal access to essential services in Saskatchewan, both regional differences in policies and availability of services may influence one's use of services (8). Furthermore, CA status may not be a predictor of differential use of health care services for regional health authorities in Saskatchewan. When the same regional difference is found for both children with and without CAs (i.e. as is seen for Northern Saskatchewan in Table 4.10 for all outcome variables) for a health outcome, this indicates that the reason for this difference is not related to CA status, but instead to a different factor that was not measured by this particular study.

5.1.4 Study Question 3: What Factors Influence the Level of Health Care Utilization?

As can be seen in Table 5.1, while many of the same variables predict different outcomes related to health care utilization, the predictive model for each outcome is different.

Table 5.1: Statistically significant variables in models for level of health care utilization

Variable	Model for Physician Visits	Model for Hospital Admissions	Model for Length of Stay
Main Effects			
Need Characteristics			
Congenital Anomaly	X	X	X
Multiple Congenital Anomalies	X	X	X
Birth Weight		X	X
Gestational Age			X
Predisposing Characteristics			
Baby's Sex	X	X	X
Registered Indian Status	X	X	X
Mother's Age Group		X	X
Mother's Marital Status		X	
Parity	X	X	X
Enabling Characteristics			
RHA of Residence in Birth Year	X	X	X
Family Ever Received Income Assistance	X	X	X
Child Ever Traveled Outside of Home RHA for Hospitalization or Physician Visit	X	X	X
Child Moved to Another RHA		X	X
Interaction Terms			
Home RHA x Child Ever Traveled Outside of Home RHA for a Hospitalization or Physician Visit	X		X
Congenital Anomaly x Registered Indian			X

According to Anderson and Newman's model of the individual determinants of health services utilization, three general categories of variables work together to determine what level of health care an individual will use: need characteristics (i.e. factors related to how ill one is), predisposing characteristics (i.e. intrinsic factors that make some individuals more likely than others to seek out treatment) and enabling characteristics (i.e. factors in one's environment that allow them to access services) (11, 41). All variables tested in the models for health care utilization were grouped according to these characteristics. As expected, congenital anomaly status and multiple congenital

anomaly status were significant in all three models. In addition, baby's sex, Registered Indian status, RHA of residence, receipt of income assistance and the need to travel outside of one's home RHA to access services were statistically significant in all three models.

As need for care is the most pressing determinant of use of health care services, it is not surprising to see that children with birth defects have more physician visits, more hospitalizations and longer lengths of stay than children without CAs. Research has shown that children with CAs require more and longer hospitalizations than children without CAs (66). Furthermore, as expected both gestational age and birth weight achieved statistical significance in the model for length of stay. The last few weeks of pregnancy are essential for the healthy development of children, and babies that are born too soon are at a higher risk of developing health problems immediately after birth (i.e. respiratory distress syndrome, intraventricular hemorrhage) and later in life (i.e. cerebral palsy) (67). The birth weight variable achieved statistical significance in the model for number of hospitalizations. This indicates that not only are low-birth weight babies more likely to stay in the hospital longer, they are also more likely to be re-hospitalized throughout their first five years of life compared to their normal birth weight peers. The same pattern was not seen for gestational age.

Infants are admitted to the hospital for a variety of reasons. Research conducted in Saskatchewan found that the leading causes of hospitalization for children in the first 28 days of life between 1989 and 1994 are: perinatal conditions (88%) [this includes jaundice (38%), asphyxia and hypoxia (20%), prematurity and low birth weight (16%)] and congenital anomalies (7%) (66). The leading causes of congenital anomaly related

hospitalizations for children under 10 were for CAs of the musculoskeletal system (25%), the heart and circulatory system (20%) and the digestive system (12%) (66).

While the overall health of the child is the greatest predictor in postnatal hospital readmissions, other factors seen in the literature that impact readmission are: mother's concerns regarding infant care, the amount of help a new mother receives at home following her initial discharge and whether the baby was seen by a health professional for a physical check-up after the initial hospital discharge (67). To the extent that this study was able to measure these characteristics, the findings are consistent with the literature as regions that had high number of physician visits for the most part also had low numbers of hospital admissions

In all three models, female children used less health care services than male children. This contradicts the general trends in the literature which show that females exhibit more health seeking behaviours than males and consequently use more health services (68). However, the literature has tended to focus on adult men and women who can choose to access or not access care, male and female children do not have that same level of autonomy. It is possible that this difference reflects a greater need for care for male children than female children in the study population.

Registered Indian children have more physician visits, more hospitalizations and longer lengths of stay than the general population of children in the study. This could also be a reflection of an increased need for services, since as a group, individuals with Registered Indian status suffer from poorer health than individuals in the general population (33).

Mother's age group reached statistical significance in the models for hospitalizations and length of stay and in both cases not being a teen mother was protective. This is consistent with other findings. The ability of parents to manage their child's care outside of a hospital setting and their ability to recognize when care is needed influences the number of hospitalizations for their children and the amount of time their children are required to spend in the hospital (46). This finding could also be due to the fact that teen mothers are more likely to have low birth weight babies than older mothers. Low birth weight babies require more hospitalizations and on average have longer lengths of stay (46). Similarly, a child moving to another RHA during the study period was protective in the models for hospital admissions and length of stay. This is most likely because parents tended to move their children to larger urban centres where more specialized care was available on an out-patient basis, thus reducing the need for hospitalizations and time in hospital.

Mother's marital status only attained statistical significance in the model for the total number of hospitalizations. While results are expected (children whose mothers were married or in a common law relationship had fewer hospitalizations than children born to single mothers), it is surprising that this variable did not reach significance in the other models, especially the model for the total number of physician visits. Mother's marital status acts as a proxy for social support which should, in theory, increase a mother's ability to obtain care for her child, and as she seeks out more physician visits for her child, her child would, in turn, require fewer hospitalizations.

Somewhat surprisingly, parity was the only maternal variable to achieve statistical significance in all three models; however, not unexpectedly, while a dose-

response relationship was observed in all models, an inverse relationship was seen for physician visits and hospitalizations/length of stay. As the number of children a woman has increases, the number of physician visits per child decreases. This could be reflective of the increasing demands on her time brought about by having additional children to care for and a lack of social support. As the number of children a woman has increases, the number of hospitalizations per child and the length of stay per hospitalization increases. Both of these factors go hand-in-hand with the pattern seen for the relationship between maternal parity and number of physician visits for her children. As women have more children they may have less time on their hands to focus on preventative care, consequently, these children require more hospitalizations (it is unknown whether the rate of ambulatory case sensitive [ACS] conditions also increase with maternal parity).

Children whose families ever received some sort of governmental income assistance during the study period had higher numbers of physician visits, hospitalizations and longer lengths of stay. This is consistent with other studies as the literature indicates that children who live in low-income families tend to experience more chronic health problems and developmental disabilities (66). This results in a greater need for health care services than is experienced by children from higher income families.

Of the six interaction terms tested in the models, only two achieved statistical significance in any of the models. No interaction terms were significant in the model for hospitalizations. Children's health region of residence and traveling outside of the region showed a significant interaction for the outcomes related to length of stay and

total number of physician visits. Children's congenital anomaly status and Registered Indian status also showed a significant interaction for the length of stay outcome variable. By examining the interaction between a child's home RHA and the need for a child to travel outside of one's home RHA for a physician visit in the model for physician visits, one can see that, with the exception of children in Northern Saskatchewan, children who have to travel outside of their home RHA to see a physician, have fewer physician visits than children who do not have to travel outside of their home RHA. A different pattern is seen when examining this interaction term in the length of stay model. Children who live in the Saskatoon Health Region, the Regina Qu'Appelle Health Region, the Prince Albert Parkland Health Region and the Prairie North Health Region and need to travel outside of their home RHA for a hospital admission, have longer lengths of stay than children who live in these same health regions who do not need to travel. This is could be an indication of need. The opposite pattern is seen for children who live in the Heartland Health Region – children who live in the Heartland Health Region and need to travel outside of their home RHA for a hospitalization have shorter lengths of stay than children who do not have to travel outside of the Heartland Health Region for a hospitalization. This could reflect different regional policies surrounding length of stay. Larger regions that can offer more specialized care could be more likely to have stricter policies on length of stay as they have a greater need for beds, and children in these regions have easier access to follow-up care on an outpatient basis.

The interaction term between congenital anomaly status and Registered Indian status acts synergistically in the length of stay model indicating that the relationship

between Registered Indian status and length of stay is different for children with and without CAs. Children with Registered Indian status and congenital anomalies had longer lengths of stay than children with congenital anomalies but without Registered Indian status. This is expected as the same pattern is seen for the constituent parts of this interaction term in all three models (i.e. Registered Indian status children use significantly more health care than the general population of children and children with CAs use significantly more health care than children without CAs).

What's particularly important about examining the models that predict a child's use of various health care services in the first five years of life; is that after adjustment for need characteristics, predisposing factors and enabling factors, a different pattern of regional differences emerges than in the previous question when one was merely examining crude differences. These adjusted differences are far more meaningful as they eliminate many differences between regions that could account for the differential use of health care.

5.2 Practical Implications of Results and Directions for Future Research

In order to prevent CAs, it is first essential to know what the baseline measures are for the prevalence of specific CAs in a population and the dispersion of CAs within that population (7, 69). Once baseline measures have been established prevention programs can be developed for both the principal condition as well as any associated co-morbidities that can reduce the quality of life for affected individuals (69). This is the first study of its kind in Saskatchewan to examine the regional differences in rates of all categories of CAs. This study found that regional differences do exist in rates of certain types of CAs. These differences in and of themselves require additional studies with a

finer level of detail for the definition of these types of CAs to determine exactly what regional differences exist. Furthermore, future studies need information on rates of prenatal testing for certain conditions to determine whether we are seeing a true regional difference in rates of CAs or merely a regional difference in the live birth rate for CAs. These studies also require information on whether or not these conditions are genetically induced or environmentally induced to determine if these regional differences could potentially be prevented. While more information is necessary to fully assess the extent of these regional differences, the prevalence estimates from the current study are important to plan for both the special educational and the health care needs of this population (63). To continually assess the regional differences in rates of CAs, there is a need for the routine monitoring of CAs, such as seen in other provinces with congenital anomalies registries, to track regional disparities over time and monitor trends in real time.

Regional differences were also found to exist for all outcome variables related to the use of health care services and in travel for health care services. These regional differences need to be further examined by Saskatchewan Health as it may indicate that residents are receiving different levels of care based on where they live in the province. Future studies are needed to also look at regional differences in access to care within health regions. As hospital care is more costly than ambulatory care, regional differences in hospitalizations should be closely examined to see why regional differences exist and what can be done about them (i.e. increased numbers of nurse practitioners and physicians in rural/remote areas, increased focus on the prevention of disease, etc).

While this study was able to determine that regional differences do in fact exist for rates of congenital anomalies and use of health care services, it was not able to answer the more meaningful questions "Are these differences also inequities?" and "Are the differences seen as unfair?" Future studies that involve both quantitative and qualitative methodologies will be required to provide a clear answer to this question.

