# 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**
**University of Saskatchewan**
**Saskatoon**

**By**
**Amy Lynn Metcalfe**

## PERMISSION TO USE STATEMENT

In presenting this thesis in partial fulfillment of the requirements for a Postgraduate degree from the University of Saskatchewan, I agree that the Libraries of this University make it freely available for inspection. I further agree that permission for copying of this thesis in any manner, in whole or in part, for scholarly purposes may be granted by the professor or professors who supervised my thesis work or, in their absence, by the Head of the Department or the Dean of the College in which my thesis was done. It is understood that any copying or publication or use of this thesis or parts thereof for financial gain shall not be allowed without my written permission. It is also understood that due recognition shall be given to me and to the University of Saskatchewan in any scholarly use which may be made of any materials in my thesis.

Requests for permission to copy or to make other use of material in this thesis in whole or in part should be addressed to:

Head of the Department of Community Health and Epidemiology
University of Saskatchewan
107 Wiggins Road
Saskatoon, Saskatchewan
S7N 5E5

## 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.

## ACKNOWLEDGEMENTS

A thesis is a huge undertaking, and one that I could not have completed without the assistance of many others. I'd like to begin by thanking the members of my thesis committee Sylvia Abonyi (chair), Pat Blakley, Nazeem Muhajarine (supervisor), and William Osei for their valuable feedback and guidance. I'd also like to gratefully acknowledge the contribution of Tracey Carr, who undertook the onerous task of editing the entire document and for acting as a sounding board and helping me to sort out my thoughts on countless occasions.

I would like to thank Saskatchewan Health for the provision of data, and the Department of Pediatrics at the University of Saskatchewan for a grant that allowed me to access the data. Again, I'd like to acknowledge Pat Blakley for her role in securing that grant – this thesis would not have been possible without that contribution.

Many thanks to Sandra Meeres and Winanne Downey at Saskatchewan Health for their efforts in linking the data and answering all of my questions.

Finally, I'd like to thank my family and friends for their support and encouragement throughout this entire process – I couldn't have made it to the end without you.

## TABLE OF CONTENTS

PERMISSION TO USE STATEMENT ..... i

ABSTRACT ..... ii

ACKNOWLEDGEMENTS ..... iv

TABLE OF CONTENTS ..... v

LIST OF TABLES ..... vii

LIST OF FIGURES ..... ix

CHAPTER ONE: INTRODUCTION ..... 1
1.1 Study Rationale ..... 1
1.2 Objectives and Research Questions ..... 4

CHAPTER TWO: LITERATURE REVIEW ..... 5
2.1 Congenital Anomalies ..... 5
2.1.1 What is a Congenital Anomaly? ..... 5
2.1.2 Causes of Congenital Anomalies ..... 7
2.1.3 Types of Congenital Anomalies ..... 9
2.1.4 Prevention of Congenital Anomalies ..... 10
2.2 Health Disparities ..... 13
2.2.1 What is a Health Disparity? ..... 13
2.2.2 Geographic Health Disparities and Healthy Child Development ..... 13
2.3 Health Services Utilization ..... 16
2.4 Saskatchewan Context ..... 23
2.5 Utility, Validity and Reliability of Administrative Databases ..... 28

CHAPTER THREE: METHODOLOGY ..... 31
3.1 Study Population ..... 31
3.2 Study Design ..... 32
3.3 Data Sources ..... 33
3.4 Study Variables ..... 35
3.5 Ethics Approval and Confidentiality ..... 40
3.6 Software Used ..... 40
3.7 Data Analysis ..... 40
3.7.1 Characteristics of the Study Population ..... 40
3.7.2 Regional Differences in the Prevalence of Congenital Anomalies ..... 40
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? ..... 41

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? ..... 42
3.7.5 Study Question 3: What Factors Influence the Level of Health Care Utilization? ..... 42

CHAPTER FOUR: RESULTS ..... 44
4.1 Description of the Study Population ..... 44
4.2 Bivariate Analysis by Congenital Anomaly Status ..... 51
4.3 Regional Differences in the Prevalence of Congenital Anomalies ..... 56
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? ..... 64
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? ..... 67
4.5.1 Regional Differences in Use of Health Care Services for All Children ..... 67
4.5.2 Regional Differences in Use of Health Care Services for Children With and Without Congenital Anomalies ..... 70
4.6 Study Question 3: What Factors Influence the Level of Health Care Utilization? 75
4.6.1 Model for Number of Physician Visits ..... 76
4.6.2 Model for Total Number of Hospitalizations ..... 78
4.6.3 Model for Total Length of Stay ..... 80

