EFFECTS OF HYDROCEPHALUS ON HOSPITAL USE, ASSOCIATED COSTS, AND ACCESS TO CARE AMONG CHILDREN WITH SPINA BIFIDA

by

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ABSTRACT

ELIZABETH RADCLIFF. Effects of hydrocephalus on hospital use, associated costs, and access to care among children with spina bifida. (Under the direction of DR. SARAH B. LADITKA and DR. CYNTHIA H. CASSELL)

Objectives: This study examined hospital resource use, including timeliness of surgical repair of spina bifida (SB), and geographical access to hospital care, by the presence of hydrocephalus, isolated or non-isolated SB, and selected sociodemographic characteristics among children with SB.

Methods: This was a retrospective, statewide, population-based study of children with SB, born in Florida 1998-2007, identified by the Florida Birth Defects Registry and linked to hospital discharge records. Information about hospitalizations, lengths of stay, and costs were obtained from hospital discharge data for infants (<1 year) and children ages one to four years. Time to SB surgical repair was calculated using procedural codes and hospital discharge data. One-way travel time and distance to access hospital care were calculated using geocoded maternal residential addresses, hospital addresses, and Florida road networks. Chi-square tests and logistic, Poisson, generalized linear regression, and ordinary least squares were used to examine the study objectives. Models were adjusted for hydrocephalus; isolated (no other major birth defect) vs. non-isolated SB, and selected predisposing, enabling, and need characteristics

Results: Of 614 children, 42.4% of children had isolated SB and hydrocephalus; 32.3% had isolated SB without hydrocephalus; 14.5% had non-isolated SB and hydrocephalus, and 10.9% had non-isolated SB without hydrocephalus. In adjusted results, infants with isolated SB and hydrocephalus had 53% more hospitalizations and

2.6 times the number of hospitalized days and costs compared with infants with isolated SB without hydrocephalus. Infants and children with non-isolated SB and hydrocephalus had twice the number of post-birth hospitalizations and hospitalized days than children with isolated SB without hydrocephalus, but only about 40% higher costs. Regarding timeliness of surgical repair, of 299 infants with a recorded repair, 68.6% had repair by day two, 15.1% had repair days three through seven, and 16.4% had repair after day seven. In adjusted results, hydrocephalus was the only characteristic associated with repair by day two (adjusted prevalence ratio=1.80, 95% confidence interval: 1.31-2.48). Of 612 children with a geocoded address, 56.4% of infants and 61.4% of children had a one-way average travel time of ≤30 minutes to hospitals. Infants with non-isolated SB and hydrocephalus traveled the longest to hospitals (mean: 60.8; median: 34.2; range 5-494 minutes). In adjusted results, non-isolated SB, maternal minority race/ethnicity, lower maternal education, and rural residence were associated with lower likelihood of traveling ≤30 minutes to hospitals during infancy.

Discussion: Comorbidities substantially increase hospital resource use for children with SB, particularly during infancy. Results also showed that the majority of infants with SB had a timely repair. Infants with non-isolated SB and hydrocephalus traveled the longest to access hospital care. Findings underscore the need to consider comorbidities when examining hospital resource use for children with SB and other birth defects. Results also demonstrate that birth defects registry data and GIS-based methods are useful to evaluate geographical access to hospitals for children with birth defects.

DEDICATION

To my beloved husband Frank

who gave me freedom to try and

who traveled each day of this journey with me, patiently and with love.

To my daughters,

for being my beautiful cheering section.

And to my parents,

who always believed I could.

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LIST OF ABBREVIATIONS

The following list includes acronyms used in this dissertation:

AHCA	Agency for Health Care Administration
AHRQ	Agency for Healthcare Research and Quality
CDC	Centers for Disease Control and Prevention
CI	Confidence interval
CSHCN	Children with special health care needs
FBDR	Florida Birth Defects Registry
FDOH	Florida Department of Health
GIS	Geographical Information Systems
ICD-9-CM	International Classification of Disease, Ninth revision; Clinical Modification
IRB	Institutional Review Board
NTD	Neural tube defect
NBDPN	National Birth Defects Prevention Network
NCBDDD	National Center on Birth Defects and Developmental Disabilities
OR	Odds ratio (uOR=unadjusted, aOR=adjusted)
SB	Spina bifida
VP	Ventriculoperitoneal (shunt)

CHAPTER 1: INTRODUCTION

1.1 Background of Birth Defects and Spina Bifida

An infant is born with a birth defect every 4.5 minutes, and major birth defects are diagnosed in 3% of all live births in the United States [1-3]. Birth defects are conditions present at birth that involve structural or functional abnormalities in one or more parts of the body, and may result in adverse effects on a child's health, development, and functional capacities [2, 4, 5]. Birth defects are among the leading causes of pediatric hospitalizations and contribute substantially to the health care costs in the United States [2, 6-8]. Birth defects are also the leading cause of mortality during the first year of life, accounting for 20% of all infant deaths [9].

Spina bifida (SB), one type of major birth defect, is a neural tube defect (NTD) that results from a failure of the caudal neural tube to fuse early in embryonic development [10, 11]. This complex birth defect affects approximately 1,500 live-born U.S. infants each year [1] and typically requires life-long, multidisciplinary health care. Children with special health care needs (CSHCN), a broader group that includes children with birth defects, typically use more health services, have greater costs, and face more barriers when accessing health services than children without special health care needs [2, 7, 12-15]. We know that children with SB have greater hospital costs than children who do not have SB and that they incur their greatest costs during their first year of life [14]. Spina bifida may be accompanied by multiple comorbidities. The presence of

comorbidities may influence health care use and costs [16, 17]. Timely surgical repair of SB may reduce the effects of certain comorbidities and mortality associated with SB [16, 18-21].

1.2 Statement of Problem

Gaps exist in our understanding of how specific comorbidities affect hospital use and access to care among children with SB. In addition, little information is available about how the effects of comorbidities may change across payer types and through childhood. Few studies have examined timeliness of services for children with SB, including the percentage of infants who have a timely surgical repair of SB.

The objective of my dissertation research was to explore the effects of comorbidities including hydrocephalus, one of the most common comorbidities associated with SB, on hospital resource use (number of hospitalizations, number of hospitalized days, and associated inpatient costs) and access to hospital care for children with SB. My study also explored predisposing, enabling, and need characteristics associated with hospital resource use and access to hospital care. In addition, my study explored predisposing, enabling, and need characteristics associated with timely surgical repair of SB and the effect of hydrocephalus and isolated versus non-isolated SB on timely surgical repair of SB.

1.3 Overview of Data Sources

This dissertation research was a retrospective, statewide, population-based analysis of inpatient hospital use and access to care for children with SB ages birth through four years born in Florida between January 1, 1998, and December 31, 2007. Florida was chosen because it provided a robust and diverse study population. The state

of Florida was the fastest growing and fourth most populous state according to the 2000 U.S. Census [22, 23]. In addition, Florida was fourth in number of annual live births, second in number of live births to non-Hispanic Black women, and third in number of live births to Hispanic women during the study period of 1998-2008 [22-24].

The state of Florida also supports a statewide, population-based birth defects registry and a statewide agency for the collection of hospital discharge data that provided information for this project. The statewide, population-based study sample used in this study included linked, longitudinal data from the Florida Birth Defects Registry (FBDR) and the Florida Bureau of Vital Statistics, both within the Florida Department of Health (FDOH), and from the Florida Agency for Health Care Administration (AHCA). These agencies provided robust, diverse sources of information for this project.

The Florida AHCA is a statewide organization that oversees Florida's Medicaid program and the licensure of the state's 41,000 health care facilities [25]. The Florida AHCA provided the hospital discharge data for this research project. The Florida AHCA data included information on inpatient and ambulatory hospital use and charges for registered Florida hospitals, birth centers, and surgical centers in the state [25].

The FBDR is a statewide, population-based surveillance system that uses passive, case-finding techniques to identify infants having at least one FBDR-eligible *International Classification of Disease, Ninth revision; Clinical Modification* (ICD-9-CM) code diagnosed during the first year of life [26-28]. The FBDR includes live-born infants whose mothers are Florida residents at the time of the infant's birth. The FBDR excludes infants who were adopted or prospective adoptees or whose mothers delivered out-of-state [27]. The Florida Bureau of Vital Statistics provided official birth and death

records for the FBDR [27]. In addition to vital statistics data, the FBDR identifies diagnosis codes present in several other datasets to increase case ascertainment and provide a more complete registry of birth defects in Florida. These data include information from the *Centers for Medicare and Medicaid Services Minimum Data Set* and from the *Early Steps Program* data set. The FBDR merges information from these datasets to create a single, non-duplicated dataset of Florida infants with birth defects.

Algorithms involving maternal, paternal, and child social security numbers were used by the FBDR as the primary linking variables; additional linkages were based on demographic and medical data. The linking rates varied across data sets from 85% to 95%; the overall linkage rates provided a robust study size for this research [28, 29]. An evaluation of the FBDR found that the program correctly identified 86.6% of infants born with selected birth defects between 2003 and 2006 [29]. FBDR case ascertainment for SB without anencephaly during 2003-2006 was 87.9% [29].

The FDOH, the FBDR, the Florida AHCA, and the University of South Florida have collaborated for over 15 years to create the state of Florida's birth defects registry. As part of a collaborative project with the University of North Carolina at Charlotte (UNC Charlotte), the University of South Florida and the FDOH created a subset of infants in the FBDR with selected major birth defects that linked FDBR records to the Florida AHCA discharge records. The longitudinal data for this project included inpatient and outpatient hospitalizations that were initiated between January 1, 1998 and December 31, 2008. This allowed for at least one year of hospitalizations for each infant with SB. Data linkage was conducted by the FDOH and the University of South Florida using a step-by-step linking strategy. Linkage was conducted in stages that ranged from

high to lower levels of confidence in the linkage. For example, stage 1 consisted of an exact match between infant Social Security Number (SSN), maternal SSN, infant date of birth, and infant sex. Subsequent stages included linkages based on less exact matches between infant and maternal SSNs; crossover matching between infant, maternal, and paternal SSN; and "fuzzy" matching on date of birth (e.g., a one or two day variability in date of birth or reversed month and day digits). When a link was established during a given step, the record was then removed from the pool of available records to be linked during subsequent, lower-confidence stages. Linkage was conducted separately for infants born as singletons versus those born as part of a multiple (twin or triplet) birth because multiple births increase the complexity of linkages steps. Details of this stepwise linking strategy have been described previously [30].

Following secure transmission of these linked data sets to UNC Charlotte, data from children with neural tube defects were merged with the two Florida AHCA data sets. Neural tube defects, specifically SB without anencephaly, were identified using the ICD-9-CM codes 740.0, 740.1, and 741.00-741.93. The first Florida AHCA data set was an infancy dataset that contained data on hospitalizations initiated during the first year of life. The second Florida AHCA data set was a longitudinal dataset that contained data for hospitalizations initiated after the first year of life. Hospital discharge data from January 1, 1998 through December 31, 2008 were used to allow for at least one year of hospitalizations for each infant with SB. Because of increasingly smaller numbers over the years, I only used data from birth through four years for the statistical analyses in my dissertation.

In the study period of 1998-2007, Florida reported 2,135,079 live births [27]. Among the 2.1 million infants, the FBDR identified about 70,000 infants who were born with a major birth defect [27]. The University of South Florida and the FDOH created a subset of 52,759 FBDR infants with selected major birth defects, including SB, which linked to the Florida AHCA discharge records. Figure 1.1 (page 9) shows the process for identification of infants for the final study samples.

1.4 Data Management

This dissertation used data from a larger research project funded by the March of Dimes Foundation grant #5-FY09-533, conducting research under protocols approved by the FDOH Institutional Review Board (IRB), the Centers for Disease Control and Prevention's (CDC) National Center on Birth Defects and Developmental Disabilities (NCBDDD) IRB, and by the UNC Charlotte IRB) (Protocol #12-07-12). The UNC Charlotte IRB Protocol #12-07-12 is valid through July 15, 2014. For the purpose of my dissertation, a new protocol was submitted and approved by the UNC Charlotte IRB (Protocol approval #12-12-24).

The data acquired from the FBDR and the Florida Bureau of Vital Statistics, within the FDOH, and from the Florida AHCA, and provided by the University of South Florida were de-identified except for maternal residential address at the time of the infant's birth and corresponding longitudinal and latitudinal (X, Y) coordinates. Appropriate measures, including storage of data on a secure network, remained in place throughout this project to ensure confidentiality of the data. Data use agreements were also in place, signed by the respective agencies necessary to carry out this dissertation

project. The agencies signing the data use agreements included the FDOH, the University of South Florida, the CDC's NCBDDD, and UNC Charlotte.

1.5 Relevance to Current Health Care Priorities

The goals of *Healthy People 2020* highlight the need to increase the proportion of CSHCN who have access to a medical home [31]. Similarly, the Spina Bifida Association's 2012 Congressional Policy Agenda called for ensured access to care for individuals with SB, especially through provision of adequate insurance [32]. Experts convened by the CDC helped establish public health research priorities, including timeliness of services and access to care, for selected birth defects including orofacial clefts, craniosynostosis, congenital heart defects, and Down syndrome [33-36]. Finally, the National Institute of Health has recognized pediatric hydrocephalus as an under-researched area [37]. Findings from my dissertation addressed these identified health care priority areas by examining access to care and standards of care for children with SB. Findings from my dissertation research contribute new information to our understanding of how hydrocephalus and isolated vs. non-isolated SB influence health resource use and access to care as comorbidities to SB.