5.3 Study Strengths

While this study has shortcomings, there are strengths associated with the study design and methodology. Foremost, this is a longitudinal cohort study, which is one of the most robust epidemiological study designs. As cohort studies begin with an exposed and unexposed group and follow the groups over time to assess an independent outcome variable, cohort designs are free from many biases that can plague case-control and cross-sectional studies. The second major strength to this study is its large sample size which increases the likelihood that the sample population will be representative of the general population.

An additional strength of this study is the five year ascertainment period for congenital anomalies. While all congenital anomalies are present at birth, many of them are not diagnosed until much later (1, 2, 12, 13). The longer ascertainment period is especially important for this particular study as the diagnosis of a CA at any point in time could result in a higher level of health care services around the time of diagnosis. If this had not been accounted for it could have resulted in an overestimate of the level of health care services for children 'without' CAs. Along this same line, using separate outcome variables (physician visits, hospital admissions and length of stay) is a study

strength, as it provides a more complete picture of the factors that predict the use of multiple health care services.

Finally, using administrative data strengthens this study because this data was not collected with a specific hypothesis in mind, it is free of certain types of bias that are common in studies that rely on survey data such as recall bias and selection bias.

5.4 Study Limitations

This study has certain limitations, and the results of the study must be interpreted with these in mind. The administrative categorization of congenital anomaly status was large and encompassed a wide range of conditions and therefore it was not possible to determine the possible causes of specific conditions. Also, it was not possible to ascertain the severity of a specific anomaly, which limits the practicality of this study in health care planning (63). It is unknown in what direction this may bias the results. While the adaptation of the ICD-10 system will partially address the issue related to the categorization of CAs in future studies, it was not possible to use ICD-10 coding for this study as the coding changed part-way through the study period. It was not possible to reclassify the ICD-9 codes into ICD-10 codes based on the available data. Additionally, it is not known who made the diagnosis of a CA (i.e. a family physician versus a specialist). This may have caused some normal variations to be classified as CAs which would bias the results towards the null if 'healthy' people were erroneously classified as 'diseased'.

In addition to the definition of CA status, there are also limitations associated with only using Registered Indian status to differentiate between those of Aboriginal ancestry and those who do not self-identify as Aboriginal. The term Aboriginal is an

umbrella term that includes all people of Aboriginal origin (First Nations, Métis and Inuit) (33). Within this group, people of First Nations or Inuit ancestry may or may not be registered under the Indian Act (there is not a similar piece of registration for the Métis) (33). The Indian Registry is a national database of all Registered First Nations who are eligible to receive benefits from the First Nations and Inuit Health Branch (FNIHB)'s Non-Insured Services (32). As not all people who self-declare as Aboriginal are registered under the Indian Act (and Métis people cannot be registered under this act), any definition of 'Aboriginal' that is limited to Registered Indians is an underestimate. A study conducted in Manitoba attempted to more completely ascertain the number of First Nations people living in Manitoba than was identified in the Manitoba Health Registry alone (a database that includes all Manitoba residents who receive universal health insurance) (32). After combining data from the Medical Services Branch of FNIHB, from Manitoba Health and from the Manitoba Health Registry (as maintained by the Manitoba Centre for Health Policy), they found that while there was an increase in every age group, most groups gained approximately 25% more individuals (32). Additionally, while the researchers did not feel that there was a geographic pattern to these increases, the largest geographical increase was in Registered First Nations people living off reserve (32). While it is not known if similar increases would be seen with a comparison of data from Saskatchewan Health and FNIHB, it is probable. As this study was only using Registered Indian status as a measure of Aboriginals in Saskatchewan, it is likely that this underestimates the difference between Aboriginals and non-Aboriginals for all outcome variables.

Another limitation associated with the use of administrative data in this particular study, is only obtaining information on physician visits from the medical services database, as this does not capture all ambulatory care visits. The information in this database is based on fee-for-service physicians' payment claims (61). While physicians under an alternative payment plan have the option of shadow billing, this is not mandatory; additionally, ambulatory care provided by nurse practitioners is not captured in this database. This will result in an underestimate in the number of physician visits throughout the province, but this underestimate will be greatest in the north where there are fewer physicians, and the majority of the day-to-day care is provided by nurses. Moreover, there is also no information on the type of physician (i.e. specialists versus general practitioner) that a child saw, which could influence the number of visits required or on the length of stay policies in the various hospitals. This could also influence the amount of time children spent in the hospital based on where they lived.

Another limitation associated with using administrative data is that it does not allow researchers to account for some potential confounders such as maternal education level, etiology of the congenital anomaly (i.e. genetically induced versus environmentally induced), use of non-traditional health care services, etc – all of which might have influenced the outcome variables.

Another limitation of this study is the use of a static reference group. While selecting the Saskatoon Health Region as the reference group eased the data analysis and interpretation, the use of one reference group does not provide a true picture of all the regional differences in the prevalence of congenital anomalies or the level of health

care utilization. When calculating the prevalence of congenital anomalies, it is possible that these rates are affected by an ascertainment bias as some conditions may be more or less likely to be diagnosed prenatally in certain regions than others, and this could have influenced the live birth rate. Also, due to small sample sizes, it was not possible to do region specific and congenital anomaly specific analyses.

5.5 Conclusions

While a number of variables (both outcome and predictor) were examined in the course of this study, this study is really only one of many that can, and should, be done in this area. This study was able to describe what is currently happening, but it was not able to explain why things were currently happening, or what could be done to change the current situation.

This study was able to conclusively show that as expected children with CAs use a higher level of health care services (physician visits, hospitalizations and length of stay) than children without CAs in the first five years of life; and that regional differences are present for the total population and for children with and without CAs. When examining the factors that predict a child's use of health care services, region of residence remained a significant predictor of all outcome variables even after adjusting for a variety of factors related to a child's need for health care, and the factors that predispose and enable a child and his or her mother to access care. This significant finding indicates that there is a need for further studies in this area and closer real-time monitoring of children with CAs in Saskatchewan to ensure that this vulnerable population is receiving the best (and most timely and accessible) care possible.

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APPENDIX A: ICD-9 AND ICD-10 CODES FOR CONGENITAL ANOMALIES

A description of all the ICD-9 codes for congenital anomalies found in the study population can be found in Table A-1. Due to small sample sizes, the children with either ICD-9 740 and/or ICD-9 741 were grouped together into the new category ICD-9 740+741. Also due to small sample sizes, all ICD10 codes found in the study population were recoded to ICD-9 codes, please see Table A-2 for a description of how ICD10 codes were reclassified.

Table A.1: ICD-9 and MSB codes for congenital anomalies found in the study population

ICD-9 Codes	Description
740 + 741	**Neural Tube Defects** Includes: 740 Anencephalus and similar anomalies 740.0 Anencephalus 740.1 Craniorachischisis 740.2 Iniencephaly 741 Spina bifida 741.0 With hydrocephalus 741.9 Without mention of hydrocephalus
742	**Other congenital anomalies of nervous system** Includes: 742.0 Encephalocele 742.1 Microcephalus 742.2 Reduction deformities of brain 742.3 Congenital hydrocephalus 742.4 Other specified anomalies of brain 742.5 Other specified anomalies of spinal cord 742.8 Other specified anomalies of nervous system 742.9 Unspecified anomaly of brain, spinal cord, and nervous system
743	**Congenital anomalies of eye** Includes: 743.0 Anophthalmos 743.1 Microphthalmos 743.2 Buphthalmos 743.3 Congenital cataract and lens anomalies 743.4 Coloboma and other anomalies of anterior segment 743.5 Congenital anomalies of posterior segment

	743.6 Congenital anomalies of eyelids, lacrimal system, and orbit 743.8 Other specified anomalies of eye 743.9 Unspecified anomaly of eye
744	**Congenital anomalies of ear, face, and neck** Includes: 744.0 Anomalies of ear causing impairment of hearing 744.1 Accessory auricle 744.2 Other specified anomalies of ear 744.3 Unspecified anomaly of ear 744.4 Branchial cleft cyst or fistula; preauricular sinus 744.5 Webbing of neck 744.8 Other specified anomalies of face and neck 744.9 Unspecified anomalies of face and neck
745	**Bulbus cordis anomalies and anomalies of cardiac septal closure** Includes: 745.0 Common truncus 745.1 Transposition of great vessels 745.2 Tetralogy of Fallot 745.3 Common ventricle 745.4 Ventricular septal defect 745.5 Ostium secundum type atrial septal defect 745.6 Endocardial cushion defects 745.7 Cor biloculare 745.8 Other 745.9 Unspecified defect of septal closure
746	**Other congenital anomalies of heart** Includes: 746.0 Anomalies of pulmonary valve 746.1 Tricuspid atresia and stenosis, congenital 746.2 Ebstein's anomaly 746.3 Congenital stenosis of aortic valve 746.4 Congenital insufficiency of aortic valve 746.5 Congenital mitral stenosis 746.6 Congenital mitral insufficiency 746.7 Hypoplastic left heart syndrome 746.8 Other specified anomalies of heart 746.9 Unspecified anomaly of heart
747	**Other congenital anomalies of circulatory system** Includes: 747.0 Patent ductus arteriosus 747.1 Coarctation of aorta 747.2 Other anomalies of aorta 747.3 Anomalies of pulmonary artery 747.4 Anomalies of great veins 747.5 Absence or hypoplasia of umbilical artery 747.6 Other anomalies of peripheral vascular system

	747.8 Other specified anomalies of circulatory system
748	**Congenital anomalies of respiratory system** Includes: 748.0 Choanal atresia 748.1 Other anomalies of nose 748.2 Web of larynx 748.3 Other anomalies of larynx, trachea, and bronchus 748.4 Congenital cystic lung 748.5 Agenesis, hypoplasia, and dysplasia of lung 748.6 Other anomalies of lung 748.8 Other specified anomalies of respiratory system 748.9 Unspecified anomaly of respiratory system
749	**Cleft palate and cleft lip** Includes: 749.0 Cleft palate 749.1 Cleft lip 749.2 Cleft palate with cleft lip
750	**Other congenital anomalies of upper alimentary tract** Includes: 750.0 Tongue tie 750.1 Other anomalies of tongue 750.2 Other specified anomalies of mouth and pharynx 750.3 Tracheoesophageal fistula, esophageal atresia and stenosis 750.4 Other specified anomalies of esophagus 750.5 Congenital hypertrophic pyloric stenosis 750.6 Congenital hiatus hernia 750.7 Other specified anomalies of stomach 750.8 Other specified anomalies of upper alimentary tract 750.9 Unspecified anomaly of upper alimentary tract
751	**Other congenital anomalies of digestive system** Includes: 751.0 Meckel's diverticulum 751.1 Atresia and stenosis of small intestine 751.2 Atresia and stenosis of large intestine, rectum, and anal canal 751.3 Hirschsprung's disease and other congenital functional disorders of colon 751.4 Anomalies of intestinal fixation 751.5 Other anomalies of intestine 751.6 Anomalies of gallbladder, bile ducts, and liver 751.7 Anomalies of pancreas 751.8 Other specified anomalies of digestive system 751.9 Unspecified anomaly of digestive system
752	**Congenital anomalies of genital organs** Includes: 752.0 Anomalies of ovaries 752.1 Anomalies of fallopian tubes and broad ligaments