CHAPTER FIVE: DISCUSSION AND CONCLUSIONS ..... 84
5.1 Interpretation of Results ..... 84
5.1.1 Regional Differences in the Prevalence of Congenital Anomalies ..... 85
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? ..... 86
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? ..... 86
5.1.4 Study Question 3: What Factors Influence the Level of Health Care Utilization? ..... 88
5.2 Practical Implications of Results and Directions for Future Research ..... 95
5.3 Study Strengths ..... 97
5.4 Study Limitations ..... 98
5.5 Conclusions ..... 101

REFERENCES ..... 102

APPENDIX A: ICD-9 AND ICD-10 CODES FOR CONGENITAL ANOMALIES .. 108

APPENDIX B: REGIONAL DIFFERENCES IN THE MEAN NUMBER OF CASES OF CONGENITAL ANOMALIES BY REGIONAL HEALTH AUTHORITY ..... 115

## LIST OF TABLES

**Table 2.1:** Barriers and facilitators to accessing health services 19

**Table 2.2:** Amalgamation of health districts into regional health authorities 27

**Table 3.1:** Description of study variables 35

**Table 3.2:** Interaction terms tested in all predictive models for health care utilization 43

**Table 4.1:** Non-time-varying covariates for the study population 44

**Table 4.2:** RHA of residence for the study population by year 47

**Table 4.3:** Non-time-varying predictor variables for children with congenital anomalies and children without congenital anomalies 51

**Table 4.4:** Comparison of time-varying predictor variables for children with congenital anomalies and children without congenital anomalies 53

**Table 4.5:** Regional differences in the mean number of cases of CAs compared to the Saskatoon Health Region 56

**Table 4.6:** Differences in use of health care for children with and without congenital anomalies 65

**Table 4.7:** Differential use of health care services for children with specific types of CAs versus children without that particular CA 66

**Table 4.8:** Crude regional differences in use of health care services 68

**Table 4.9:** Crude regional differences in travel for health care services 70

**Table 4.10:** Crude regional differences in health care utilization for children with and without congenital anomalies 71

**Table 4.11:** Crude regional differences in travel for health care utilization for children with and without congenital anomalies 75

**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 76

**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 78

**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 80

**Table 5.1:** Statistically significant variables in models for level of health care utilization 89

**Table A.1:** ICD-9 and MSB codes for congenital anomalies found in the study population 108

**Table A.2:** Reclassification of ICD-10 codes found in the study population to ICD-9 codes 113

## LIST OF FIGURES

**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) 3

**Figure 2.1:** Sensitive stages of development 7

**Figure 2.2:** Causes of congenital anomalies 8

**Figure 2.3:** Prevalence of the most common types of CAs in Canada (*excluding Nova Scotia and Quebec) per 10,000 births 9