1.6 Objectives of my Dissertation Research

My dissertation research focused on health resource use, timeliness of care, and access to care among children with SB, a type of birth defect that is included under the broader category of CSHCN. My research had three specific objectives. The first objective was to examine the effects of hydrocephalus, as well as other predisposing, enabling, and need characteristics, on hospital use and associated costs among children with SB (Chapter 3). The second objective was to explore predisposing, enabling, and

need characteristics associated with the timeliness of primary surgical repair of SB (Chapter 4), including the role of hydrocephalus and isolated versus non-isolated SB. The third objective was to investigate the effects of hydrocephalus, as well as other predisposing, enabling, and need characteristics, on access to hospital care in terms of travel time and distance for children with SB (Chapter 5). Aday and Anderson's *Framework for the Study of Access to Medical Care* provided the theoretical structure for my research. Chapter 2 reviews the literature that is relevant to my research topics.

My research improved upon previous studies in several ways. First, it addressed topics in health care priority areas by providing a better understanding of the influence of the comorbidity hydrocephalus on hospital resource use and access to care among children with SB. Second, by reporting the percentage of children who had timely surgical repair of SB, the results of this research contributed to our knowledge of adherence to standards of care for children with SB. Findings also suggested factors associated with timeliness of surgical repair among children with SB. Understanding factors associated with timely care are important because of known associations between timing of the surgical repair of SB and later comorbidities. Finally, findings of this research added to our understanding of geographic access to care for children with SB with findings based on Geographic Information Systems (GIS) methods.

Increasing our understanding in each of these areas can help inform opportunities for improved health service delivery, health outcomes, and quality of life for children with SB and their families. My research can also help inform research for children with other types of birth defects.

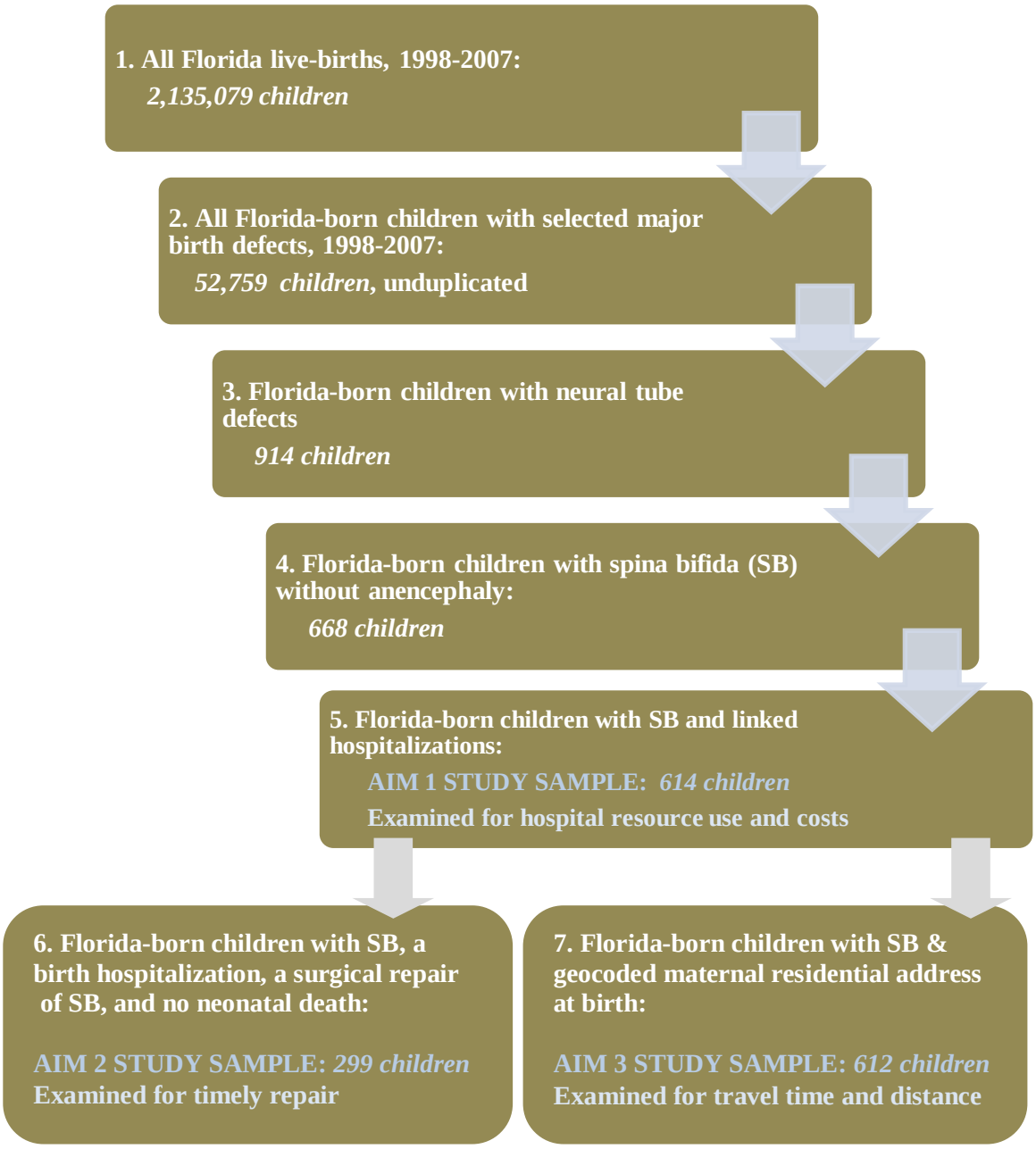

Figure 1.1 Selection of the three study samples for examination of Aim 1) hospital resource use, Aim 2) timely surgical repair of SB, and Aim 3) geographical access to hospital care among Florida-born children with spina bifida, birth through four years of age, 1998-2007

CHAPTER 2: LITERATURE REVIEW

My review of the literature begins with an overview of the nature, prevalence, and significance of birth defects in the U.S. health care system, with a specific focus on the epidemiology of spina bifida (SB). I continue with a description of the current management and treatment of SB and an explanation of isolated versus non-isolated SB and associated comorbidities, including hydrocephalus. I then describe research that examined the influence of comorbidities on health outcomes for children with SB and literature related to the timeliness of the primary surgical repair of SB. Next, I review current literature on hospital resource use, including measures such as charges, costs, and numbers and lengths of hospital admissions for children with birth defects, including SB. Finally, I describe findings on the role of predisposing, enabling, and need characteristics on hospital resource use, timeliness of care, and access to care for children with birth defects, including SB. Chapter 2 concludes with a description of how this literature, coupled with the Aday and Anderson *Framework for the Study of Access to Medical Care*, provided a theoretical and literature-informed framework for my dissertation research.

2.1 Overview of Birth Defects: Definition, Prevalence, and Significance

Birth defects are conditions present at birth that involve structural or functional abnormalities in one or more parts of the body [2, 4, 5]. Birth defects can result in adverse effects to a child's health, development, and functional capacities [2]. Most birth

defects occur early in pregnancy, typically during the first trimester [4, 5]. Some birth defects are due to genetic or chromosomal abnormalities (e.g., an extra chromosome 21 results in Down syndrome). Other birth defects may be caused by environmental exposures (e.g., fetal alcohol spectrum disorders may result from drinking alcohol during pregnancy) or by deficiencies in certain micronutrients (e.g., a folic acid deficiency is associated neural tube defects) [5, 38, 39]. Birth defects can also be associated maternal age, diabetes, obesity, or infection [4, 39]. The specific causes of most birth defects, however, remain unknown. Most birth defects are thought to be the result of multiple factors [4, 5].

In the United States, an infant is born with a birth defect every 4.5 minutes [5]. The overall prevalence estimate of major birth defects in the United States is 1 in 33 or approximately 3% of all live births [1, 3]. Birth defects are a leading contributor to disability and pediatric hospitalizations and accounted for more than $2.6 billion in annual hospital costs in the United States in 2004 [2, 6-8]. In addition, birth defects are the leading cause of death during the first year of life, accounting for more than 20% of all infant mortality [9]. The impact of birth defects on children, families, and health care systems make birth defects an important focus for public health and health services research.

2.2 Overview of Spina Bifida, a Specific Major Birth Defect

2.2.1 Description of Spina Bifida

Neural tube defects (NTDs) are a type of birth defect that affect the central nervous system and that occur very early in embryonic development, usually by the 28th day post-conception [10, 11]. The birth defect category of NTDs includes several

different types of central nervous system malformations, such as omphalocele, encephalocele, anencephaly, and SB [10, 11, 40, 41]. Anencephaly, a defect not typically compatible with life, and SB without anencephaly are the most common forms of NTDs [11, 41, 42].

Spina bifida is specifically a defect in which the caudal neural tube does not close completely at some point along the spine from the cervical to the sacral regions [10, 11]. Spina bifida occulta ("closed") occurs when a small gap in the spine exists, but no opening occurs on the back, thus the spinal cord and nerves remain essentially intact [10, 11, 41]. This type of SB presents few health care problems and may not be diagnosed at birth [43]. Meningomyelocele, an "open" type of SB, involves the herniation of both the meninges (the membranes that surround the central nervous system) and the spinal cord into a sac outside the vertebral column [10, 43]. Meningomyeloceles are the most serious and most common presentation of SB and may result in significant disability [11, 41]. A 2012 National Birth Defects Prevention Study used population-based birth defects surveillance data from a 10-state area to examine infants with various presentations of SB [42]. The researchers found that 85.6% of infants with SB had the subtypes of SB that included myelomeningocele, meningocele, and myelocele [42]. Other research reports as many as 90% of children with SB have the myelomeningocele type [11, 41].

The severity of neurologic impairment is related to the position of the defect along the spinal column, which then directly affects the child's mobility and ability to maintain bowel and bladder control [44]. In addition, a child with SB is at risk for related challenges, such as hydrocephalus, scoliosis and other orthopedic issues, urinary tract infections and chronic renal disease, and obesity [44-46]. A child with SB may face

challenges with educational, social, and psychological development [47] and typically requires life-long, multidisciplinary health care.

2.2.2 Epidemiological Profile of Spina Bifida

The occurrence of NTDs has notably declined in the United States because of the availability of prenatal diagnosis [46, 48, 49]. The occurrence of NTDs has also declined substantially since the mandatory fortification of the U.S. cereal grain supply with folic acid [48, 50-53]. In 1998, the U.S. government passed legislation that required mandatory fortification of the nation's cereal grain supply with folic acid. In the years following mandatory folic acid fortification, the occurrence of NTDs decreased significantly [48, 50-53]. Research reported a SB prevalence estimate of 2.62 cases per 10,000 live births prior to mandatory folic acid fortification (October 1995 through December 1996) compared to a SB prevalence estimate of 2.02 cases per 10,000 live births for October 1998 through December 1999, a decrease in prevalence of 22.9% [52]. More recent reports on post-fortification trends found an additional decrease of 6.9% in SB prevalence between 1999-2000 compared to 2003-2005 data [54]. The most recent annual U.S. prevalence estimate for SB is approximately 1,500 infants or 1 in 2,858 live births per year [1]. According to Florida Birth Defects Registry (FBDR) data, an average of 70 infants with SB were born each year in the state of Florida between 1998-2007 [27]. Table 2.1 (page 34) shows the distribution of births of children with SB included in the study by year of birth. Figure 2.1 (page 35) shows the distribution with an overall trend line included.

Differences in prevalence estimates of SB exist across racial and ethnic groups. A study using 2003-2005 birth certificate data from 46 U.S states and the District of

Columbia (representing 90% of all live births in the U.S. during the study period) found 2.00 cases of SB per 10,000 live births among infants of non-Hispanic White mothers, 1.96 cases of SB per 10,000 live births among infants of Hispanic mothers, and 1.74 cases of SB per 10,000 live births among infants of non-Hispanic Black mothers [54]. A multi-site, population-based study of children born between 1997-2005 with non-syndromic SB reported that Hispanic mothers had a higher prevalence rate of each type and subtype of SB than either non-Hispanic White or non-Hispanic Black mothers [42]. Another study using 1991-2002 population-based, birth defects surveillance data from 10 U.S. regions reported that prevalence of SB among children 0 to 19 years was lowest among male and non-Hispanic Black children [55].

The mortality rate for SB without anencephaly is approximately 10%, with the majority of deaths occurring in the first year of life [56-58]. A study using 1979-2003 data from 10 U.S. birth defects registries reported one-year survival probabilities across the study period for infants with SB [59]. Results showed improvements in survival rates for all racial and ethnic categories [59]. However, differences remained in overall adjusted, one-year survival probabilities (non-Hispanic White: 96%; non-Hispanic Black: 88%; Hispanic: 93%) [59]. These results suggested differences in survival across race/ethnicity. Slightly lower mortality rates may be associated with more aggressive and early treatment of SB, including early surgical repair of SB [18, 20, 21, 56]. These studies did not address payer type and access to care, both of which could have influenced the outcomes reported.

2.2.3 Initial Management and Treatment of Infants with Spina Bifida

Fifty years ago, children born with SB received only palliative care because of lack of viable options for clinical treatment [46, 60-62]. New medical and surgical interventions now provide hope for both survival and improved quality of life and health outcomes for children born with SB [49, 63, 64].

The Spina Bifida Association's publication *Guidelines for Spina Bifida Health Care Services throughout the Lifespan* comes closest to standardized guidelines for the care of individuals with SB [65]. Postnatal surgical closure of the defect within the first 48 hours of life is the current recommended standard of treatment for SB, particularly for infants with myelomeningocele [65]. Surgical closure includes closing the opening in the spinal column as well as restoring skin and muscle tissues that cover the site [66]. Prompt closure of the site is important because it prevents infection from developing in the exposed nerves and tissues [66]. Prompt surgical repair also protects the exposed nerves and structures from additional trauma [66, 67]. Surgical closure, however, does not restore function lost because of damage to exposed neural tissue.