	752.2 Doubling of uterus 752.3 Other anomalies of uterus 752.4 Anomalies of cervix, vagina, and external female genitalia 752.5 Undescended and retractile testicle 752.6 Hypospadias and epispadias and other penile anomalies 752.7 Indeterminate sex and pseudohermaphroditism 752.8 Other specified anomalies of genital organs 752.9 Unspecified anomaly of genital organs
753	**Congenital anomalies of urinary system** Includes: 753.0 Renal agenesis and dysgenesis 753.1 Cystic kidney disease 753.2 Obstructive defects of renal pelvis and ureter 753.3 Other specified anomalies of kidney 753.4 Other specified anomalies of ureter 753.5 Exstrophy of urinary bladder 753.6 Atresia and stenosis of urethra and bladder neck 753.7 Anomalies of urachus 753.8 Other specified anomalies of bladder and urethra 753.9 Unspecified anomaly of urinary system
754	**Certain congenital musculoskeletal deformities** Includes: 754.0 Of skull, face, and jaw 754.1 Of sternocleidomastoid muscle 754.2 Of spine 754.3 Congenital dislocation of hip 754.4 Congenital genu recurvatum and bowing of long bones of leg 754.5 Varus deformities of feet 754.6 Valgus deformities of feet 754.7 Other deformities of feet 754.8 Other specified nonteratogenic anomalies
755	**Other congenital anomalies of limbs** Includes: 755.0 Polydactyly 755.1 Syndactyly 755.2 Reduction deformities of upper limb 755.3 Reduction deformities of lower limb 755.4 Reduction deformities, unspecified limb 755.5 Other anomalies of upper limb, including shoulder girdle 755.6 Other anomalies of lower limb, including pelvic girdle 755.8 Other specified anomalies of unspecified limb 755.9 Unspecified anomaly of unspecified limb
756	**Other congenital musculoskeletal anomalies** Includes: 756.0 Anomalies of skull and face bones 756.1 Anomalies of spine

	756.2 Cervical rib 756.3 Other anomalies of ribs and sternum 756.4 Chondrodystrophy 756.5 Osteodystrophies 756.6 Anomalies of diaphragm 756.7 Anomalies of abdominal wall 756.8 Other specified anomalies of muscle, tendon, fascia, and connective tissue 756.9 Other and unspecified anomalies of musculoskeletal system
757	**Congenital anomalies of the integument** Includes: 757.0 Hereditary edema of legs 757.1 Ichthyosis congenital 757.2 Dermatoglyphic anomalies 757.3 Other specified anomalies of skin 757.4 Specified anomalies of hair 757.5 Specified anomalies of nails 757.6 Specified anomalies of breast 757.8 Other specified anomalies of the integument 757.9 Unspecified anomaly of the integument
758	**Chromosomal anomalies** Includes: 758.0 Down syndrome 758.1 Patau's syndrome 758.2 Edward's syndrome 758.3 Autosomal deletion syndromes 758.4 Balanced autosomal translocation in normal individual 758.5 Other conditions due to autosomal anomalies 758.6 Gonadal dysgenesis 758.7 Klinefelter's syndrome 758.8 Other conditions due to chromosome anomalies 758.9 Conditions due to anomaly of unspecified chromosome
759	**Other and unspecified congenital anomalies** Includes: 759.0 Anomalies of spleen 759.1 Anomalies of adrenal gland 759.2 Anomalies of other endocrine glands 759.3 Situs inversus 759.4 Conjoined twins 759.5 Tuberous sclerosis 759.6 Other hamartoses, NEC 759.7 Multiple congenital anomalies, so described 759.8 Other specified anomalies 759.9 Congenital anomaly, unspecified
MSB Z60	**Congenital dysplasia of hip**
MSB Z61	**Clubfoot**

Table A.2: Reclassification of ICD-10 codes found in the study population to ICD-9 codes

ICD-10 Code	Description	Recoded to ICD-9
	Q00-Q07: Congenital malformations of the nervous system	
Q00	Anencephaly and similar malformations	(740)
Q02	Microcephaly	(742)
Q03	Congenital hydrocephalus	(742)
Q04	Other congenital malformations of brain	(742)
Q05	Spina bifida	(741)
Q06	Other congenital malformations of spinal cord	(742)
Q07	Other congenital malformations of nervous system	(742)
	Q10-Q18: Congenital malformations of eye, ear, face and neck	
Q10	Congenital malformations of eyelid, lacrimal apparatus and orbit	(743)
Q11	Anophthalmos, microphthalmos and macrophthalmos	(743)
Q17	Other congenital malformations of ear	(744)
Q18	Other congenital malformations of face and neck	(744)
	Q20-Q28: Congenital malformations of the circulatory system	
Q20	Congenital malformations of cardiac chambers and connections	(746)
Q21	Congenital malformations of cardiac septa	(745)
Q22	Congenital malformations of pulmonary and tricuspid valves	(747)
Q23	Congenital malformations of aortic and mitral valves	(746)
Q24	Other congenital malformations of heart	(746)
Q25	Congenital malformations of great arteries	(747)
Q27	Other congenital malformations of peripheral vascular system	(747)
Q28	Other congenital malformations of circulatory system	(747)
	Q30-Q34: Congenital malformations of the respiratory system	
Q30	Congenital malformations of nose	(748)
	Q35-Q37: Cleft lip and cleft palate	
Q35	Cleft palate	(749)
Q37	Cleft palate with cleft lip	(749)
	Q38-Q45: Other congenital malformations of the digestive system	
Q38	Other congenital malformations of tongue, mouth and pharynx	(750)
Q39	Congenital malformations of oesophagus	(750)
Q43	Other congenital malformations of intestine	(759)
	Q50-Q56: Congenital malformations of genital organs	
Q52	Other congenital malformations of female genitalia	(752)
Q53	Undescended testicle	(752)
Q54	Hypospadias	(752)

Q55	Other congenital malformations of male genital organs	(752)
	Q60-Q64: Congenital malformations of the urinary system	
Q61	Cystic kidney disease	(753)
Q62	Congenital obstructive defects of renal pelvis and congenital malformations of ureter	(753)
	Q65-Q79: Congenital malformations and deformations of the musculoskeletal system	
Q66	Congenital deformities of feet	(754)
Q68	Other congenital musculoskeletal deformities	(754)
Q69	Polydactyly	(755)
Q70	Syndactyly	(755)
Q74	Other congenital malformations of limb(s)	(755)
Q75	Other congenital malformations of skull and face bones	(756)
Q76	Congenital malformations of spine and bony thorax	(756)
Q78	Other osteochondrodysplasias	(756)
Q79	Congenital malformations of the musculoskeletal system, not elsewhere classified	(756)
	Q80-Q89: Other congenital malformations	
Q82	Other congenital malformations of skin	(757)
Q86	Congenital malformation syndromes due to known exogenous causes, not elsewhere classified	(759)
Q87	Other specified congenital malformation syndromes affecting multiple systems	(759)
Q89	Other congenital malformations, not elsewhere classified	(759)
	Q90-Q99: Chromosomal abnormalities, not elsewhere classified	
Q90	Down syndrome	(758)
Q91	Edwards' syndrome and Patau's syndrome	(758)
Q93	Monosomies and deletions from the autosomes, not elsewhere classified	(758)
Q96	Turner's syndrome	(758)

APPENDIX B: REGIONAL DIFFERENCES IN THE MEAN NUMBER OF CASES OF CONGENITAL ANOMALIES BY REGIONAL HEALTH AUTHORITY

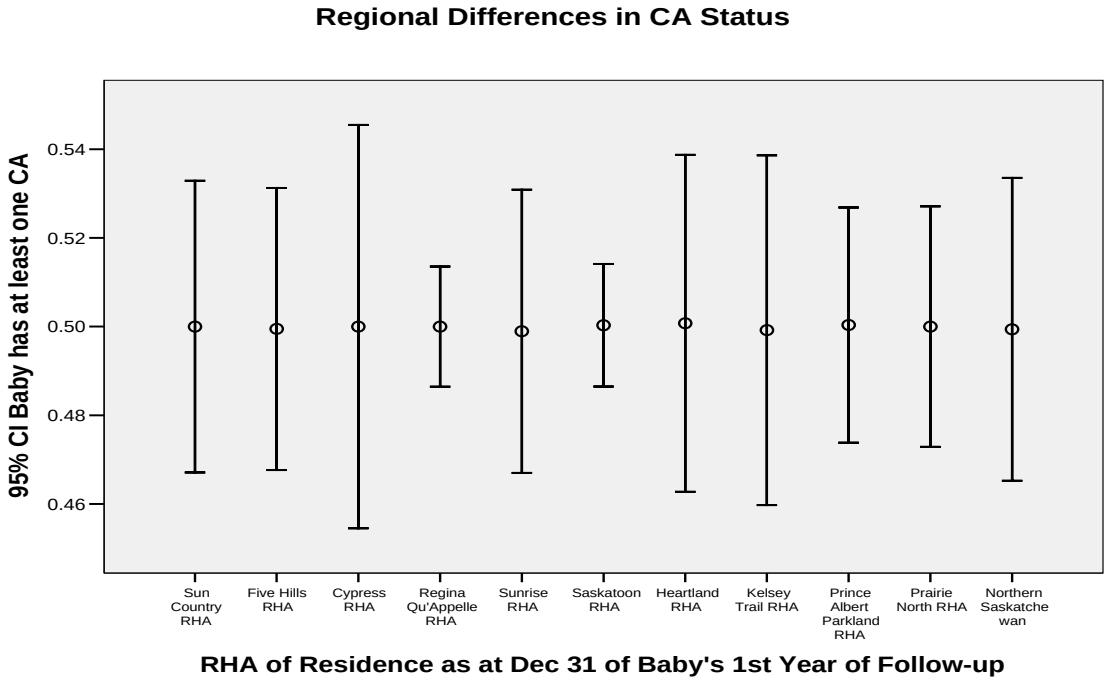

Figure B.1: Regional differences in the mean number of cases of children with CAs