**Figure 2.4:** Individual determinants of health services utilization 18

**Figure 2.5:** Map of Saskatchewan health districts 26

**Figure 2.6:** Map of Saskatchewan regional health authorities 28

**Figure 4.1:** Percentage of families receiving income assistance by year in the study population 48

**Figure 4.2:** Percentage of children who traveled outside of their home RHA for a physician visit by year 48

**Figure 4.3:** Percentage of children who traveled outside of their home RHA for a hospital admission by year 49

**Figure 4.4:** Prevalence of congenital anomalies in the study population per 1000 live births 50

**Figure 4.5:** Prevalence of categories of CAs in the study population 50

**Figure 4.6:** Prevalence of any CAs by RHA 57

**Figure 4.7:** Prevalence of CAs of the eye (ICD-9 743) by RHA 58

**Figure 4.8:** Prevalence of bulbus cordis anomalies and anomalies of cardiac septal closure (ICD-9 745) by RHA 58

**Figure 4.9:** Prevalence of other CAs of the heart (ICD-9 746) by RHA 59

**Figure 4.10:** Prevalence of other CAs of the circulatory system (ICD-9 747) by RHA 59

**Figure 4.11:** Prevalence of cleft lip and cleft palate (ICD-9 749) by RHA 60

**Figure 4.12:** Prevalence of CAs of the upper alimentary tract (ICD-9 750) by RHA 60

**Figure 4.13:** Prevalence of certain congenital musculoskeletal deformities (ICD-9 754) by RHA 61

**Figure 4.14:** Prevalence of other CAs of limbs (ICD-9 755) by RHA 61

**Figure 4.15:** Prevalence of other musculoskeletal anomalies (ICD-9 756) by RHA 62

**Figure 4.16:** Prevalence of CAs of the integument (ICD-9 757) by RHA 62

**Figure 4.17:** Prevalence of chromosomal anomalies (ICD-9 758) by RHA 63

**Figure 4.18:** Prevalence of other and unspecified CAs (ICD-9 759) by RHA 63

**Figure 4.19:** Prevalence of congenital dysplasia of hip (MSB Z60) by RHA 64

**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 68

**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 69

**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 69

**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 72

**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 73

**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 74

**Figure B.1:** Regional differences in the mean number of cases of children with CAs 115

**Figure B.2:** Regional differences in the mean number of cases of children with multiple CAs 115

**Figure B.3:** Regional differences in the mean number of cases of children with neural tube defects (ICD-9 740 and 741) 116

**Figure B.4:** Regional differences in the mean number of cases of children with other congenital anomalies of nervous system (ICD-9 742) 116

**Figure B.5:** Regional differences in the mean number of cases of children with congenital anomalies of eye (ICD-9 743) 117

**Figure B.6:** Regional differences in the mean number of cases of children with congenital anomalies of ear, face, and neck (ICD-9 744) 117

**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) 118

**Figure B.8:** Regional differences in the mean number of cases of children with other congenital anomalies of heart (ICD-9 746) 118

**Figure B.9:** Regional differences in the mean number of cases of children with other congenital anomalies of circulatory system (ICD-9 747) 119

**Figure B.10:** Regional differences in the mean number of cases of children with congenital anomalies of respiratory system (ICD-9 748) 119

**Figure B.11:** Regional differences in the mean number of cases of children with cleft palate and cleft lip (ICD-9 749) 120

**Figure B.12:** Regional differences in the mean number of cases of children with other congenital anomalies of upper alimentary tract (ICD-9 750) 120

**Figure B.13:** Regional differences in the mean number of cases of children with other congenital anomalies of digestive system (ICD-9 751) 121

**Figure B.14:** Regional differences in the mean number of cases of children with congenital anomalies of genital organs (ICD-9 752) 121

**Figure B.15:** Regional differences in the mean number of cases of children with congenital anomalies of urinary system (ICD-9 753) 122

**Figure B.16:** Regional differences in the mean number of cases of children with certain congenital musculoskeletal deformities (ICD-9 754) 122

**Figure B.17:** Regional differences in the mean number of cases of children with other congenital anomalies of limbs (ICD-9 755) 123

**Figure B.18:** Regional differences in the mean number of cases of children with other congenital musculoskeletal anomalies (ICD-9 756) 123

**Figure B.19:** Regional differences in the mean number of cases of children with congenital anomalies of the integument (ICD-9 757) 124

**Figure B.20:** Regional differences in the mean number of cases of children with chromosomal anomalies (ICD-9 758) 124

**Figure B.21:** Regional differences in the mean number of cases of children with other and unspecified congenital anomalies (ICD-9 759) 125

**Figure B.22:** Regional differences in the mean number of cases of children with congenital dysplasia of hip (MSB Z60) 125