A growing body of research suggests an alternate prenatal surgical repair of the SB defect [68-73]. The Management of Myelomeningocele Study (MOMS) conducted a randomized control trial in 2003 to compare the safety and efficacy of prenatal surgical repair of the SB defect with that of postnatal surgical repair [68, 74]. Prenatal surgical repair was associated with a reduced need for shunt placement for treatment of hydrocephalus and improved mobility in early childhood [56, 68]. After recruiting 183 of the projected 200 participants, researchers halted the trial because of the efficacy of the prenatal surgical repair [68]. Related to my dissertation research, no Florida hospitals participated in the MOMS clinical trial [68, 74].

Prenatal surgical repair of SB may pose risks to both mother and fetus. Risks include abruption of the placenta and uterine scarring among mothers, and preterm births among infants [56, 68]. In addition, the numbers of facilities in the United States that can perform the surgery are limited [75]. Thus, the adoption of prenatal surgical repair of SB is not yet widespread [56, 76, 77]. The Spina Bifida Association recommends caution in the acceptance of a new standard of care based on a single study [75] with a sample size that is small and not representative of the U.S. population. The post-natal surgical repair of SB therefore remains the primary method of initial treatment.

2.2.4 Isolated versus Non-Isolated Spina Bifida

Birth defects may present as a single condition or may present in conjunction with other major or minor birth defects. In general, children are classified as having an isolated birth defect if they: 1) have a single major birth defect; 2) have a major birth defect and a minor birth defect; 3) have multiple major defects that affect a single organ system; or 4) have a major defect accompanied by a documented sequence of related defects and no additional unrelated major defects [78].

Similar to other birth defects, SB can present as a single condition in a newborn or can occur with other conditions diagnosed at birth or later in life. Isolated SB is SB with the single SB malformation or SB with sequential or associated malformations such as hydrocephalus, hip dislocation, or defects of the urinary system [12, 79, 80]. Isolated SB can also include SB with other minor anomalies, such as low set ears, skin tags, or bent fingers (clinodactyly) [12, 80]. Non-isolated SB is defined as SB with another major, unrelated malformation and without a syndromic diagnosis [12, 79, 80].

Approximately 15% to 25% of cases of SB are non-isolated; that is, they occur with another anomaly not related to the central nervous system defect [81, 82]. Children with non-isolated SB most commonly have orofacial clefts, cardiac defects, and abdominal wall anomalies [81, 82].

In my dissertation research, classification of each infant as having isolated or non-isolated SB was informed by discussions with expert clinical consultants from the Centers for Disease Control and Prevention's (CDC) National Center on Birth Defects and Developmental Disabilities (NCBDDD), as well as previous research [12, 79]. An expert clinical consultant from the CDC's NCBDDD manually reviewed about 15% of selected individual cases that required additional consideration because of multiple conditions. For example, patent ductus arteriosus (PDA) is a heart condition common among premature infants. If an infant with SB had a PDA, the infant was considered to have isolated SB if the infant was premature, but non-isolated SB if the infant was born at term. These and other similar situations required a case-by-case review. I referenced surveillance guidelines from the National Birth Defects Prevention Network (NBDPN) for *International Classification of Disease, Ninth revision; Clinical Modification* (ICD-9-CM) diagnostic codes for major birth defects [83]. Table A in the appendix shows a list of ICD-9-CM diagnostic codes considered as major birth defects by the NBDPN for its surveillance and research purposes.

2.2.5 Comorbidities Associated with Spina Bifida

One of the most common comorbidities associated with SB is hydrocephalus [56, 66]. Hydrocephalus is an abnormal accumulation of cerebrospinal fluid in the ventricles of the brain that can cause swelling and increased intracranial pressure [37, 66]. This

increased pressure can cause multiple central nervous system-related symptoms and may result in seizures, brain damage, and if untreated, death [56, 84]. Even when treated, hydrocephalus is associated with chronic conditions, including cognitive and developmental disabilities [47, 85-87]. Hydrocephalus is diagnosed in approximately 80-90% of children with SB whose defect type is a meningomyelocele [43, 66, 88-90] and is the leading cause of death among children with SB [21].

The National Institute of Health recognizes hydrocephalus as a serious, but under-researched health issue [37]. Few standardized protocols exist for managing hydrocephalus among children with SB [91]. Treatment for hydrocephalus typically involves the surgical placement of a ventriculoperitoneal (VP) shunt, which is tubing that travels from the ventricles of the brain into the peritoneal cavity to continually drain excess cerebrospinal fluid [43, 66, 92]. The optimal timing for shunt placement has not been established [91]. Options for timing of the shunt include 1) immediate placement in conjunction with a high spinal level of SB; 2) placement later in the child's life depending on clinical symptoms; or 3) "expectant monitoring" in the presence of asymptomatic but expanded ventricles [61, 91]. Among children with SB and hydrocephalus, approximately 15% undergo immediate VP shunt placement in conjunction with the surgical repair of their SB defect, and as many as 80-90% eventually undergo surgical placement of the VP shunt [56].

A child with SB also faces comorbidities that develop over time as an indirect result of their condition. Additional comorbidities include neuropathic bladder with urinary tract infections and chronic renal disease, orthopedic problems, chronic skin ulcerations, and obesity [46, 93-95]. A child with SB also faces challenges with

educational, psychological, and social development [47, 95-97]. Children with SB face higher risks of learning disorders and attention deficit hyperactivity disorder than children without SB, and academic challenges in secondary and postsecondary education [47, 95]. As children with SB move into adolescence and adulthood, individuals face challenges with social interaction, anxiety, and depression [47, 95, 98].

In general, individuals who have multiple comorbidities have poorer health outcomes and higher health care costs than those without comorbidities [17]. One study found that children with multiple medical conditions were eight times as likely to be high users of physician services as those without multiple conditions [99]. Another study reported that among children with special health care needs (CSHCN), those with more than one chronic condition were 27% more likely to be hospitalized in the course of a year than those with only one chronic condition [100]. Children with SB and the comorbidity hydrocephalus can experience lower scores of intelligence than children with SB only, and scores may decrease with increasing numbers of shunt revisions [47]. Researchers using 2000-2005 administrative datasets from the Children's Hospital of Alabama found that shunt revisions for hydrocephalus resulted in a median reimbursement by insurers of over $5000 per admission, thus contributing to higher health costs for these children [101].

In general, individuals who have multiple comorbidities have poorer health outcomes and higher health care costs than those without comorbidities [17]. Health resource use by individuals with multiple conditions may appear different when the conditions are examined separately as compared with examining the conditions together

[17]. Thus, a concurrent examination of comorbidities is important to understanding health resource use among individuals with more than one health condition.

2.2.6 Timeliness of Care for Children with Spina Bifida

Experts convened by the CDC helped establish public health research priorities, including timeliness of services, for selected birth defects, including orofacial clefts, craniosynostosis, congenital heart defects, and Down syndrome [33-36]. In addition to being a core component of the Institute of Medicine's quality of care framework, timely care is important in the reduction of comorbidities among children with birth defects and can contribute to improved quality of life and lower health care use and costs [12, 16, 17, 102-105].

One recent study examined timeliness of care among children with birth defects. The study reported the timeliness of primary surgical repair among Medicaid-eligible children with cleft lip and/or cleft palate who were born between 1995- 2002 in North Carolina [106]. In that study, between 58% and 90% of children with orofacial clefts had timely repair, depending on maternal demographics and other factors, such as prenatal care at a local health department and region of residence in the state [106].

Among children with SB, researchers found that timely post-natal surgical repair of SB reduces the risk of injury to the exposed neural tissues and reduces the risk of central nervous system or other infections [56]. Timely surgical repair of SB has also been associated with a reduction in the risk of comorbidities, including infection of a VP shunt [18], neurogenic bladder [16], and neurodevelopmental delays [19]. In addition, mortality rates appear lower if SB repair occurs in the first 36-48 hours of life [18, 20, 21].

However, timely care among children with birth defects remains an understudied area. To my knowledge, no peer-reviewed research exists on adherence to recommended standards of care of timely surgical repair of SB among infants with SB. Further, no studies have examined factors associated with the timing of the initial surgical repair for infants with SB. A better understanding of timely surgical repair of SB is useful because early surgical repair reduces mortality and decreases the likelihood of certain comorbidities associated with SB, such as neurogenic bladder, neurodevelopmental delays, and VP shunt infections [16, 18-21, 107].

2.3 Health Resource Use and Access to Care for Children with Spina Bifida

2.3.1 Hospitalizations and Hospitalized Days among Children with Spina Bifida

Children with special heath care needs use more health care services and have higher health care expenditures than those without special health care needs [13, 108, 109]. A study using the 2000 Medical Expenditure Panel Survey Data (MEPS) found that CSHCN had four times the number of hospital admissions and seven times the number of hospital days than those without special health care needs [13]. In addition, CSHCN accounted for over 52% of pediatric hospital days, despite accounting for only 16% of the pediatric population [13].

Researchers using the 2004 Agency for Healthcare Research and Quality (AHRQ) Healthcare Cost and Utilization Project (HCUP) Nationwide Inpatient Sample (NIS) data found that hospitalizations for birth defects were longer and more costly than hospitalizations unrelated to birth defects [8]. In the same study, SB was listed as a diagnosis in 28,300 separate hospital admissions nationwide, making it the sixth most commonly listed birth defect diagnosis among all hospitalizations [8]. Using the 2003

Agency for Healthcare Research and Quality Healthcare Cost and Utilization Project Kids' Inpatient Database (KID) data, researchers found that newborns with SB had an average length of stay for birth hospitalizations of 15.1 days, in comparison to an average hospital stay of 2.1 days for newborns with uncomplicated births [2]. Ouyang et al. used 2001-2003 national private health MarketScan Commercial claims data to examine health care expenditures of both children and adults with SB [14]. They found that the majority of children with SB had at least one hospital re-admission in their first year of life following a birth hospitalization and that the highest percent of hospitalizations per year occurred in the first year of life [14]. Another study used a population-based sample of infants born in Florida 1998-2007 to examine hospital use during infancy [110]. These researchers found that infants with SB were hospitalized an average of 2.4 times during infancy and spent an average of 25 days in the hospital during the first year of life [110]. In another study, researchers used data from the 2000, 2003, 2006, and 2009 Kids Inpatient Databases, and found that children with SB ages 1-20 years were more likely to receive inpatient care at children's hospitals or in pediatric units when compared to all children [111]. These researchers also found that the proportion of children with SB covered by Medicaid rose during the course of the study period, as did the proportion for all children in the study [111].

2.3.2 Health Care Costs among Children with Spina Bifida

The study of health care costs associated with any medical condition is complex because costs can be reported in multiple ways [112]. Health care charges refers to the fees that a health care provider requests for performance of a particular health care service [113, 114], whereas health care expenditures are actual dollars paid for health-

related services by an individual or by a public or private payer [113, 114]. Health care cost is a general term that reports the dollar amount a health care provider incurs to deliver health services [114]. Hospital charges are facility fees that do not typically include professional fees. Charges are usually higher than costs or expenditures. Costs, charges, and expenditures cannot be directly compared.

It is also important to recognize that hospital costs, charges, or expenditures do not capture the full health care economic costs associated with any medical condition. To better estimate the total cost of care for a specific medical condition, information on other direct costs are needed; these include physician fees, outpatient costs, and prescription drug costs. Including an estimate of indirect costs, such as the value of care provided by the family in the home or the value of lost parental work time, can also help to provide a more complete understanding of financial costs.

A number of studies have explored health care costs for children with SB compared with children who did not have SB [2, 8, 12-15]. Using nationally representative 2000 MEPS data, researchers found that children born with SB from birth to 18 years had medical expenditures three times those of the average for special needs children [13]. Using 2001-2003 national private health claims MarketScan Commercial data, researchers found that average medical expenditures for the first 18 years of life were thirteen times higher for a child with SB than for a child without SB [14]. In the study just described, the authors also explored changes in health care costs over the lifespan of individuals with SB and found that individuals had the highest average total expenditures during their first year of life [14]. Economic evaluations estimated lifetime direct costs of $279,000 and lifetime total costs of $636,000 for individuals with SB [115,

116]. A more recent study calculated lifetime direct costs for a child with SB of approximately $730,000 in 2010 dollars [117]. Another study used data from the Florida Birth Defects Registry and hospital discharge data to examine inpatient hospital costs during infancy for Florida children with SB born from 1998 through 2007. Researchers found that during the first year of life, children with SB had average total hospital costs of $39,000 across all payer types (in 2011 dollars) [110]. The majority of these costs occurred during the birth hospitalization [110]. Using the North Carolina birth defects registry and Medicaid data, Cassell et al. (2011) compared health care expenditures among North Carolina Medicaid-enrolled children with SB with and without hydrocephalus for different age groups, including during the first year of life, for children who were born in North Carolina 1995-2002. The authors found that infants born with SB who developed hydrocephalus had Medicaid health care expenditures 2.6 times higher than infants born with SB who did not develop hydrocephalus [12]. However, this study only examined one payer type, a public payer perspective, and only examined children from birth to five years.

Little research has explored health service use and specifically hospital resource use among children with SB in presence of common comorbidities. No research has examined the number of hospital admissions or lengths of stay for children with SB by the presence or absence of hydrocephalus or by isolated (no other coded major birth defect) vs. non-isolated SB. Further, no studies have examined these characteristics over the years of early childhood and across various health insurance payer types.

2.3.3 Access and Barriers to Health Care

The Institute of Medicine's quality of care framework includes equity in access to care as a fundamental measure of quality health care [102]. Lack of access based on inadequate health insurance coverage, race/ethnicity, educational level, or geographic barriers may result in increased morbidity and increased mortality [102, 118].