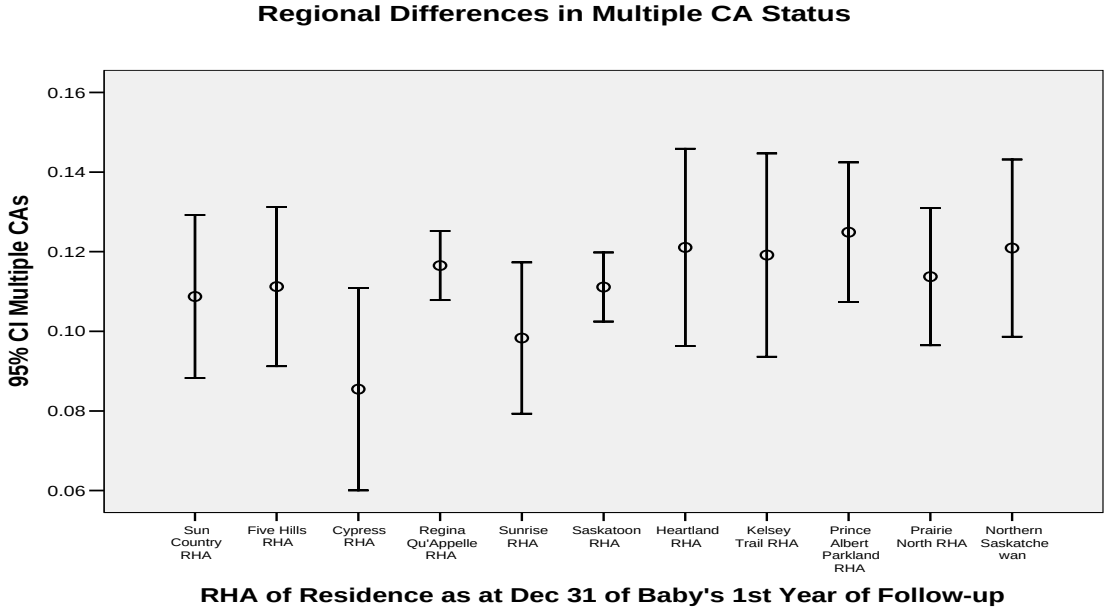

Figure B.2: Regional differences in the mean number of cases of children with multiple CAs

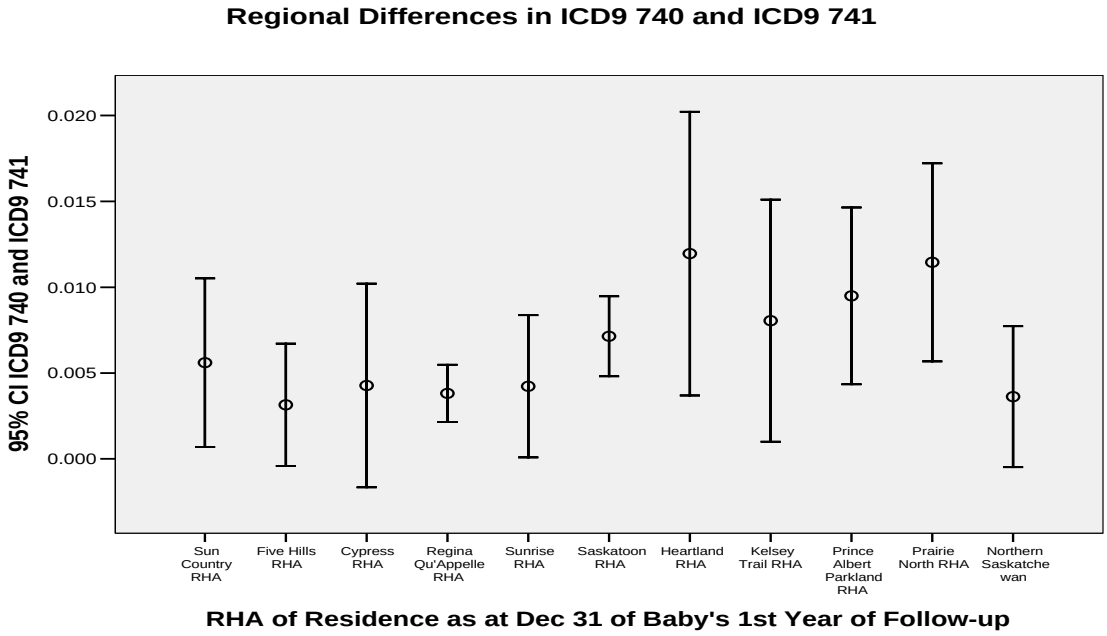

Figure B.3: Regional differences in the mean number of cases of children with neural tube defects (ICD-9 740 and 741)

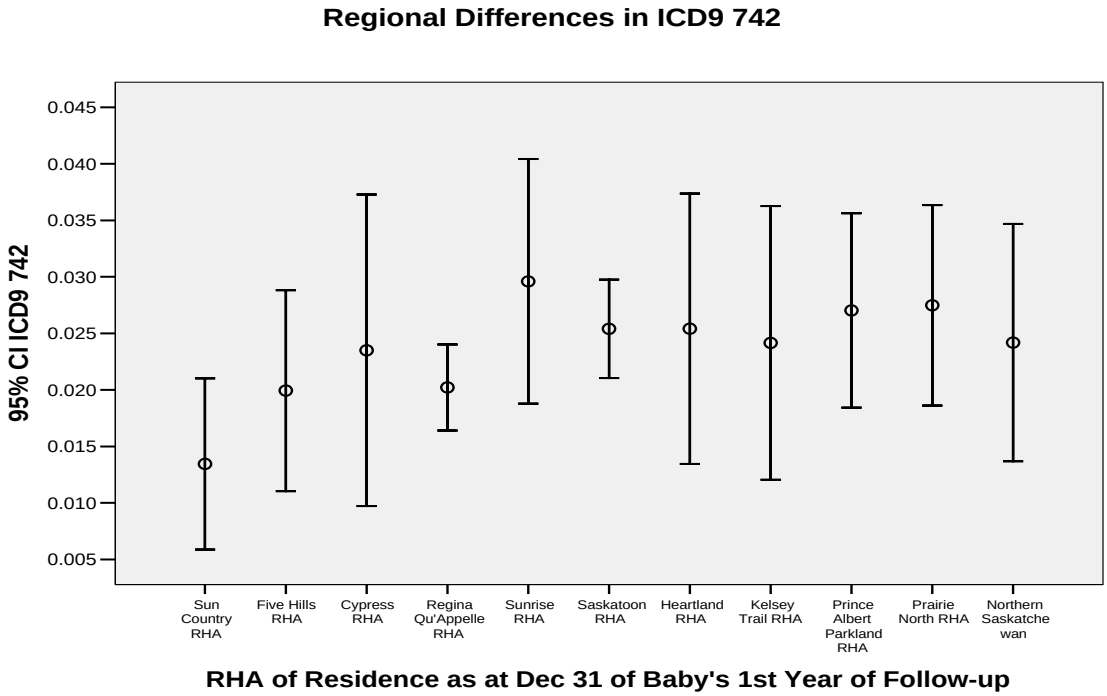

Figure B.4: Regional differences in the mean number of cases of children with other congenital anomalies of nervous system (ICD-9 742)

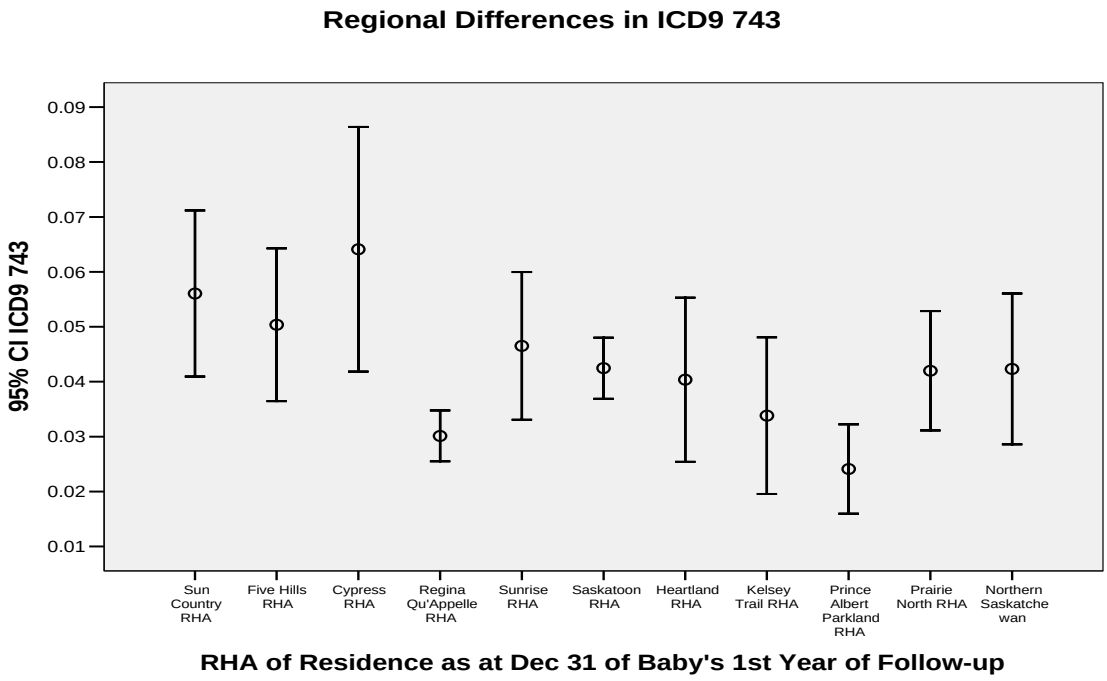

Figure B.5: Regional differences in the mean number of cases of children with congenital anomalies of eye (ICD-9 743)

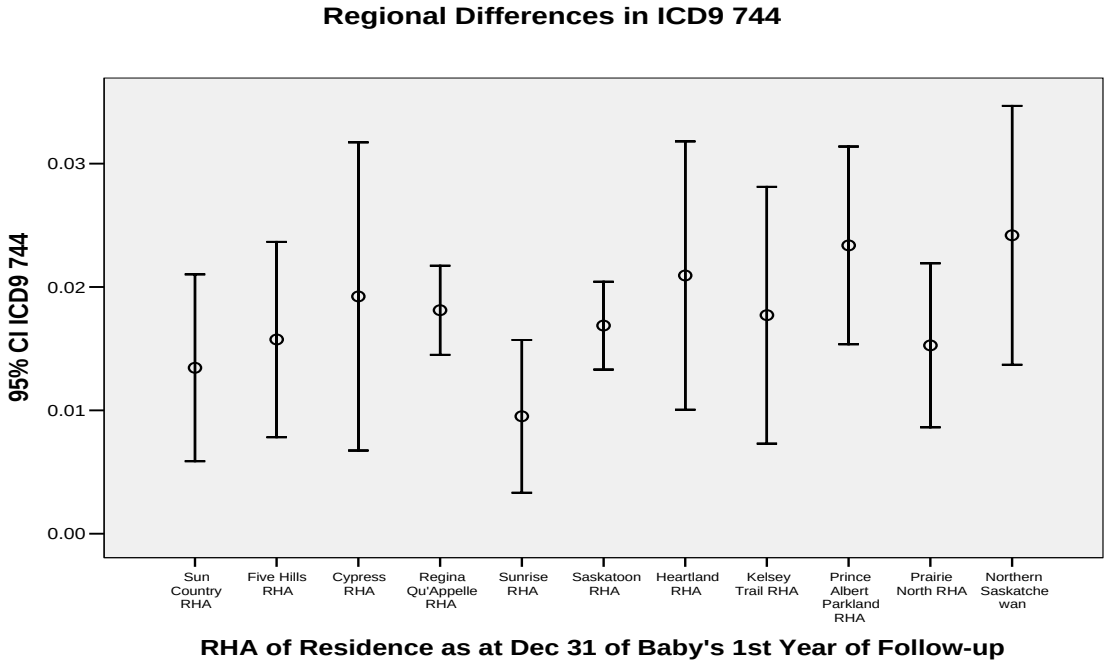

Figure B.6: Regional differences in the mean number of cases of children with congenital anomalies of ear, face, and neck (ICD-9 744)