**Figure B.23:** Regional differences in the mean number of cases of children with clubfoot (MSB Z61) 126

# 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).

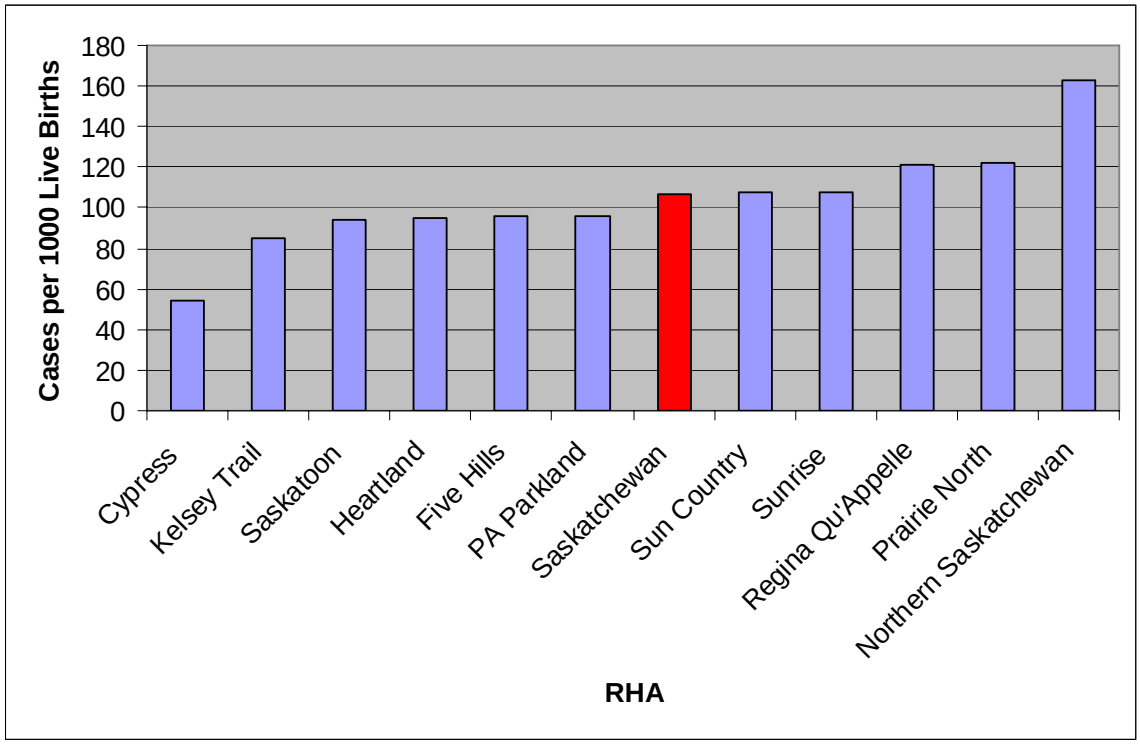


**Figure 1.1:** Combined prevalence of five selected CAs (neural tube defects, limb reduction deficits, Down syndrome, cleft lip/cleft palate and congenital heart defects) by regional health authority (1990-1999) (9)

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.