The *Healthy People 2020* program overseen by the U.S. Department of Health and Human Services included access to care in their series of Maternal-Infant-Child Health (MICH) goals [31]. MICH Goal 30.2 states that *Healthy People 2020* programs will "increase the proportion of children with special health care needs who have access to a medical home" [31]. MICH Goal 31 states that another objective of *Healthy People 2020* is to "increase the proportion of children with special health care needs who receive their care in family-centered, comprehensive, coordinated systems" [31]. Similarly, the Spina Bifida Association's 2012 Congressional Policy Agenda calls for ensured access to care for individuals with SB, especially through provision of adequate insurance [32].

Previous studies have examined five interdependent dimensions of access to care [119-121]. Those dimensions include availability, accessibility, accommodation, affordability, and acceptability [119-121]. Availability is associated with the adequacy of health care personnel, facilities, and special services [120, 121]. Accommodation describes the relationship the health care providers' organizational plans to accept clients, and clients' perceptions of the plans' suitability and appropriateness [120, 121]. Affordability addresses health care costs, clients' abilities to pay those costs, as well as clients' perceptions of the value of the costs [120, 121]. Acceptability is the relationship between attitudes of clients and providers about personal and practical characteristics that influence both seeking and providing care [120, 121]. The final dimension of access to

care is accessibility. Accessibility describes the relation between the location of the health care service or provider and the potential health care client, and examines measures such as transportation resources, travel time and distance, and travel costs [120, 121]. My dissertation research focused on the dimension of accessibility.

A number of studies have explored barriers to accessing health care for children [122-128]. Barriers to care can fall into the categories of personal barriers, financial barriers, or organizational (structural) barriers [129], generally paralleling the dimensions of access to care [121]. Personal barriers result from individual perceptions of health care need or personal health beliefs. Personal barriers can also include cultural and social influences, such as language barriers and expectations of care [127, 129]. Financial barriers occur when an individual has insufficient monetary resources or health insurance coverage to access adequate health care [108, 127, 129-131]. Organizational or structural barriers are factors related to the health care system and include such characteristics as capacity, transportation, and geographic location of resources [129, 132]. Factors in each of these three categories (personal, financial, or organizational and structural) influence the ability of an individual to access health care. The inability to access health care can result in missed or delayed opportunities for health services and can ultimately result in poorer health outcomes and higher health care costs [127].

Personal and financial barriers are commonly reported barriers to accessing care for children with or without special health care needs and include such barriers as low income, minority status, and lack of insurance [125, 130, 133, 134]. In a study of Latino children, researchers found that children of immigrant parents were less likely to have insurance and less likely to access routine health care than Latino children of parents born

in the United States [135]. Studies have found that CSHCN are particularly likely to face barriers to accessing health care [122, 124-127, 130]. Among CSHCN, adequate insurance has been reported to be the most critical determinant of access to care [127].

Less research exists on organizational or structural barriers to accessing health care, and specifically for children with birth defects, a subset of CSHCN. In one study, researchers used data from the *2001 U.S. National Household Travel Survey* and found that the average distance traveled to access medical or dental care was 10.2 road miles or 22.0 minutes [136]. This study also found that rural residents traveled 31.4% longer time to access care than residents of urban areas [136]. Specifically examining access to care for CSHCN, researchers used 2000-2002 data from the *National Survey of Children with Special Health Care Needs* and found that CSHCN who lived in rural areas were less likely to be seen by a pediatrician than children living in urban areas [128]. Another study using data from the 2005-2006 *National Survey of Children with Special Health Care Needs* found that geographic disparities existed in access to care for CSHCN in the western and northeastern regions of the United States [137].

While a number of studies have used survey data to examine access to care for CSHCN and specifically for children with birth defects, a subset of CSHCN [123, 128, 137], fewer studies have used GIS methods to examine access to care. Using a statewide, population-based birth defects registry data and geographic information system (GIS) methods, researchers found geographic disparities in access to pediatric genetic clinics among children born with major structural or chromosomal anomalies in Texas between 1999 and 2004 [138]. Using a statewide, population-based birth defects registry and survey data in North Carolina, a qualitative study of perceived barriers to care among

mothers of children with orofacial clefts born between 2001-2004 found multiple perceived barriers to accessing care, including location of services, and lack of transportation [124]. Using the same North Carolina data, researchers also found that children with orofacial clefts traveled an average of 80 miles and 92 minutes one-way, to access cleft and craniofacial care [123]. The travel distance varied by maternal education, child's age, and cleft type [123]. In another study, researchers used data from the statewide, population-based Texas birth defects registry data and GIS methods to examine mortality among infants with congenital heart disease born between 1996- 2003. These researchers found no association between increased mortality rates and home distance to a cardiac center [139]. Using linked Florida birth defects registry and hospital discharge data from 1998-2007, researchers calculated one-way travel time and distance to access hospital care for infants with SB [140]. Researchers found that 56.4% of infants traveled less than 30 minutes to access hospital care, while 22.4% traveled more than 60 minutes to access hospital care [140].

Collectively, the studies reviewed in the preceding paragraph contribute to our understanding of the role that structural barriers play in accessing to health care, especially for CSHCN, including children with birth defects. The findings of these studies suggest that geographic location is associated with the use of health care services. These studies also suggest that CSHCN may travel longer times and distances to access health care than individuals without these conditions.

Notwithstanding previous research, the role of geography remains an important and under-researched component to understanding heath care access [141]. Few studies have examined the role of geography and access to health care among children with birth

defects. Siffel et al. examined the role of GIS in birth defects surveillance, noted that 'place' is the least researched of the three epidemiological components (person, place, and time) [142] because of the challenges inherent in geographic research methods. These challenges include standardizing and defining spatial features and maintaining individual confidentiality [143]. Siffel et al. recommended an expanded and wide-ranging use of GIS in collaborative birth defects research to better understand the role of place in birth defect interventions [143]. Similarly, Kirby noted that the evaluation of the spatial component of disease occurrence, specifically intellectual and developmental disabilities, could address previously unanswered issues related to geographical distribution of incidence and prevalence, and of distribution of appropriate health care providers or health services use [144]. To date, researchers have used GIS methods to examine risk factors or geographic distribution of birth defects [145-150], but limited research has used GIS methods to examine access to care for children with birth defects [138, 140].

2.4 Framework for Study of Health Resource Use and Access

Access to care is the use of appropriate and adequate health care services and encompasses all the factors that may facilitate or hinder an individual's use of those services [151]. Access to care involves linkages to a health care provider, and also includes the assurance that the services rendered are appropriate and delivered in a timely manner [151].

Aday and Andersen's *Framework for the Study of Access to Medical Care* is a conceptual model that suggests that health policy decisions, health system characteristics, as well as predisposing, enabling, and need characteristics can be used to describe and

predict health care use [152]. Aday and Andersen suggest that health policy has a direct influence on both the characteristics of a health delivery system and on the population at risk [152]. The model suggests that health policy plays an important role in use of health services and is often the ultimate target of health services research [152].

The Aday and Andersen model is a commonly used framework for research on health care use among CSHCN. Researchers have examined child and family level variables, as well as system characteristics to explore access to care, unmet health care needs, and the economic burden of health care for CSHCN using the framework of the Andersen and Aday model [134, 137, 153, 154]. The Aday and Andersen model lends itself to research that uses surveys or administrative data to provide information on individual and system level characteristics that may influence health care use [155] and thus was selected as the framework for this dissertation research.

In the Aday and Andersen model, the health delivery system is comprised of all the components required for providing health care to consumers [152]. These components broadly include two categories of components: resource and organizational. Resource components include the facilities, workforce, equipment, and capital required to deliver services. Resources are evaluated in terms of volume and distribution. Organizational resources for health delivery are divided into entry and structure components. Entry components correspond to the concept of access and are the means by which an individual gains access to the medical care system [152]. The structural components are system characteristics that describe a health consumer's experience following entry into a health care system.

Aday and Andersen also describe characteristics that affect the population at risk. These characteristics are the predisposing, enabling, and need factors that affect an individual's heath care utilization [152, 156]. Predisposing characteristics include individual demographic characteristics as well as health values, and explain a person's propensity to use health care services. Enabling characteristics include those characteristics that facilitate an individual's use of health services and include measures such health insurance and geographic proximity to care. Need characteristics are the perceived or evaluated measures indicating that health services are required. Characteristics of the population at risk are all individual characteristics. Some of these individual characteristics may be mutable such as health insurance, while others such as ethnicity are not.

Heath policy, system, and individual characteristics are all health inputs in the Aday and Andersen *Framework for the Study of Access to Medical Care*. The outputs of this model are health care utilization and satisfaction of the health consumer. Health care utilization describes the type, location, and purpose of health care services, and time interval between use of services [152]. The final component of the model is consumer satisfaction. The satisfaction of the health consumer with the services provided represents the consumer's attitudes and perceptions of the health services they actually received. Convenience, time, personal interactions, and cost may each be dimensions in the measure of satisfaction.

For my dissertation research, I examined three of the five components of the Aday and Andersen framework: 1) characteristics of the health delivery system; 2) characteristics of the population at risk; and 3) use of health services. These components

are shown in Figure 2.2 (page 36). I modified the Aday and Andersen model to identify both characteristics of the health delivery system and characteristics of the population at risk as predisposing, enabling, and need characteristics. My primary outcomes were selected measures of the use of health services. Figure 2.3 (page 37) shows the conceptual model for my dissertation.

2.5 Summary of Background and Significance of Research

Although the numbers of children born with SB are decreasing, the severity, costs, and challenges of this birth defect continue to make it a major factor in health care economic and societal costs in the United States and for children with SB and their families. My dissertation addresses several knowledge gaps by examining hospital resource use for children with SB with and without hydrocephalus and by isolated (no other coded major birth defect) versus non-isolated SB from birth through year four. In addition, my research reports the percentage of children who have timely surgical repair of SB, and examines predisposing, enabling, and need characteristics associated with timely repair. Finally, my research uses geographic systems information to address the access to care research priorities recommended by *Healthy People 2020* and the Spina Bifida Association. I report one-way travel time and distance to access hospital care for children with SB from birth through age four.

This information contributes to our understanding of the influence of comorbidities and of predisposing, enabling, and need characteristics on hospital resource use and geographical access to hospital care among children with SB. A more complete understanding of hospital use and costs, of access to hospital services, and of timeliness

of care can contribute to improved health care service delivery and improved health outcomes for children with SB.

Table 2.1: Infants with spina bifida without anencephaly born in Florida by birth year, 1998-2007

Year of birth	Number (n=614)	Percent of study sample
1998	64	10.4
1999	81	13.2
2000	60	9.8
2001	64	10.4
2002	58	9.4
2003	51	8.3
2004	63	10.3
2005	62	10.1
2006	50	8.1
2007	61	9.9

Note: Infants in this table (n=614) had at least one matched hospital discharge record. All infants with spina bifida without anencephaly= 668.

Data sources: Florida Birth Defects Registry, 1998-2007 and hospital discharge data from the Florida Agency for Health Care Administration, 1998-2008.

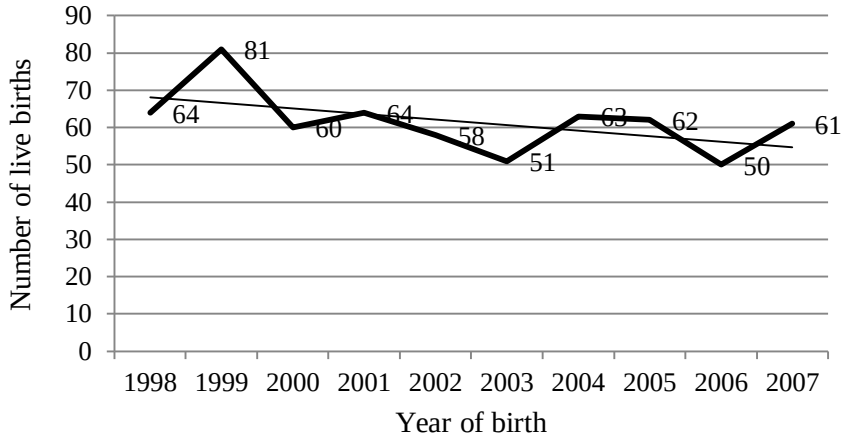

Figure 2.1 Number and trend line for Florida-born infants with spina bifida without anencephaly with at least one hospitalization initiated during first year of life, 1998-2007. (Note: Mandatory dietary folic acid fortification in U.S. began in January 1998)

Data sources: Florida Birth Defects Registry, 1998-2007 and hospital discharge data from the Florida Agency for Health Care Administration, 1998-2008.