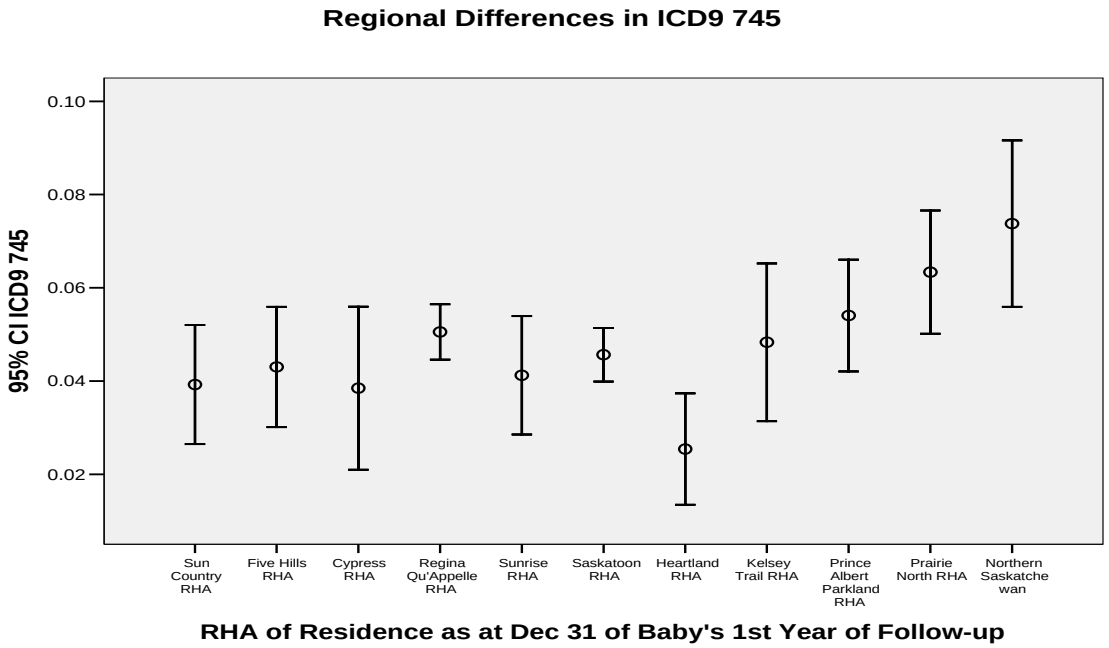

Figure B.7: Regional differences in the mean number of cases of children with bulbus cordis anomalies and anomalies of cardiac septal closure (ICD-9 745)

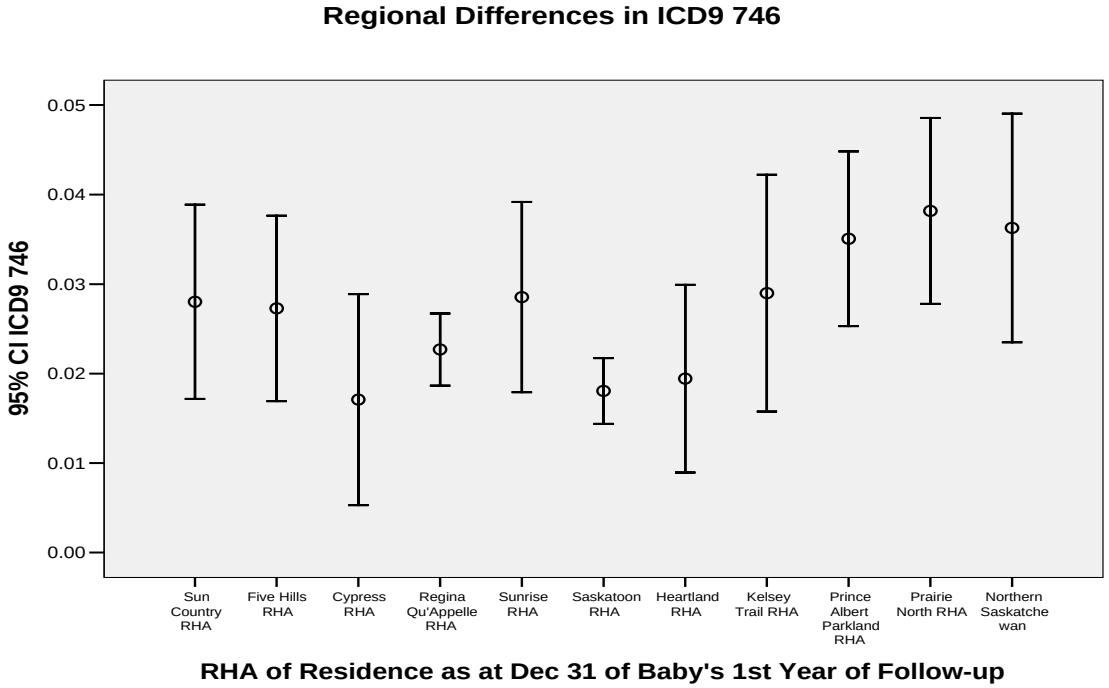

Figure B.8: Regional differences in the mean number of cases of children with other congenital anomalies of heart (ICD-9 746)

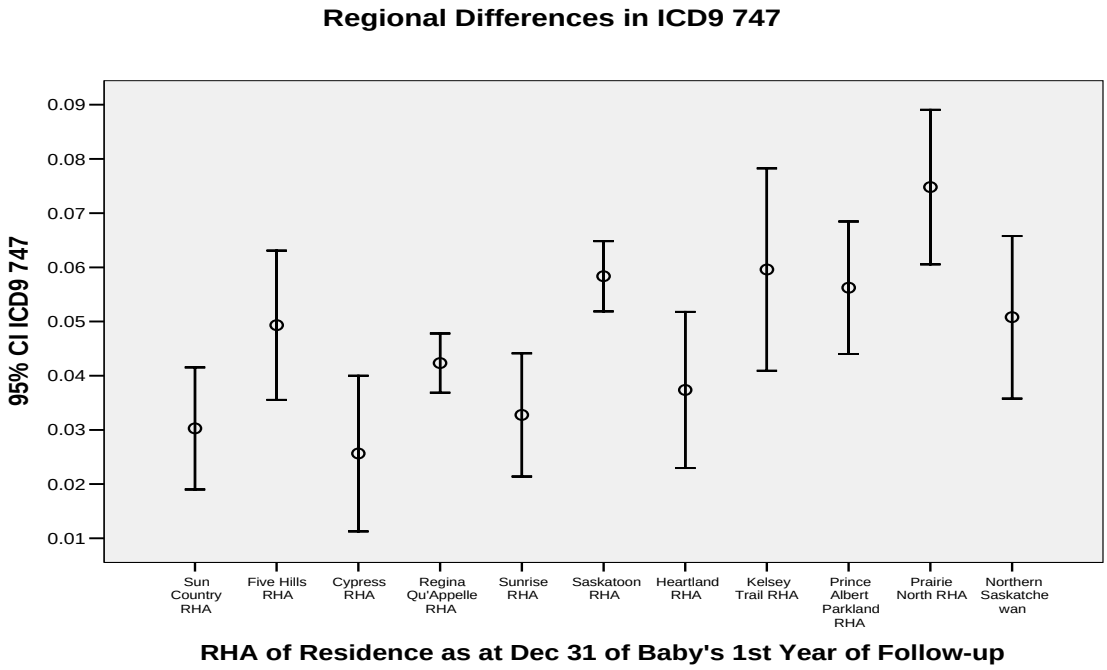

Figure B.9: Regional differences in the mean number of cases of children with other congenital anomalies of circulatory system (ICD-9 747)

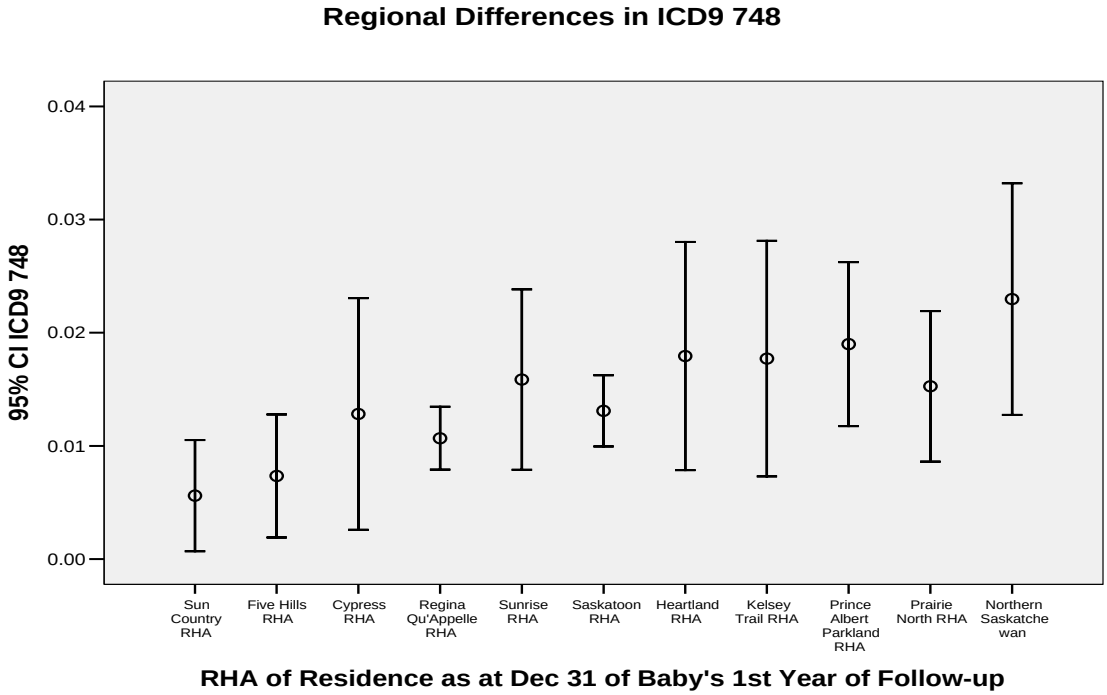

Figure B.10: Regional differences in the mean number of cases of children with congenital anomalies of respiratory system (ICD-9 748)