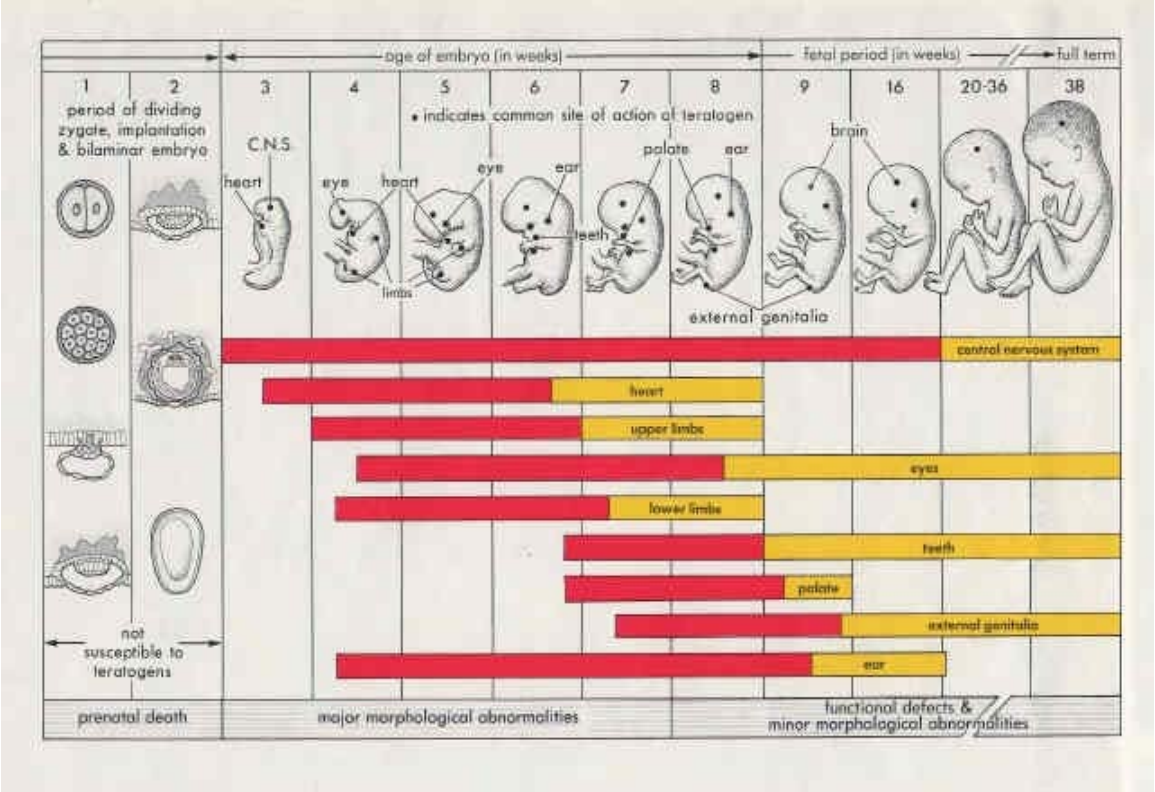


**Figure 2.1:** Sensitive stages of development (1)

### 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).

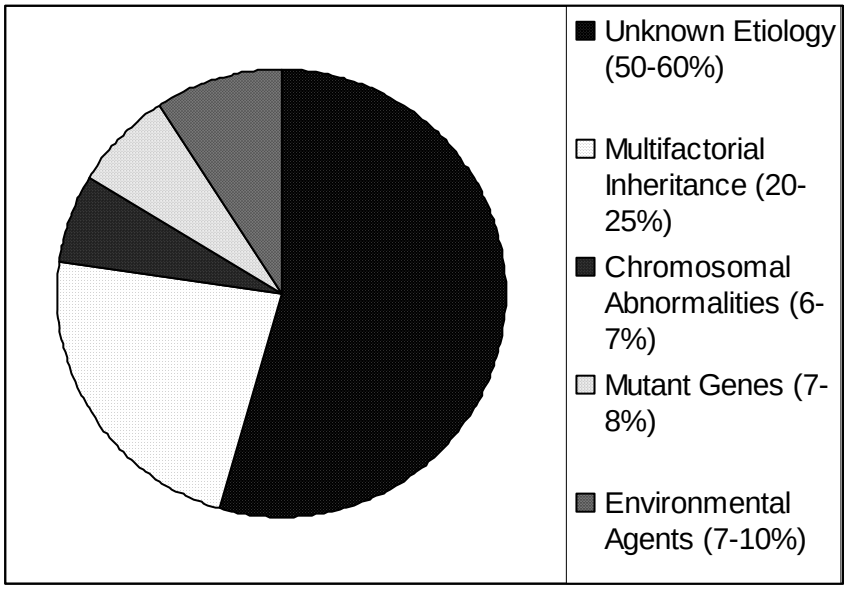


**Figure 2.2:** Causes of congenital anomalies (2)

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).

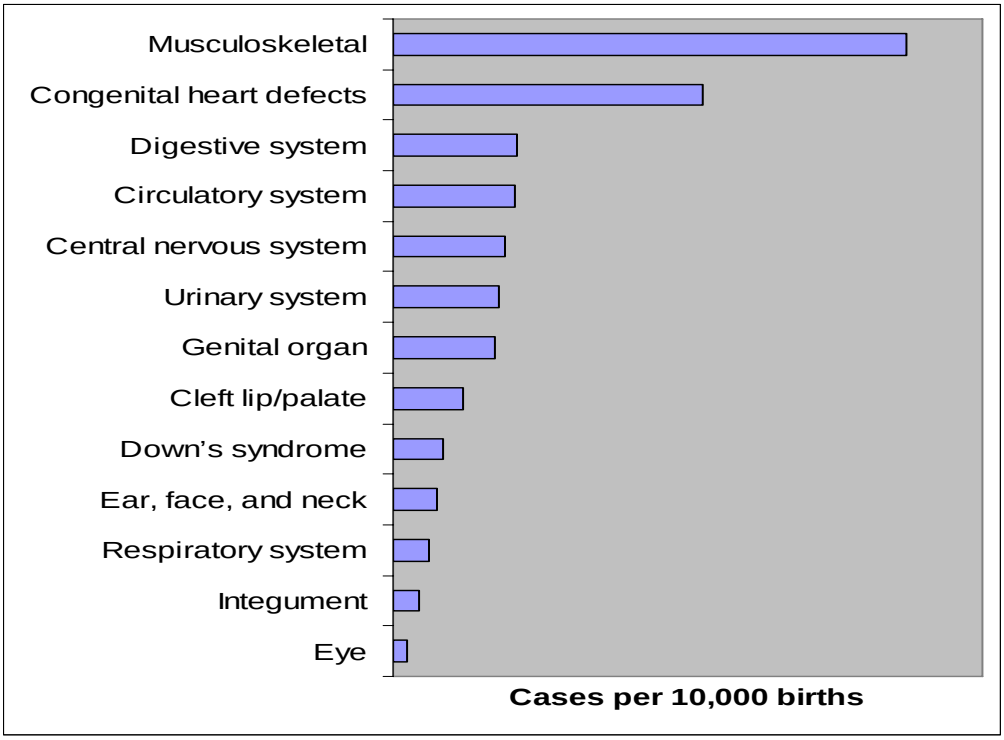


**Figure 2.3:** Prevalence of the most common types of CAs in Canada (*excluding Nova Scotia and Quebec) per 10,000 births (15)

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 15$^{th}$ birthday compared to 98% of subjects without birth defects (i.e. children with CAs were more likely to die before their 15$^{th}$ 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).