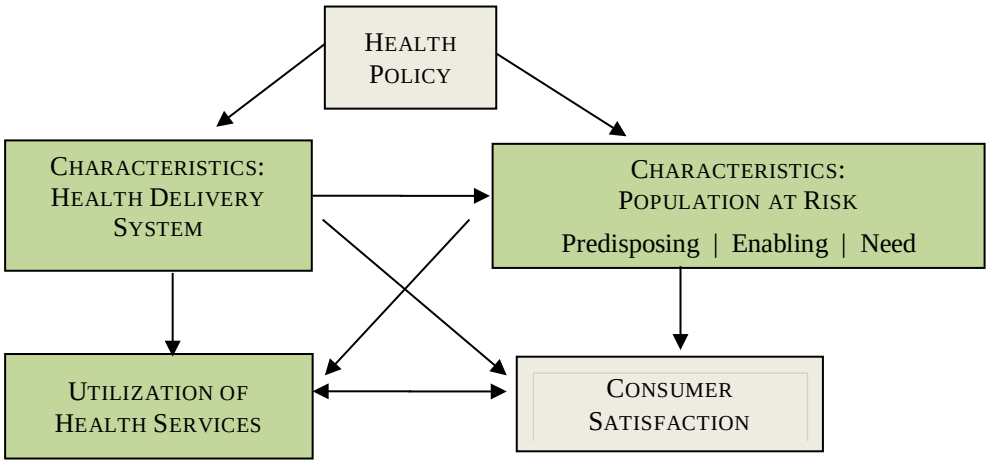

Figure 2.2 Aday and Andersen's *Framework for the Study of Access to Medical Care*

Note: boxes in green were addressed in this dissertation

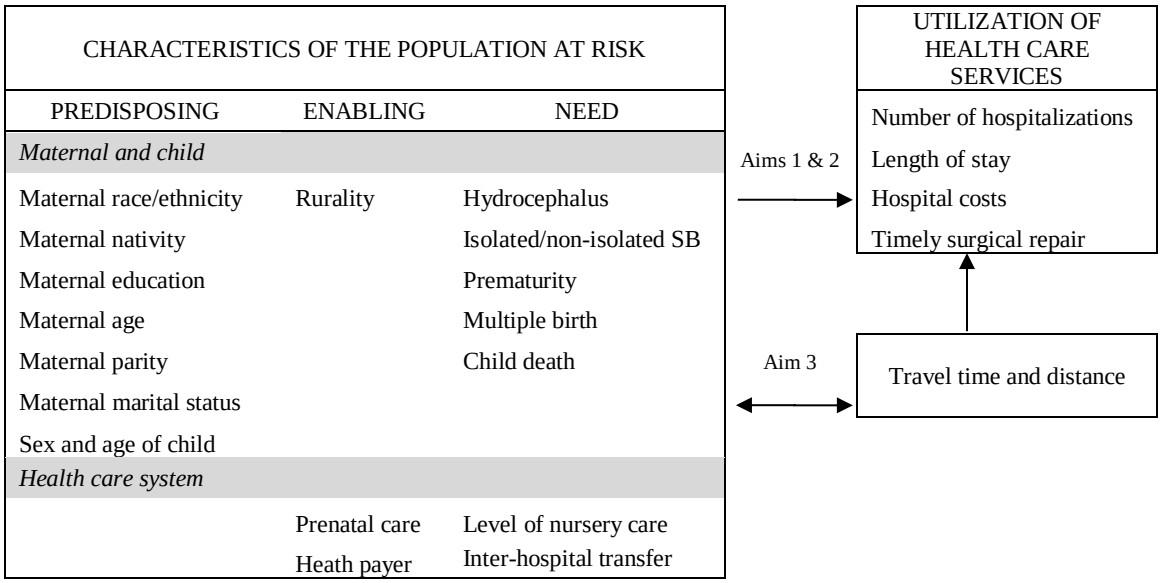

Figure 2.3 Conceptual framework for examination of hospital use, costs, and access to care among Florida-born infants with spina bifida, 1998-2007, adapted from the Aday and Andersen *Framework for the Study of Access to Medical Care*

CHAPTER 3: EFFECTS OF HYDROCEPHALUS ON HOSPITAL USE AND ASSOCIATED COSTS AMONG CHILDREN WITH SPINA BIFIDA

3.1 Introduction

Birth defects are a leading contributor to disability and pediatric hospitalizations in the United States and account for more than $2.6 billion in annual hospital costs [2, 6-8]. Comorbidities influence both hospital resource use and health care costs for children with birth defects [17, 99, 101, 157, 158]. This research focused on one type of major birth defect, spina bifida (SB), and examined the effect that hydrocephalus had on hospital use and costs during the first four years of life. Hydrocephalus is one of the most common comorbidities of SB. This study also examined predisposing, enabling, and need characteristics associated with hospital use and costs.

Findings of this study contribute to our understanding of health care resource use by children with SB. Findings identify predisposing, enabling, and need characteristics that may affect hospital use and costs. Finally, by quantifying the difference in health resource use by the presence of hydrocephalus, results from this research can inform program planning and policy development, which contributes to improved health care delivery and improved health outcomes for children with SB

3.2 Literature Review

3.2.1 Epidemiology of Neural Tube Defects and Spina Bifida

Spina bifida is a neural tube defect (NTD) that results from a failure of the caudal

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neural tube to fuse early in embryonic development [10, 11]. Spina bifida is one of the most severe birth defects compatible with life [46, 159]. Spina bifida affects a child's mobility and ability to maintain bowel and bladder control [97]. A child with SB is also at risk for comorbidities associated with SB such as hydrocephalus, neurogenic bladder and decreased renal function, orthopedic problems including scoliosis and lower limb issues, and obesity [46, 62]. A child with SB may also face challenges with educational, social, and psychological development [47] and typically requires life-long, multidisciplinary health care.

The most recent annual U.S. prevalence estimate for SB is approximately 1,500 infants or 1 in 2,858 live births per year [1]. According to the Florida Birth Defects Registry (FBDR) data, an average of 70 infants with SB were born each year in the state of Florida between 1998- 2007 [27].

3.2.2 Isolated or Non-isolated Spina Bifida

Spina bifida may present as a single condition in a newborn or it may be accompanied by other conditions diagnosed at birth or later in life. Isolated SB is SB with the single SB malformation or SB with sequential or associated malformations, such as hydrocephalus, hip dislocation, or defects of the urinary system [12, 79, 80]. Isolated SB can also include SB with other minor anomalies, such as low set ears, skin tags, or abnormally bent or curved fingers (clinodactyly) [12, 80]. Non-isolated SB is defined as SB with another major, unrelated birth defect and without a syndromic diagnosis [12, 79, 80]. Children with non-isolated SB most commonly have orofacial clefts, cardiac defects, and renal or abdominal wall anomalies [81, 82]. Approximately 15% to 25% of children with SB have non-isolated SB [81, 82, 160-162].

3.2.3 Spina Bifida and Hydrocephalus

One of the most common comorbidities associated with SB is hydrocephalus [20, 56, 66, 86]. Hydrocephalus is an abnormal accumulation of cerebrospinal fluid in the ventricles of the brain that causes swelling and increased intracranial pressure [37, 49, 56, 66, 85]. This increased pressure can cause central nervous system-related symptoms such as seizures and can cause death if untreated [56, 84]. Even if treated, hydrocephalus may be associated with chronic conditions such as seizures and cognitive and developmental disabilities [47, 85-87].

Hydrocephalus is diagnosed in approximately 80 to 90% of children with SB who have a meningomyelocele [43, 66]. Meningomyeloceles are the most common and most severe form of SB [11, 41]. Among children with SB and hydrocephalus, approximately 15% undergo immediate ventriculoperitoneal (VP) shunt placement at the same time as the repair of their SB defect [56]. As many as 80 to 90% of children eventually undergo surgical placement of a VP shunt [56].

Hydrocephalous presents additional risks to a child with SB, most notably the risks of shunt infections and shunt failures [18, 20, 66, 85, 87, 92, 111, 163]. These complications may require hospitalizations for treatment, thus increasing hospital use and costs by children with SB and hydrocephalus. Previous research found that about half of all children with SB and hydrocephalus required a shunt revision during their first year of life [20, 164]. The initial placement of a shunt increases the costs associated with SB and hydrocephalus [163]. Subsequent revisions for shunt failure or infections can further add to health care use and costs [12, 92, 101, 163].

In general, individuals who have multiple comorbidities have poorer health outcomes and higher health care costs than those without comorbidities [17]. Health resource use by individuals with multiple conditions may appear different when the conditions are examined separately as compared with examining the conditions together [17]. Thus, a concurrent examination of comorbidities is important to understanding health resource use among individuals with more than one health condition.

3.2.4 Hospital Resource Use

Previous research has found that children with special heath care needs (CSHCN) use more health care services and have higher health care expenditures compared with children without special health care needs [13, 108, 109]. A study of CSHCN using the 2000 Medical Expenditure Panel Survey Data (MEPS) reported that CSHCN had four times the number of hospital admissions and seven times the number of hospital days than children without special health care needs [13]. In addition, CSHCN accounted for over 52% of pediatric hospital days, while accounting for only 16% of the pediatric population [13].

Adequate insurance coverage is an important determinant of health resource use and may serve as a proxy for access to care [108, 127, 157, 165-167]. For CSHCN adequate health insurance may be the most critical determinant of access to health care [127]. Gaps in insurance coverage, especially multiple gaps, may also affect children's access to health care [165]. Other characteristics such as maternal age and education, maternal race/ethnicity, and nativity may also influence access to care [125, 126, 133, 135, 168, 169].

The severity of a child's medical condition is also an important factor in health resource use [99, 157]. One study found that children with multiple medical conditions were eight times as likely to be high users of physician services as those without multiple conditions [99]. Another study reported that among CSHCN, those with more than one chronic condition were 27% more likely to be hospitalized in the course of a year than those with only one chronic condition [100]. The same study found that CSHCN with poor or fair perceived health were over twice as likely to be hospitalized in the course of a year as those with perceived good health [100].

A number of studies have explored the economic health care burden for children with birth defects, a subset of CSHCN [2, 8, 12-15, 111]. Researchers using the 2004 Agency for Healthcare Research and Quality (AHRQ) Healthcare Cost and Utilization Project (HCUP) Nationwide Inpatient Sample (NIS) data found that hospitalizations for birth defects were longer and more costly than hospitalizations unrelated to birth defects [8]. These researchers found that SB was a diagnosis in 28,300 separate hospital admissions nationwide, making it the sixth most commonly listed birth defect diagnosis among all hospitalizations [8].

In the following paragraphs, I provide a brief review of studies that have examined health resource use by persons with SB. A study of 2003 Agency for Healthcare Research and Quality Healthcare Cost and Utilization Project Kids' Inpatient Database (KID) data found that newborns with SB had an average length of stay for *birth* hospitalizations of 15.1 days [2]. In comparison, newborns with uncomplicated births had an average hospital stay of 2.1 days [2]. Ouyang et al. used 2001-2003 national private health MarketScan Commercial claims data to examine health resource use of

both children and adults with SB [14]. The researchers found that the majority of children with SB had at least one hospital re-admission in their first year of life following a birth hospitalization [14]. They also found that the highest percent of hospitalizations per year occurred in the first year of life [14].

Another study used data from the 2000, 2003, 2006, and 2009 Kids Inpatient Databases to examine hospital care for individuals with SB, ages one to twenty years [111]. These researchers found that individuals with SB under twenty years of age were three times more likely to receive inpatient care at children's hospitals and twice as likely to receive care in pediatric units compared with all children [111]. They also found that the most common reason for readmission was repair of a malfunctioning shunt for hydrocephalus [111]. In addition, the authors found that the proportion of children with SB covered by Medicaid rose from 45.1% in 2000 to 49.7% in 2009, similar to findings for the entire study sample [111].

Other researchers examined nationally representative 2000 Medical Expenditure Panel Survey (MEPS) data and found that children with SB ages birth to 18 years had medical expenditures three times those of the average for special needs children [13]. Another study used 2001-2003 national private health claims MarketScan Commercial data and found that average medical expenditures for the first 18 years of life were thirteen times higher for a child born with SB than for a child born without SB [14]. In the same MarketScan study, researchers explored changes in health care costs over the lifespan of individuals with SB. They found that individuals with SB incurred an average of $49,602 in total expenditures during the first year of life compared with $15,911 average yearly expenditures for ages 0-64 years [14].

Based on nationally weighted data from the Agency for Healthcare Research and Quality (AHRQ) Healthcare Cost and Utilization Project (HCUP) 2003 Kids' Inpatient Database (KID), the mean hospital charges per neonatal admission for infants with SB was $65,342 [2]. In comparison, the mean hospital charge per neonatal admission for uncomplicated births was $1,844 [2]. Economic evaluations estimated lifetime direct costs (primarily medical) associated with SB at $279,000 per individual with SB, and lifetime total costs were estimated at $636,000, both reported in 2002 dollars [115, 116]. A recent study reported the lifetime direct costs for a child with SB of approximately $730,000 in 2010 dollars [117].

Few studies describe health service use among children with SB in the presence of common comorbidities. One study used the North Carolina Birth Defects Registry and Medicaid data to compare health care expenditures among North Carolina Medicaid-enrolled children with SB with and without hydrocephalus for children birth through five years who were born between 1995 and 2002 [12]. The authors found that infants with SB who developed hydrocephalus had Medicaid health care expenditures 2.6 times higher than infants born with SB who did not develop hydrocephalus [12]. However, this study only examined expenditures for children insured by Medicaid.

Gaps remain in our understanding of this economic burden. To my knowledge, no peer-reviewed research has examined the number of hospitalizations, lengths of stay, and associated costs for children with SB with and without hydrocephalus and by isolated versus non-isolated SB. Further, no studies have compared these factors during infancy and childhood, and across various health insurance payer types.

3.3 Study Objectives and Hypotheses

The purpose of this study was to examine the effect of hydrocephalus and other selected predisposing, enabling, and need characteristics on hospital use and costs among children with SB. The research questions were:

1. What are the differences in number of hospitalizations, lengths of stay, and associated costs among children with SB by the presence of hydrocephalus, age group, and payer type?
2. What predisposing, enabling, and need characteristics are associated with number of hospitalizations, lengths of stay, and costs among children with SB by the presence of hydrocephalus?

I hypothesized that children with SB and hydrocephalus would incur greater costs, have more hospitalizations, and spend more days in the hospital compared to children with SB without hydrocephalus, and that highest hospital use for all children with SB would be during infancy (from birth through one year) [12, 14, 17, 157].

Finally, I hypothesized that differences in hospital use and costs would vary based on predisposing characteristics (maternal nativity [135, 169, 170], maternal race/ethnicity, age, educational level, and marital status [125, 126, 168, 169, 171]), enabling characteristics (adequacy of prenatal care [132] and health care payer source [108, 125, 134, 157]), and need characteristics (preterm birth [172-174], isolated or non-isolated SB, and the presence of hydrocephalus).

3.4 Conceptual Framework

The Aday and Andersen *Framework for the Study of Access to Medical Care* provided the conceptual basis for this project [152, 156, 175, 176]. I adapted the Aday

and Andersen model to the research questions presented above, and included the specific predisposing, enabling, and need variables used in each component of the model. This adapted model is shown in Figure 2.3 (Chapter 2, page 37).