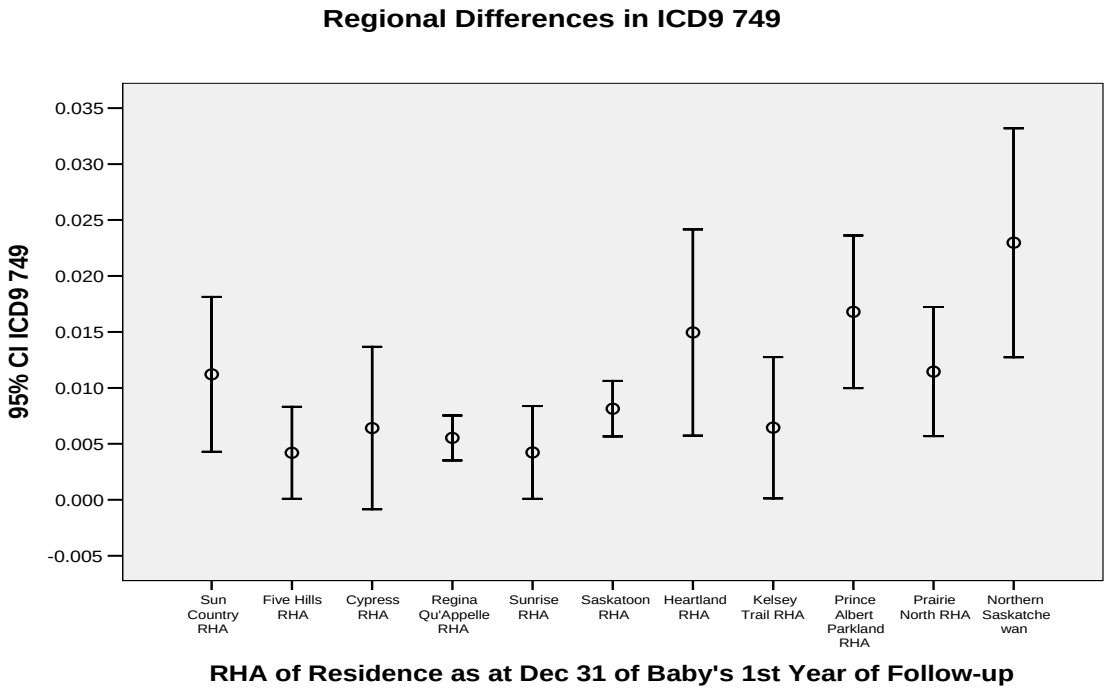

Figure B.11: Regional differences in the mean number of cases of children with cleft palate and cleft lip (ICD-9 749)

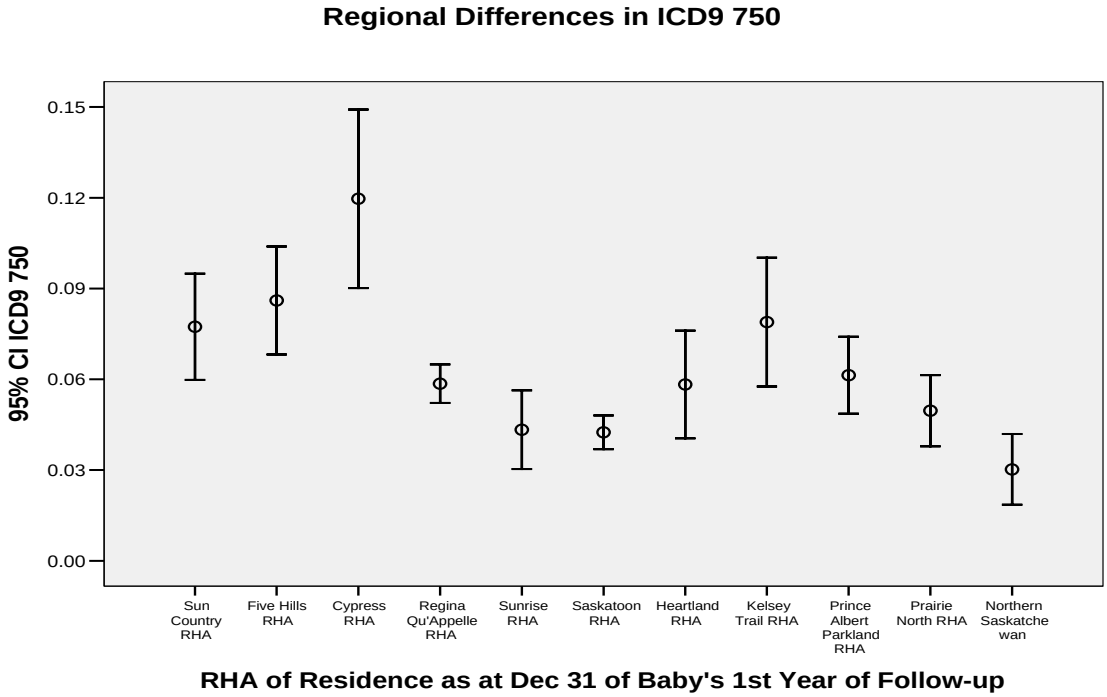

Figure B.12: Regional differences in the mean number of cases of children with other congenital anomalies of upper alimentary tract (ICD-9 750)

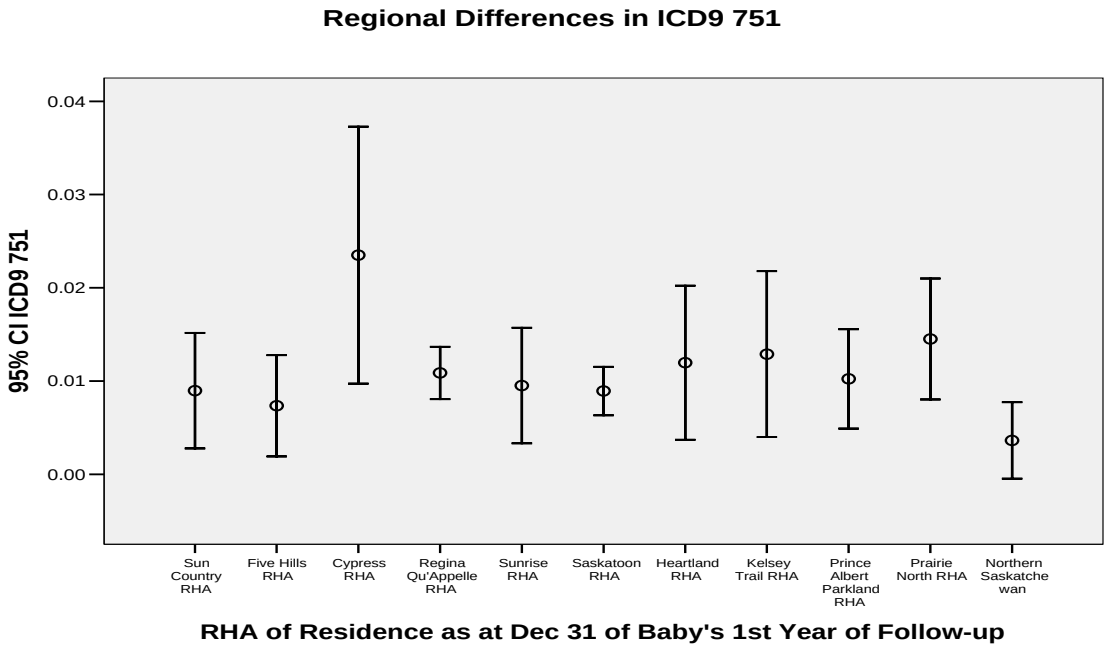

Figure B.13: Regional differences in the mean number of cases of children with other congenital anomalies of digestive system (ICD-9 751)

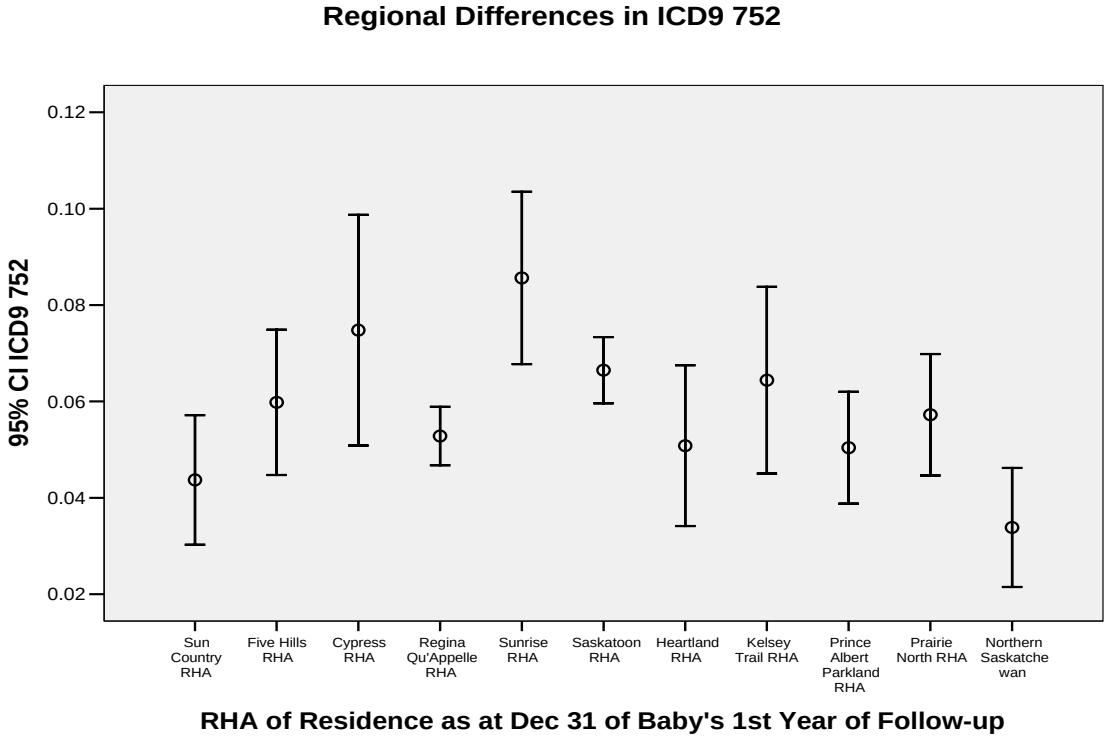

Figure B.14: Regional differences in the mean number of cases of children with congenital anomalies of genital organs (ICD-9 752)