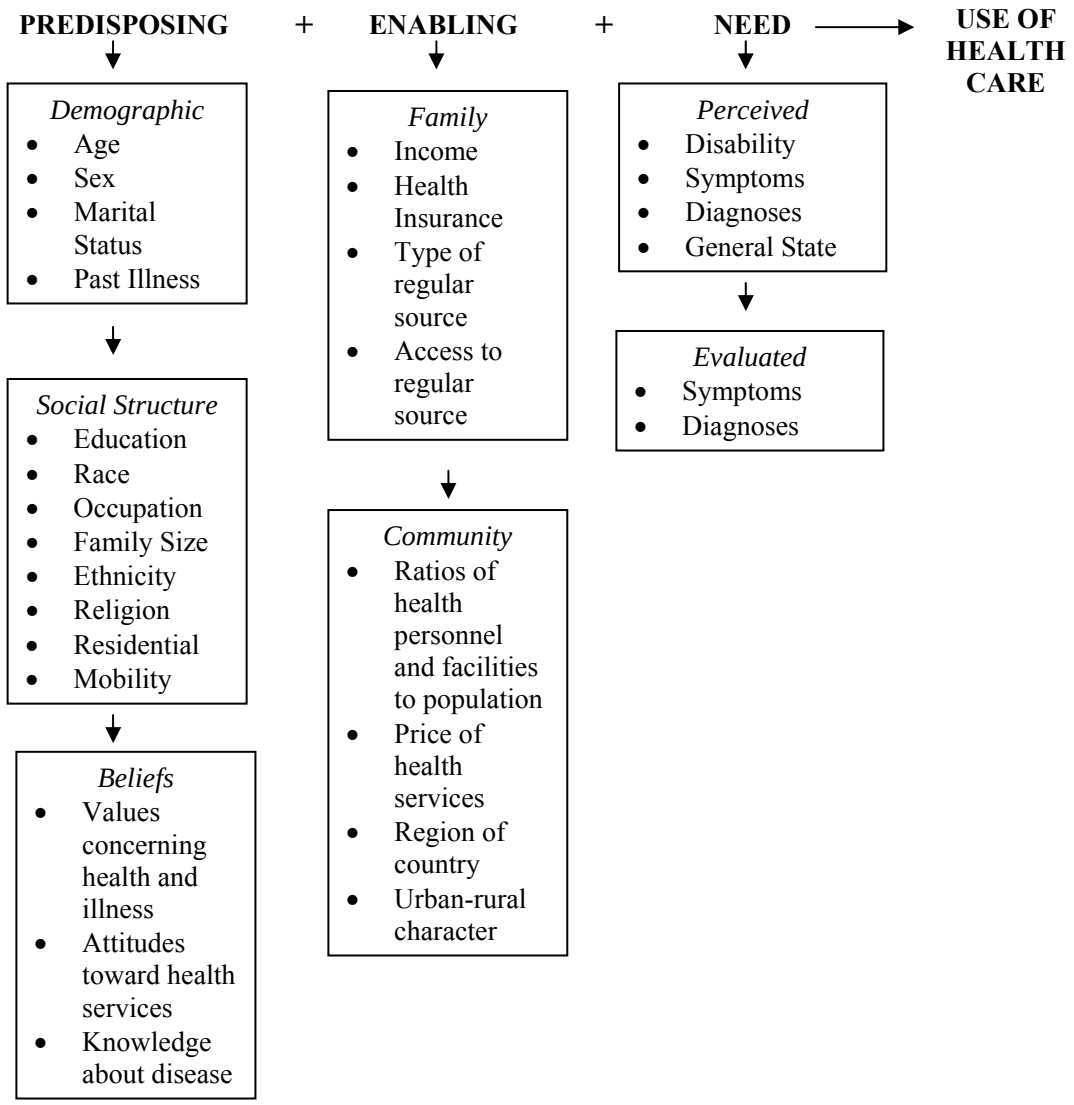


**Figure 2.4:** Individual determinants of health services utilization (11).

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