3.5 Study Design and Methods

3.5.1 Study Design

This study was a retrospective, statewide, population-based cohort analysis of inpatient hospital use and costs for children with SB from birth through four years born in Florida between January 1, 1998, and December 31, 2007.

3.5.2 Data Acquisition and Study Sample

I obtained data for this study from linked, longitudinal datasets provided by the FBDR and the Florida Bureau of Vital Statistics, both in the Florida Department of Health (FDOH), and the Florida Agency for Healthcare Administration (AHCA). The Florida AHCA provided the hospital discharge data. Infants with SB without anencephaly born in Florida between 1998 and 2007 were identified using the *International Classification of Disease, Ninth revision; Clinical Modification* (ICD-9-CM) codes 741.00-741.93. Hospital discharge data from January 1, 1998 through December 31, 2008, were used to allow for at least one year of hospitalizations for each infant with SB.

Included infants were live-born in Florida to a mother who was a Florida resident at the time of delivery. Infants who were adopted or prospectively adopted or who were born out of state were excluded by the FBDR. Included infants also matched with an inpatient hospital discharge record during the first year of life. Children who died at any point in the study period were included to capture the full extent of cost and hospital use

associated with the care for children with SB. Death was included as a control variable in the analysis. Figure 1.1 (Chapter 1, page 9) shows the process for identification of infants for the final study sample.

3.5.3 Primary Outcomes of Interest: Number of Hospitalizations, Hospital Costs, and Lengths of Stay

I examined three outcomes of interest related to the use of hospital resources: 1) number of admissions, 2) hospital costs, and 3) lengths of stay. I examined these outcomes for the birth hospitalization, for post-birth hospitalizations during infancy, across all infancy, and for ages one to four years.

3.5.3.1 Outcome of Interest #1: Number of Hospitalizations

The first outcome of interest was the number of hospitalizations per child. I reported total number of hospitalizations during infancy (birth through age one) and during ages one to four years separately. I also categorized hospitalizations during the first year of life as either birth hospitalizations or post-birth hospitalizations. I made this distinction because previous work found that birth hospitalizations among infants with SB have notably higher charges than subsequent infancy hospitalizations [14]. For birth hospitalizations, by definition, the number of hospitalizations was always one. For other ages categories of number of hospitalizations (post-birth, all infancy, or ages one to four years), the number of hospitalizations was the total number of hospitalizations a child experienced during the respective period.

I defined a hospitalization as a single episode of hospital care, whether or not the hospital admission included an accompanying inter-hospital transfer [177]. If hospital discharge records showed that an infant was admitted to a hospital on the same day the

infant was discharged from another hospital, the two admissions were merged into one hospitalization. If a one-day difference existed between a discharge from one hospital and an admission to another hospital and the records included a "transfer" code, the two admissions were also merged into one hospitalization. This definition provided a more accurate picture of a hospital experience and reduced the number of single day admissions in one facility that would result in lower average costs and lengths of stay [177].

3.5.3.2 Outcome of Interest #2: Hospital Costs

The second outcome of interest was total hospital costs. For birth hospitalizations, hospital costs were those costs incurred during the birth hospitalization only. For other age categories of hospital costs (post-birth, all infancy, or ages one to four years), the costs are the total costs a child incurred during the respective period.

I converted the total estimated hospital charges obtained from the Florida AHCA dataset to total estimated hospital costs, using the 2010 average all-payer inpatient hospital cost-to-charge ratio for the state of Florida, provided by AHRQ [178]. The average all-payer inpatient hospital cost-to-charge ratio among Florida hospitals ranged from 0.355 in 2001 (n=209 hospitals reporting) to 0.294 in 2008 (n=217 hospitals reporting). This average all-payer cost-to-charge ratio suggested that hospitals' costs averaged approximately 29-36% of the amount those hospitals billed to health care payers between 2001 and 2008 [178]. Because 2001 was the earliest year of data available, I used the cost-to-charge ratio for 2001 (0.355) to convert inpatient charges to estimated costs for the years 1998-2001. I then adjusted total estimated hospital costs to 2012 dollars using hospital industry data from the Producer Price Index, U.S. Department

of Labor [179]. I used the Producer Price Index, instead of the Consumer Price Index, because it measures real output and excludes services, imports, sales taxes, and distribution costs [180].

3.5.3.3 Outcome of interest #3: Length of stay

The third outcome of interest was length of stay. The Florida AHCA provided the length of stay for each admission for each child in the AHCA dataset. I measured length of stay in days. If a child had a reported hospitalization and associated charges, but a zero-day length of stay, I re-coded length of stay to one-day. Twenty-four infancy hospitalizations and 18 hospitalizations during ages one to four years had a zero-day length of stay. These 42 hospital admissions were re-coded to a one-day length of stay.

For birth hospitalizations, length of stay was the number of hospitalized days for the birth hospitalization only. For other age categories of hospitalizations (post-birth, all infancy, or ages one to four years), the length of stay was the total number of days a child was hospitalized during that period. I referred to length of stay as number of hospitalized days when referring to aggregate hospitalizations.

3.5.4 Primary Exposure of Interest: Hydrocephalus

Hydrocephalus, a need characteristic, was the primary exposure of interest. The presence of hydrocephalus was based on administrative coding and was not clinically verified. I identified hydrocephalus by the ICD-9-CM codes 741.01- 741.03 from the FBDR dataset. The selection of these ICD-9-CM codes for identification of hydrocephalus was informed by discussions with several expert clinicians from the Centers for Disease Control and Prevention's (CDC) National Center on Birth Defects and Developmental Disabilities (NCBDDD). I reported the presence of hydrocephalus as

a dichotomous variable. In selected analyses, I also stratified hydrocephalus by isolated or non-isolated SB. Thus, for selected analyses, I reported the variable "spina bifida and hydrocephalus" as "isolated SB with hydrocephalus", "non-isolated SB with hydrocephalus", "isolated SB without hydrocephalus", and "non-isolated SB without hydrocephalus."

3.5.5 Stratification by Age of Child

I stratified findings for birth through four years into two categories. First, I reported findings for all hospitalizations that were initiated when the child was <365 days old. I refer to this category as infancy. I created infancy as a separate category because the highest health care use and costs typically occur during the first year of life for children with SB [14]. I then collapsed outcomes for age one through four years into a separate, single age category to maintain an adequate sample size for meaningful results. I identified hospitalizations for age one through four years as hospitalizations that began when the child was between 365 and 1823 days old. I obtained the age of the child in days from the Florida AHCA dataset "time to admission" variable.

3.5.6 Covariables Measuring Predisposing, Enabling, and Need Characteristics

I categorized additional covariables as predisposing, enabling, and need characteristics. These characteristics corresponded to the components of the Aday and Andersen conceptual model. Consistent with the Aday and Andersen conceptual model, the following characteristics were considered predisposing characteristics: maternal age, maternal race/ethnicity, maternal nativity, parity, marital status, maternal education, and child's age and sex. Enabling characteristics included the variables that measured adequacy of prenatal care, residential rurality, and health insurance payer. In addition to

hydrocephalus, need characteristics included isolated or non-isolated SB, preterm birth, level of nursery care at the birth hospital, an inter-hospital transfer during the birth hospitalization, and death. I describe these variables and their coding below.

3.5.6.1 Predisposing Characteristics of Mothers and Children

Predisposing characteristics of mothers included maternal race/ethnicity, maternal age, education, maternal nativity, and marital status. These data were obtained from the FBDR and Florida vital statistics. I calculated maternal parity by adding the number of live born children still living and those live born but now deceased, as reported in the FBDR data.

Predisposing characteristics of the child were sex and age. I obtained the sex of the child from the FBDR data. I calculated the child's age in years using the "time to admission" variable in the Florida AHCA data, which was reported in days.

3.5.6.2 Enabling Characteristics of Mothers and Children

Enabling characteristics included adequacy of prenatal care and rurality of maternal residential address, which were obtained from Florida vital statistics data. Expected health insurance payer, another enabling characteristic, was obtained from the Florida AHCA data. I identified adequacy of prenatal care using the Kotelchuck Index. The Kotelchuck Index creates a ratio comparing the month in which prenatal care was initiated with the total number of prenatal visits prior to delivery to calculate four categories of prenatal care: inadequate (less than 50% of expected visits), intermediate (50-79%), adequate (80-109%), and adequate plus (110% or more) [181, 182]. The Kotelchuck scoring system considers scores less than 80% to be inadequate care [181, 182]. For the purpose of this research, based on an examination of the data, and to ensure

adequate cell sizes for meaningful results, I reported adequacy of prenatal care as a binary variable. I used the Kotelchuck cut point of 80% to classify adequate and adequate plus care as "adequate prenatal care," and intermediate and inadequate care as "inadequate prenatal care."

I identified maternal residential rurality by comparing the geocoded maternal residential addresses reported at birth with the 2000 U.S. Census data that reported rurality by block group level. In the 2000 U.S. Census, the Census Bureau defined "urban" as all territory, population, and housing units located within an urbanized area or in an urban cluster [183]. Urban areas and urban clusters were described by the U.S. Census Bureau as densely settled areas consisting of core census block groups or blocks that had a population density of at least 1,000 people per square mile and surrounding census blocks that had an overall density of at least 500 people per square mile [183]. The U.S. Census Bureau defined all territory, population, and housing units located outside of urban areas or clusters as "rural" [183]. The U.S. Census Bureau assigned a designation to each census block group identifying the geographic area as urban, an urban cluster area, or as rural. Following consultation with spatial research experts at the University of North Carolina at Charlotte's (UNC Charlotte) Department of Geography and Earth Sciences and examination of the data, I collapsed urban and urban cluster designations into a single "urban" category. I reported maternal residential "rurality" as a dichotomous variable, "urban" or "rural," to ensure adequate cell sizes for meaningful results. As described in published research, the FDOH conducted the initial geocoding of the maternal residential addresses reported at birth [140]. Researchers in UNC

Charlotte's Department of Geography and Earth Sciences improved the match of the initial geocoding to ultimately geocode 99.7% of the maternal addresses [140

I classified health insurance payers in two ways using hospital discharge data from the Florida AHCA. First, I classified payers for the birth hospitalization as: 1) public, 2) private, or 3) self-insured, under-insured, or no insurance. Second, I classified "payer type" across infancy. "Payer type" was classified as: 1) public payers only for all hospitalizations; 2) private payers only for all hospitalizations; 3) self-insured, under-insured, or no insurance only for all hospitalizations; or 4) multiple payer types. Multiple payer types indicated that a child had different types of health insurance coverage across more than one hospitalization; for example, a private health insurance payer covered one hospitalization and a public payer covered a subsequent hospitalization. Public payer sources included Medicare, Medicaid, KidCare (Florida's state children's health insurance program), and the Veterans Administration. Private payer sources included private or employer-based insurance, including military coverage (CHAMPUS or TriCare). The self-pay, no insurance or under-insured category was defined by the Florida AHCA as either no third party coverage or less than 30% estimated insurance coverage [184]. I did not report payer type for ages one to four years because children may have had multiple types of payers across the four-year period. For example, some children had a consistent payer for all four years, while others had one or more changes in payer types. These changes made it difficult to characterize each child by a single, meaningful payer type for ages one to four years.

3.5.6.3 Need Characteristics of Mothers and Children

In addition to hydrocephalus, need characteristics included isolated or non-isolated SB, preterm birth (less than 37 weeks gestation), low birth weight (less than or equal to 2500 grams), plurality, and death. Data for these variables were obtained from the FBDR and Florida vital statistics data. I reported SB for each infant as a dichotomous variable, isolated or non-isolated. I also stratified hydrocephalus by isolated or non-isolated SB for selected analyses. Infants were classified as having isolated SB if they met any of the following criteria: 1) had only the SB birth defect; 2) had the SB defect and another minor birth defect associated or not associated with SB, such as low set ears or skin tags; or 3) had the SB defect accompanied by a documented sequence of coded defects related to SB and no additional unrelated, coded major defects [79, 80], as verified by a clinical expert at the CDC's NCBDDD. Classification of isolated or non-isolated SB was informed by discussions with expert clinical consultants from the CDC's NCBDDD, as well as previous research [12, 79]. An expert clinician from the CDC's NCBDDD manually reviewed approximately 15% of the study sample that required additional consideration because of multiple conditions. For example, patent ductus arteriosus (PDA) is heart condition common among premature infants. If an infant with SB had a PDA, the infant was considered to have isolated SB if the infant was premature, but non-isolated SB if the infant was born at term. These and other similar situations required a case-by-case review. I referenced surveillance guidelines from the National Birth Defects Prevention Network (NBDPN) for ICD-9-CM diagnostic codes for major birth defects [83]. Table A in the appendix lists ICD-9-CM diagnostic codes considered as major birth defects by the NBDPN for its surveillance and research purposes.

I also considered the level of nursery care at the birth hospital as a *need* characteristic. The American Academy of Pediatrics classifies level of nursery care at the birth hospital as Level I, II, or III [185, 186]. Level III nursery care provides the most sophisticated care for complex cases [185, 186]. I reported the level of hospital nursery care for the birth hospitalization, even if an infant was transferred at birth to a hospital with a higher level of nursery care. I defined a birth hospitalization as a first hospitalization with age at admission of zero days or a first hospitalization with an age at admission of one day with an accompanying indication of hospital transfer [177]. I used the level of nursery care only in analyses that examined birth hospitalizations.