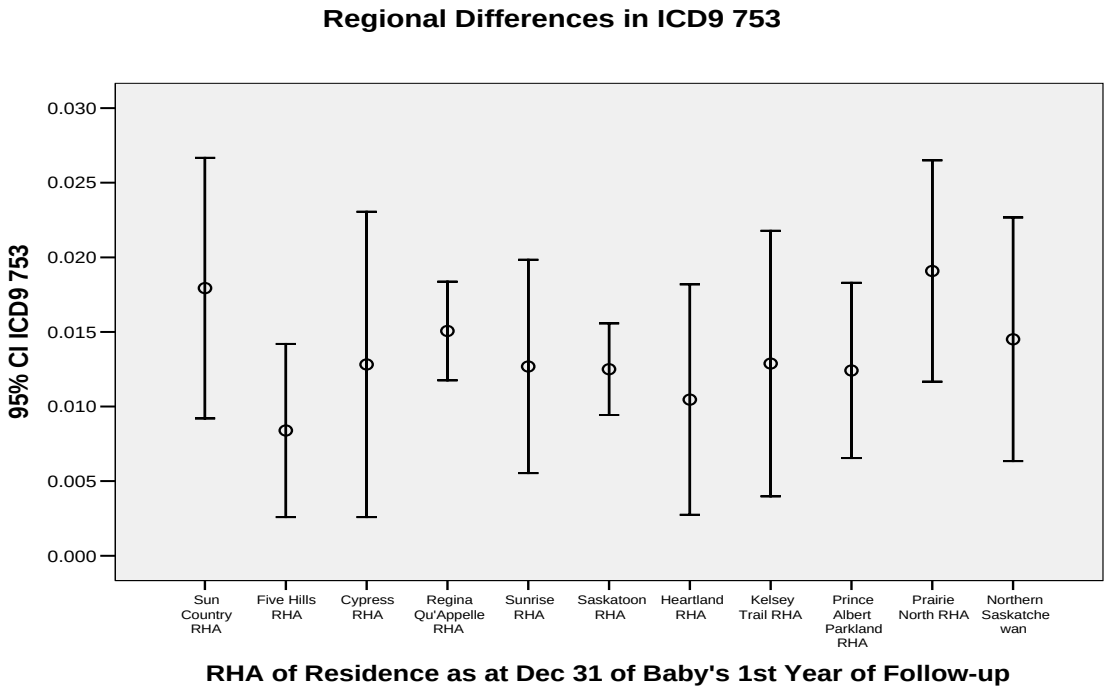

Figure B.15: Regional differences in the mean number of cases of children with congenital anomalies of urinary system (ICD-9 753)

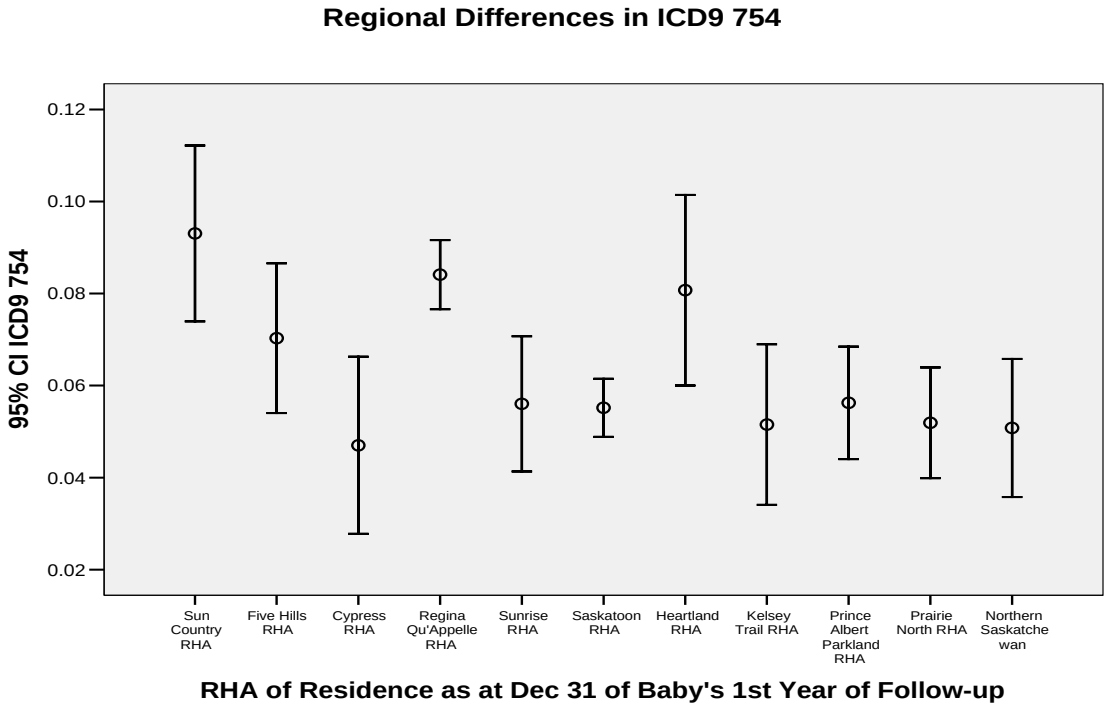

Figure B.16: Regional differences in the mean number of cases of children with certain congenital musculoskeletal deformities (ICD-9 754)

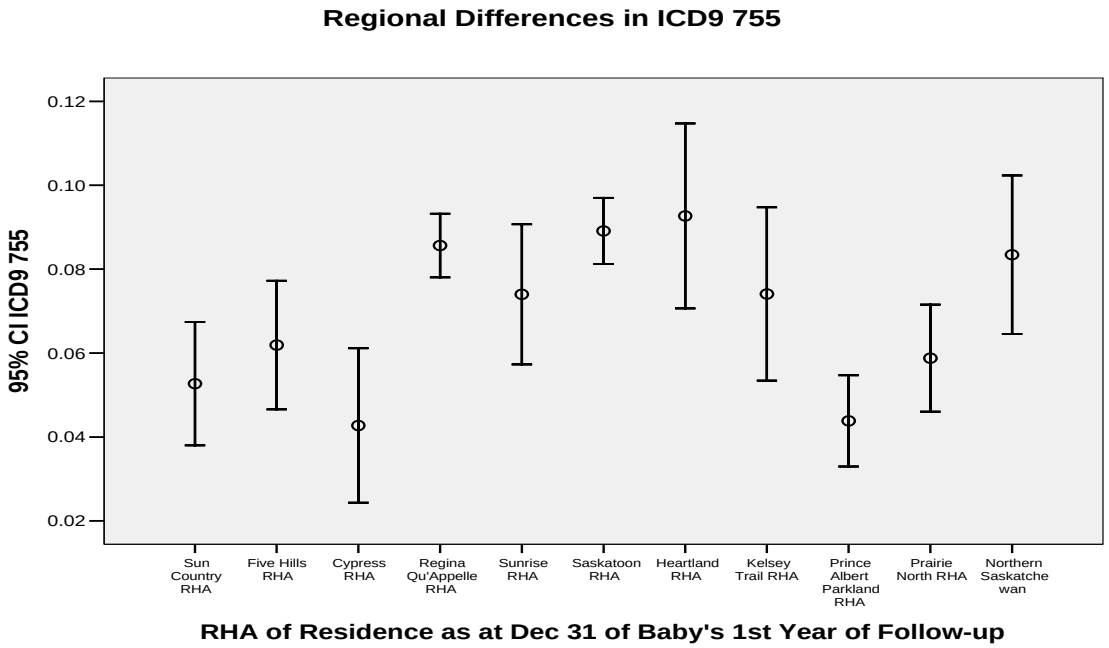

Figure B.17: Regional differences in the mean number of cases of children with other congenital anomalies of limbs (ICD-9 755)

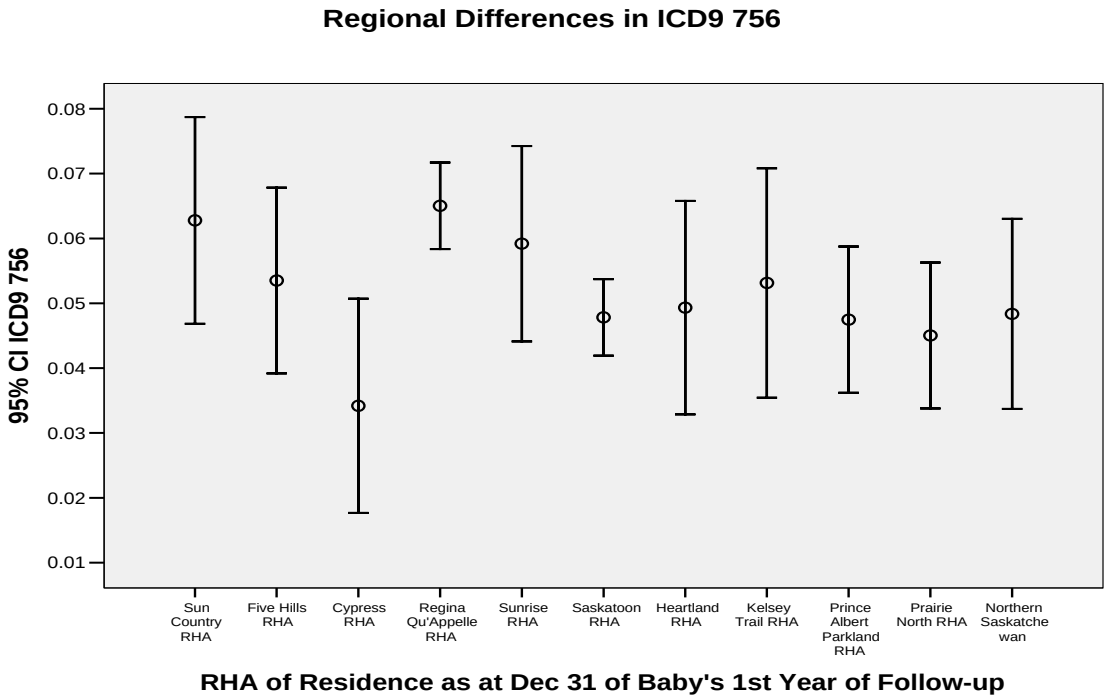

Figure B.18: Regional differences in the mean number of cases of children with other congenital musculoskeletal anomalies (ICD-9 756)

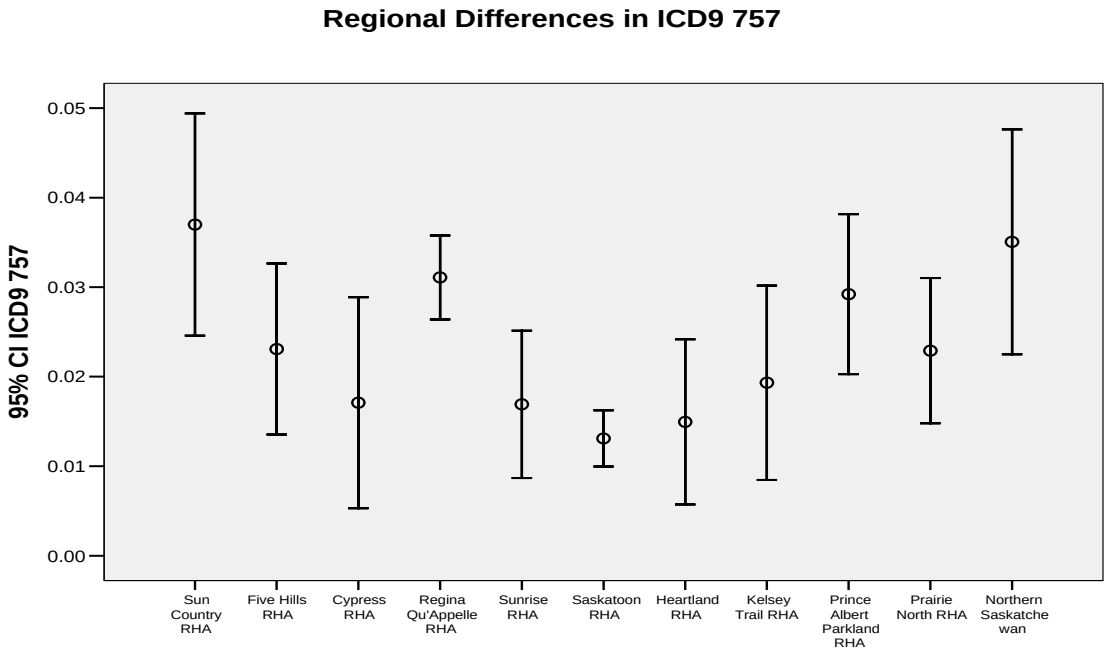

Figure B.19: Regional differences in the mean number of cases of children with congenital anomalies of the integument (ICD-9 757)