In addition, I considered an inter-hospital transfer at birth as a need characteristic because infants who are transferred at birth typically have more serious or complex medical conditions that require services that are not available at the birth hospital. Inter-hospital transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital, or if a one-day difference existed between a discharge from one hospital and an admission to another hospital and the infant's records included an indication of a transfer [177]. Only inter-hospital transfers that occurred during the birth hospitalization were included in the analysis. I did not include later transfers because the data did not include information on hospital level of pediatric care, a designation that could have helped identify need. In addition, transfers later in life could have been return trips to local hospitals for continued care or for other medical reasons beyond the scope of this dissertation research. I coded inter-hospital transfers as a dichotomous variable, transfer or no transfer.

Finally, I reported the child's death, which was categorized as no death as of December 31, 2008, neonatal death (death at <28 days), death during infancy (< 365 days), or death following infancy (≥365 days) [187]. For multivariable analyses, I collapsed the death categories into a dichotomous variable, death or no death. I collapsed the variable to ensure that cell sizes were adequate for meaningful results. All reported deaths occurred within the study period of January 1, 1998 through December 31, 2008.

3.5.7 Statistical Analyses

I conducted descriptive analyses for the predisposing, enabling, and need characteristics of the study population and health system. I reported the mean, median, and range for total hospital costs, total number of hospitalizations, and total lengths of stay respectively for birth hospitalizations, all post-birth hospitalizations during infancy, all infancy hospitalizations, and all hospitalizations during ages one to four years.

I used bivariate analyses to examine number of hospital admissions, hospital costs, and lengths of stay by the presence or absence of hydrocephalus, by isolated or non-isolated SB, by payer type, and by other predisposing, enabling, and need characteristics. I examined outcomes for hospitalizations during infancy and for ages one to four years. I also examined birth and post-birth hospitalizations during infancy separately, because previous research suggested infants with SB use more hospital resources during birth hospitalizations than during later hospitalizations [14]. Chi-square analyses were conducted on the categorical variables to determine significance level using a p-value of <0.05. Where appropriate, I used Fisher's exact test to account for small cell sizes, using a p-value of <0.05 to determine statistical significance. Because of

the skewness of the data for hospital use and costs, I conducted Wilcoxon Rank Sums tests to determine significance level using a p-value of <0.05.

For the multivariable analyses, I used Poisson regression and ordinary least squares (OLS) or generalized linear model (GLM) procedures, depending on the type and distributional qualities of the data [188]. To examine the number of hospitalizations, I used Poisson regression because the outcomes were finite count data. Costs and lengths of stay presented analytic challenges because the variables were positively skewed. This is a common challenge when analyzing health care expenditures, length-of-stay, and utilization of health care data [188]. For costs and lengths of stay, I applied a method described by Manning and Mullahy [188] to evaluate characteristics of the data, apply recommended algorithms, and selected the method most appropriate for estimating each model. For analyses of lengths of stay for post-birth hospitalizations and for those during ages one to four years, I used a generalized linear model (GLM) procedure with a Poisson distribution. For all other models, I used an ordinary least squares (OLS) procedure for the log of the outcome, using a normal distribution and robust standard error option. These calculations ensured more accurate effect estimates by increasing precision and reducing bias [188]. For the multivariable analyses, I reported effect estimates as unadjusted prevalence ratios (uPR), adjusted prevalence ratios (aPR) or the log-transformed exp (β) and corresponding 95% confidence intervals (CI) to determine if the selected predisposing, enabling, and need factors were associated with hospital use and costs. I reported exp (β) values when the continuous outcomes were log-transformed for analysis.

I constructed models for: 1) hospital costs and length of stay for birth hospitalizations; 2) total number of admissions, total hospital costs, and total lengths of stay for post-birth infancy hospitalizations; 3) total number of admissions, total hospital costs, and total lengths of stay for all infancy hospitalizations; 4) total number of admissions, total hospital costs, and total lengths of stay for all hospitalizations ages one to four years. I did not examine number of hospitalizations for birth hospitalizations because, by definition, that number was always one.

The goal of the multivariable analyses was to arrive at models that were theory-based, informed by previous research, and parsimonious given the relatively small sample size; thus, selected predisposing, enabling, and need covariables were included in the final regression model. I excluded parity because no theory or previous research supported its inclusion. I excluded low birth weight because of its close correlation with preterm birth. I excluded plurality because too few infants were part of multiple births to contribute meaningfully to the results. I excluded transfers because of their correlation with level of nursery care in the birth hospital.

My final models included the following variables: predisposing characteristics: maternal age, maternal race/ethnicity, maternal nativity, marital status, maternal education, and child's sex and age; enabling characteristics: adequacy of prenatal care, residential rurality, and health care payers; need characteristics coded hydrocephalus (the primary outcome of interest), isolated or non-isolated SB (reported separately and by presence of hydrocephalus), preterm birth, level of nursery care, and death.

I conducted three sensitivity analyses to observe for differences in selected characteristics among the study sample. First, I examined for differences between infants

who a linked inpatient hospital discharge record and those who did not. Second, I examined for differences between infants who had a recorded birth hospitalization and those who did not. Third, I examined for differences in characteristics between infants who experienced an inter-hospital transfer as part of their birth hospitalization and infants who did not. Infants with no linked discharge records, no birth hospitalization, or infants who were transferred may have been different in terms of their demographic characteristics, clinical experiences, or severity of medical conditions compared with other infants in the study population.

I assessed individual variables used in the multivariable analysis for multicollinearity. There was no evidence of notable multicollinearity based on recommended maximum levels of the variance inflation factor [189, 190].

All analyses were conducted using SAS 9.2 statistical software (SAS Institute, Inc., Cary, NC). This study was approved by the Institutional Review Boards at UNC Charlotte, the FDOH, and the CDC's NCBDDD.

3.6 Results

3.6.1 Selection of the Sample

The FBDR data identified 914 Florida-resident infants who were born between January 1, 1998 and December 31, 2007 with an ICD-9-CM code indicating an NTD. Of these 914 infants, 668 had ICD-9-CM codes for SB without anencephaly. Of the 668 infants with SB, 614 were successfully linked to at least one inpatient hospital discharge record. These infants comprised the sample for analysis. Figure 1.1 (Chapter 1, page 9) shows the process for selecting the study sample.

Infants who did not have a linked hospital discharge record and were in the FBDR were more likely to be born to mothers who were less educated (p=0.0011) and foreign-born (p<.0001), and of Hispanic ethnicity (p= 0.0044) than infants who matched with a hospital discharge record (results not shown). There were no significant differences in maternal age, marital status, infant's sex and birth weight, or death between infants who matched and did not match to hospital discharge records (results not shown).

Of the 614 infants who matched to hospital discharge records, 569 infants had a first hospitalization recorded in the Florida AHCA dataset that was also the infants' birth hospitalization. Infants with no birth hospitalization recorded in the Florida AHCA data may have been born at a Florida hospital that did not report data to the Florida AHCA, such as a military hospital or a birthing center. They may also have been born at home, although home births are relatively rare (less than 1% of births nationally) [191]. Infants in the FBDR who did not have a birth hospitalization were more likely to be born to Hispanic mothers (p=0.0272) who were rural residents (p=0.0206) and who were not born in the United States (p=0.0008) (results not shown). I found no significant differences in maternal age, maternal education, marital status, and infant's sex and birth weight, and the presence or absence of hydrocephalus between infants with and without a birth hospitalization (results not shown). Among infants without a birth hospitalization, 64.4% had hydrocephalus (n=29) (results not shown).

3.6.2 Descriptive Results

3.6.2.1 Descriptive Characteristics of the Mothers and Children

Tables 3.1 and 3.2 show selected descriptive characteristics of the mothers and infants in this study (n=614). About 53% (n=323) of infants were born to non-Hispanic

White mothers. Most mothers were born in the United States (75.9%, n=466), had at least a high school diploma (76.2%, n=468), and had received "adequate" prenatal care (72.8%, n=447). About 60% of mothers were married (n=368). The majority of mothers lived in urban or urban cluster areas (85.5%, n=525).

About 20% (n=121) of infants were born low birth weight and 26.5% infants were born preterm (n=163). About 57% of infants had hydrocephalus (n=349); 25.4% of infants had non-isolated SB (n=156). Examining hydrocephalus by isolated or non-isolated SB, 42.4% of infants had isolated SB with hydrocephalus (n=260); 14.9% had non-isolated SB with hydrocephalus (n=89). About 32% of infants had isolated SB without hydrocephalus (n=198); 10.9% had non-isolated SB without hydrocephalus (n=67). Just under 9% of the children died at any point during the study period (n=53), with the majority of deaths occurring during infancy (6.7%, n=41).

About 24% of infants were transferred to another hospital during their birth hospitalization (n=146). Infants who had an inter-hospital transfer were more likely to be born to a mother who was born in the United States (p=0.0324) and to have non-isolated SB (p=0.0012) compared with infants who were not transferred. Infants born at a hospital with Level III nursery care were less likely to be transferred at birth (p=0.0003). There were no differences between infants who were transferred and those who were not, based on maternal race/ethnicity, maternal age and education, maternal nativity or marital status, or on infant's sex or gestational age (results not shown).

3.6.2.2 Descriptive Results for Number of Hospitalizations

Table 3.3 shows descriptive results for number of hospitalizations for birth hospitalizations, all post-birth hospitalizations during infancy, all infancy

hospitalizations, and all hospitalizations during ages one to four years. The average number of hospitalizations across all infancy was 2.4 (SD 1.7; median: 2.0; range: 1-12). Infants with more than one hospitalization during infancy had an average of 2.2 hospitalizations following their birth hospitalization (standard deviation, SD: 1.7; median: 2.0; range: 1-11). During ages one to four years, children with SB had an average total of 3.0 hospitalizations (SD 3.0; median; 2.0; range: 1-19).

3.6.2.3 Descriptive Results for Hospital Costs

Table 3.3 also shows descriptive results for hospital costs for birth hospitalizations, all post-birth hospitalizations during infancy, all infancy hospitalizations, and all hospitalizations during ages one to four years. Total costs across all infancy averaged $47,884 (standard deviation, SD $86,934; median; $26,825; range: $124-1,590,268). The average cost of a birth hospitalization (mean: $30,557; SD: $52,148; median: $18,789; range: $124-706,793) was 2.3 times higher than the average cost for a post-birth hospitalization (number of hospitalizations: 2.2; total mean: $29,592; SD $64,931; total median: $11,286; total range: $720-883,476; mean for a single post-birth hospitalization: $13,450). Total costs for all hospitalizations during ages one to four years averaged $30,483 (SD $57,427; median; $11,593; range: $304-505,528). The average cost per hospitalization for children with SB ages one to four years was $10,161 or 24% less than a post-birth hospitalization during infancy.

3.6.2.4 Descriptive Results for Length of Stay

Finally, Table 3.3 shows the descriptive results for lengths of stay for birth hospitalizations, all post-birth hospitalizations during infancy, all infancy hospitalizations, and all hospitalizations during ages one to four years. Infants were

hospitalized for an average of 17.2 days for their birth hospitalization (standard deviation, SD: 23.2; median: 10.0; range: 1-221). Post-birth hospitalizations averaged 14.2 total days (SD 24.7; median: 5.0; range: 1-255). Average total number of hospitalized days across all infancy was 25.3 days (SD 34.4; median: 14.0; range: 1-476). The average total number of hospitalized days for a child during ages one to four years was 14.8 days (SD 26.8; median: 6.0; range: 1-206).

3.6.3 Bivariate Results for Primary Exposure of Interest Hydrocephalus

In response to research question one, the following sections describe the results for the number of hospitalizations, lengths of stay, and associated costs for children with SB. Based on the research question, I report the findings by the presence of hydrocephalus, by age group, and by payer type.

3.6.3.1 Bivariate Results for Number of Hospitalizations by Hydrocephalus and Age Group

Table 3.3 further shows hospital use and costs by the presence of coded hydrocephalus. Infants with hydrocephalus were hospitalized significantly more often than those without hydrocephalus. During the first year of life, infants without hydrocephalus had an average total of 1.9 hospitalizations (SD; 1.4; median: 2.0; range: 1-12). Infants with hydrocephalus had an average total of 2.7 hospitalizations (SD; 1.9; median: 2.0; range: 1-12) ($p<0.0001$). Examining all post-birth infancy hospitalizations, infants without hydrocephalus were hospitalized an average total of 1.7 times (SD: 1.4; median: 1.0; range: 1-11). Infants with hydrocephalus were hospitalized an average of total of 2.5 times (SD: 1.9; median: 2.0; range: 1-11) ($p<0.0001$).

During ages one to four years, children with SB and hydrocephalus also had significantly more hospitalizations compared with children without hydrocephalus. Children without hydrocephalus averaged 2.4 total hospitalizations during ages one to four years (SD: 2.8; median: 1.0; range: 1-15). Children with hydrocephalus had an average total of 3.2 hospitalizations (SD: 3.1; median: 2.0; range: 1-19) ($p=0.0023$).

Table 3.4 shows the results for number of hospitalizations for infants with hydrocephalus, stratified by isolated vs. non-isolated SB. In general, infants and children with simpler presentations of SB (isolated SB, without hydrocephalus, or both) were hospitalized significantly fewer times during infancy and ages one to four years than children with more complex presentations of SB (non-isolated SB, with hydrocephalus, or both). Infants with isolated SB and no hydrocephalus were hospitalized least often, 1.7 times (SD: 0.8; median: 1.5; range: 1-6). Infants with non-isolated SB and hydrocephalus were hospitalized most often, 3.0 times (SD: 2.5; median: 2.0; range: 1-12) ($p<0.0001$). During ages one to four years, children with isolated SB and no hydrocephalus were hospitalized least often, 1.7 times (SD: 1.9; median: 1.0; range: 1-12); infants with non-isolated SB and hydrocephalus were hospitalized most often, 4.0 times (SD: 3.1; median: 3.0; range: 1-13) ($p<0.0001$).