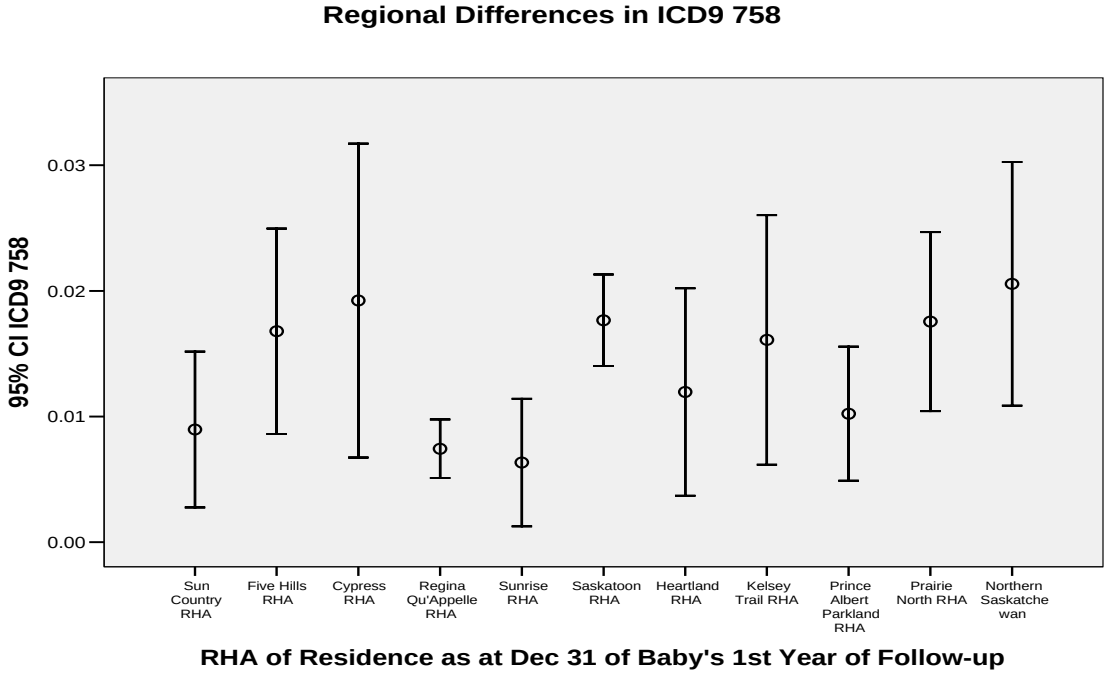

Figure B.20: Regional differences in the mean number of cases of children with chromosomal anomalies (ICD-9 758)

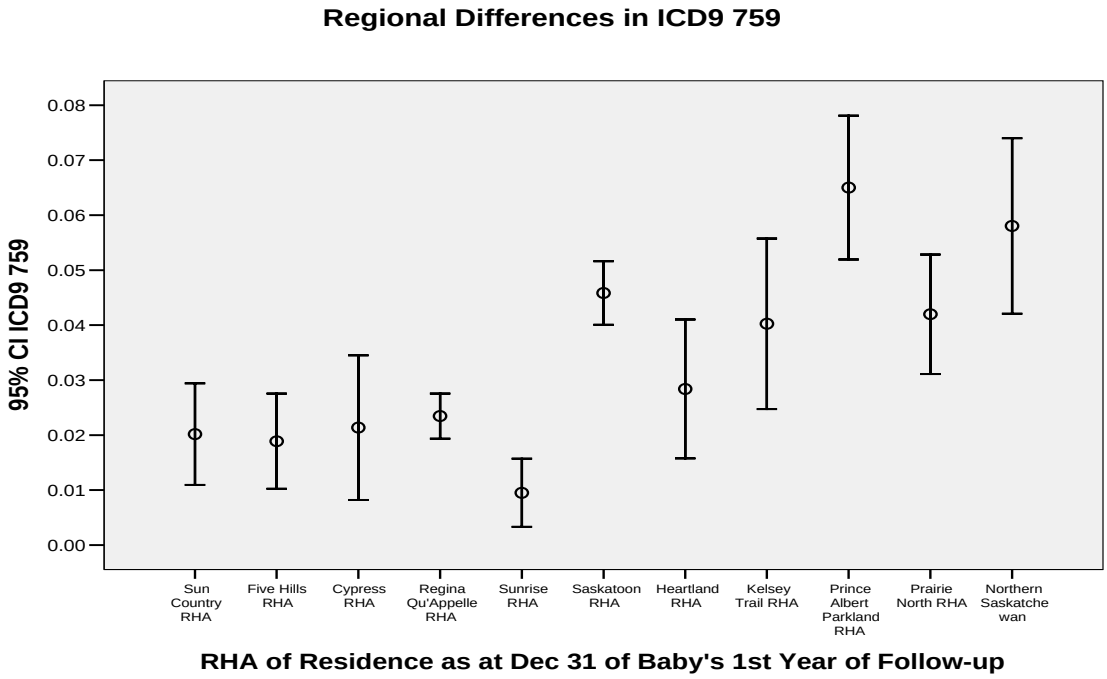

Figure B.21: Regional differences in the mean number of cases of children with other and unspecified congenital anomalies (ICD-9 759)

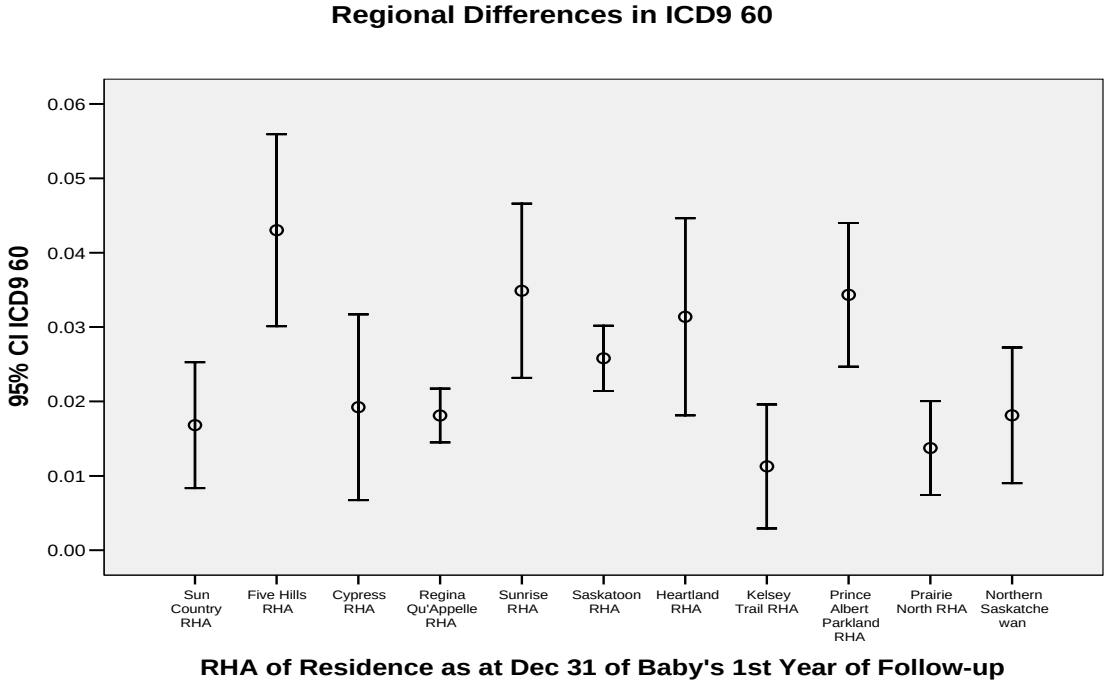

Figure B.22: Regional differences in the mean number of cases of children with congenital dysplasia of hip (MSB Z60)

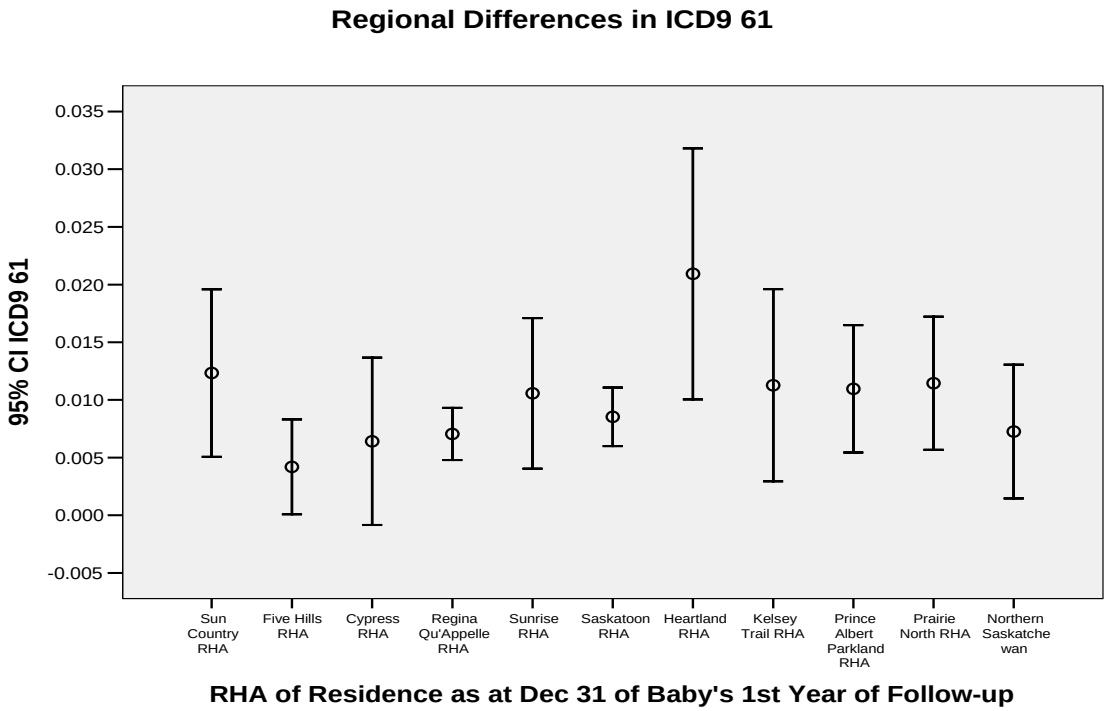

Figure B.23: Regional differences in the mean number of cases of children with clubfoot (MSB Z61)