3.6.3.2 Bivariate Results for Hospital Costs by Hydrocephalus and Age Group

Table 3.3 also shows hospital costs by the presence of coded hydrocephalus for hospitalizations for all infancy hospitalizations and for all hospitalizations during ages one to four years. Infants with hydrocephalus incurred significantly higher hospital costs compared with infants without hydrocephalus. During the first year of life, average inpatient costs for infants with hydrocephalus were 53.4% higher than average costs for

infants without hydrocephalus [mean (median) with hydrocephalus: $56,345 ($38,253); mean (median) without hydrocephalus: $36,742 ($14,838); p<0.0001].

I found the same pattern of higher costs among infants with hydrocephalus comparing birth and all post-birth hospitalizations. Infants with hydrocephalus incurred average costs for their birth hospitalizations that were 51.5% higher than the cost for infants without hydrocephalus [mean (median) cost with hydrocephalus: $35,884 ($27,491); mean (median) cost without hydrocephalus: $23,711 ($6,615); p<.0001]. Similarly, infants with hydrocephalus incurred 29.1% higher average costs for all post-birth hospitalizations compared with infants without hydrocephalus [mean (median) with hydrocephalus: $32,338 ($13,787); mean (median) without hydrocephalus: $25,050 ($8,775); p=0.0006].

For all hospitalizations during ages one to four years, children with hydrocephalus had significantly higher hospital costs compared with children without hydrocephalus, although the differences were smaller. The costs for children with hydrocephalus were 5.9% higher than children without hydrocephalus [mean (median) cost with hydrocephalus: $30,902 ($13,095); mean (median) cost without hydrocephalus: $29,177 ($9,223); p=0.0181].

Table 3.4 shows the results for costs by the presence of hydrocephalus stratified by isolated vs. non-isolated SB. Infants and children with simpler presentations of SB (isolated SB, without hydrocephalus, or both) had significantly lower hospital costs than those who had more complex presentations of SB (non-isolated SB with hydrocephalus). Infants with isolated SB and no hydrocephalus had the lowest average total costs (mean: $18,637; median: $11,974). Costs for infants with non-isolated SB without

hydrocephalus (mean: $90,247; median: $36,787) were three to four times higher than costs for infants with isolated SB and no hydrocephalus (p<.0001). During ages one to four years, average total costs for children with non-isolated SB without hydrocephalus were twice as high as total costs for children with isolated SB and no hydrocephalus ($42,423 versus $20,584, respectively; p<0.0010).

3.6.3.3 Bivariate Results for Length of Stay by Hydrocephalus and Age Group

Table 3.3 shows number of hospitalized days for infants and children ages one to four years by the presence of hydrocephalus. During the first year of life, infants with hydrocephalus were hospitalized for a total average of 30.0 days (SD 30.1; median: 20.0; range 1-216) compared with 19.0 days (SD 38.6; median: 8.0; range: 1-476) for infants without hydrocephalus (p<0.0001). Comparing birth and post-birth hospitalizations, infants without hydrocephalus were hospitalized for average of 13.2 days at birth (SD 25.7; median: 5.0; range 1-221) compared with 20.2 hospitalized days (SD 20.6; median: 15.0; range 1-149) for infants with hydrocephalus (p<0.0001). For post-birth hospitalizations, infants without hydrocephalus were hospitalized for a total average of 11.5 days (SD 26.1; median: 4.0; range 1-255), while infants with hydrocephalus were hospitalized for a total average of 15.9 days (SD 23.6; median: 7.0; range 1-206) (p=0.0044). There were no significant differences been the average total number of hospitalized days for children with and without hydrocephalus during ages one to four years.

Table 3.4 shows the results for the total number of hospitalized days by the presence of coded hydrocephalus stratified by isolated vs. non-isolated SB. Infants and children with simpler presentations of SB had significantly shorter lengths of stay than

those with more complex presentations of SB. Infants with isolated SB and no hydrocephalus spent the fewest total days in the hospital, 11.5 days (SD: 18.6; median: 7.0; range: 1-138). Infants with non-isolated SB with or without hydrocephalus had over three times the number of total hospitalized days (mean: 37.0; SD: 30.0; median: 26.0; range: 1-125 and mean: 41.5; SD: 65.1; median: 23.0; range: 1-476, respectively) compared with infants with isolated SB without hydrocephalus ($p<0.0001$).

3.6.4 Bivariate Results Stratified by Health Payer

Table 3.5 shows the outcomes of interest stratified by health payer for *infancy* hospitalizations. At birth, infants insured by a public payer spent an average of 4.2 days longer in the hospital than infants insured by private insurance (public payer: 19.1 days, private payer: 14.9 days; $p=0.0058$). Infants insured by a public payer had 5.3% higher average costs than infants insured by a private payer (public payer: $31,282; private payer: $29,712; $p=0.0151$). Examining post-birth hospitalizations, infants with multiple payers were hospitalized significantly more often that infants insured by a private payer (multiple payers: 2.6 hospitalizations; private payer: 2.0 hospitalizations; $p=0.0226$).

Across all infancy hospitalizations, payer type was significantly associated with the number of hospitalizations, costs, and length of stay. Infants with multiple payers were hospitalized on average 52% more often that infants covered by a private payer (multiple payers: 3.2 hospitalizations; private payer: 2.1 hospitalizations; $p<.0001$). Infants with multiple payers were hospitalized an average of 39%% fewer total days than infants covered by a private payer (multiple payers: 11.8 days; private payer: 19.4 days; $p<.0001$). Finally, infants with multiple payers 36% lower average total hospital costs

compared with infants covered by private payers (multiple payers: $25,179; private payer: $39,049; p<0.0007).

The number of infants who were self-pay or under-insured was small (n=24 birth hospitalizations). Among these twenty-four infants, sixteen who had a subsequent infancy hospitalization were re-classified into the "multiple payers" category after their initial hospitalization, indicating they obtained some type of health insurance after their initial hospitalization.

3.6.5 Multivariable Results

In response to research question two, the following sections describe the associations between hospital use and cost and selected predisposing, enabling, and need characteristics among children with SB.

3.6.5.1 Multivariable Results for Number of Hospitalizations and Hydrocephalus, Stratified by Isolated versus Non-Isolated Spina Bifida

Table 3.6 shows the aPR and 95% CIs for the association between total number of hospitalizations and hydrocephalus, stratified by isolated vs. non-isolated SB. Both hydrocephalus and isolated vs. non-isolated SB are need characteristics. In the adjusted models, infants with isolated SB and hydrocephalus were hospitalized 53% more often during infancy and 82% more frequently at ages one to four years than those with isolated SB without hydrocephalus. Children with non-isolated SB with hydrocephalus were hospitalized 72% more often during infancy and over twice as often during ages one to four years compared with children with isolated SB without hydrocephalus. Children with non-isolated SB with hydrocephalus were hospitalized 79% more often during

infancy and over twice as often during ages one through four years compared with children with isolated SB without hydrocephalus.

3.6.5.2 Multivariable Results for Hospital Costs and Hydrocephalus, Stratified by Isolated versus Non-Isolated Spina Bifida

Table 3.7 shows the effect estimates and 95% CIs for the association between total estimated inpatient hospital costs and coded hydrocephalus, stratified by isolated vs. non-isolated SB. In the adjusted models, total costs for infants with isolated SB with hydrocephalus were over twice as high as costs for infants with isolated SB without hydrocephalus. Infants with non-isolated SB without hydrocephalus had over two times the total costs of those of children with isolated SB without hydrocephalus. Infants with non-isolated SB with hydrocephalus had over three times the total costs than those of children with isolated SB without hydrocephalus. There was no association however, between hydrocephalus, isolated or non-isolated SB, and average total costs for children ages one to four years.

3.6.5.3 Multivariable Results for Length of Stay and Hydrocephalus, Stratified by Isolated versus Non-Isolated Spina Bifida

Table 3.8 shows the effect estimates and 95% CIs for total length or stay and coded hydrocephalus, stratified by isolated vs. non-isolated SB. In adjusted models, infants isolated SB with hydrocephalus were hospitalized over twice as many days compared with infants with isolated SB without hydrocephalus. Infants with non-isolated SB, with or without hydrocephalus, spent three times more days in the hospital than infants with isolated SB without hydrocephalus. During ages one to four years, children

with non-isolated SB with hydrocephalus had over twice the number of hospitalized days compared with children with isolated SB without hydrocephalus.

3.6.5.4 Multivariable Results for Number of Hospitalizations and Selected Predisposing, Enabling and Need Characteristics

Table 3.6 shows effect estimates and 95% CI for the selected predisposing, enabling, and need characteristics and their effect on number of hospitalizations. In adjusted models, need characteristics had the most notable associations with numbers of hospitalizations. In addition to the effects of hydrocephalus and isolated vs. non-isolated SB described in previous sections, infants who were born prematurely were hospitalized 16% more frequently following their birth hospitalization than full-term infants.

Examining predisposing characteristics, infants born to Hispanic mothers had 22% fewer post-birth hospitalizations than infants born to non-Hispanic White mothers. Across all infancy, infants born to non-Hispanic Black mothers had 14% fewer hospitalizations than those born to non-Hispanic White mothers. Children ages one to four years born to non-Hispanic Black mothers had 25% fewer hospitalizations than children born to non-Hispanic White mothers. Boys, ages one to four years, had 24% fewer hospitalizations than girls.

Among enabling characteristics, children ages one to four years with a rural residence at birth had 46% fewer hospitalizations than those living in urban areas. Infants who changed health care payers had 49% more hospitalizations than those who were insured only by a private payer.

3.6.5.5 Multivariable Results for Hospital Costs and Selected Predisposing, Enabling and Need Characteristics

Table 3.7 also shows the effect estimates and 95% CIs for selected predisposing, enabling, and need characteristics and their influence on hospital costs. In addition to the effects of hydrocephalus and isolated vs. non-isolated SB described in previous sections, infants born preterm had 34% higher costs for their birth hospitalizations than those born at full-term. Infants born at a hospital with a lower level of nursery care had 60% lower costs compared with those born at a hospital with a higher level of nursery care. Infants who died during infancy had 79% higher hospital costs for post-birth hospitalizations compared with infants who survived. Children who died after infancy had over three times the costs during ages one to four years than infants who survived.

Among predisposing characteristics, children born to non-Hispanic Black mothers had 31% higher average total costs during ages one to four years than those born to non-Hispanic White mothers. Examining enabling characteristics during ages one to four years, total costs were 54% lower for rural resident children than those living in urban areas.

3.6.5.6 Multivariable Results for Length of Stay and Selected Predisposing, Enabling and Need Characteristics

Finally, Table 3.8 shows the effect estimates and 95% CI for selected predisposing, enabling, and need characteristics and their influence on length of stay. Need characteristics again had the most notable associations with this outcome. In addition to the effects of hydrocephalus and isolated vs. non-isolated SB described in previous sections, infants born preterm had 32% more hospitalized days at birth than

infants born full-term. Infants born at a hospital with a lower level of nursery care were hospitalized 46% fewer days at birth than those born at a hospital with Level III nursery care. Children who died were hospitalized more than twice as many total days for post-birth hospitalizations and more than five times the number of total days during ages one to four years than those who survived.

Among predisposing characteristics, infants born to Hispanic mothers spent 35% fewer days in the hospital for post-birth hospitalizations compared with infants born to non-Hispanic white mothers. During infancy, boys had 43% fewer hospitalized days than little girls.

Among enabling characteristics, children with a rural residence had 72% fewer hospitalized days during ages one to four years than those with an urban residence. Infants with a public payer source had 31% more hospitalized days than infants with a private payer source.

3.7 Discussion

My dissertation research provided statewide, population-based information about the number of hospitalizations, lengths of stay, and total estimated hospital costs for hospitalizations initiated during infancy and during ages one to four years for Florida-resident children born with SB. I extended research in this area by quantifying the difference in health resource use by comorbid or other individual characteristics. This information can inform program planning and policy development.

Patterns of hospital use and costs differed substantially for infants and children who had hydrocephalus and those that did not, as well as by isolated vs. non-isolated SB. In addition, hospital use and costs differed by several other predisposing, enabling, and

need characteristics: maternal race/ethnicity, rural residence, health care payer, prematurity, and level of care of the birth hospital nursery.

In response to my first research question, infants with hydrocephalus were hospitalized significantly more often, spent more days in the hospital, and incurred higher total hospital costs than those without hydrocephalus. Examining hospital use by both hydrocephalus and isolated vs. non-isolated SB, infants and children with the simplest presentation of SB (isolated without hydrocephalus) used the least hospital resources and incurred the lowest costs. Infants and children with non-isolated SB used more hospital resources than children with isolated SB, regardless of the presence of hydrocephalus. Children ages one to four years with hydrocephalus had significantly more hospitalizations and higher costs compared with those without hydrocephalus. These differences between infants and children with and without hydrocephalus and by isolated vs. non-isolated SB were expected because hydrocephalus and other birth defects add complexity and risk factors to SB that may result in the need for additional health care. Quantifying the extra resource use and cost for infants and children in these groups can assist state governments and program planners as they evaluate needs for services and forecast health care budget needs.

Results for number of hospitalizations and costs are consistent with previous research that found that infants with hydrocephalus may require readmissions for shunt revision or infections, thus adding to the costs of SB [12, 18, 20, 66, 85, 87, 92, 101, 111, 163, 164]. I am aware of no published research that has compared length of stay for children with and without hydrocephalus; however, results for length of stay are consistent with work that suggests comorbidities may result in more health resource use

[17, 192, 193]. In addition, I am aware of no published research that has compared results for hospital resource use by isolated vs. non-isolated SB.

Also in response to research question one, infants used more hospital resources and incurred greater costs during infancy compared with hospital use and costs during ages one to four years. This expected pattern is consistent with previous research that found that children with SB use the most health care resources during infancy and particularly during their birth hospitalizations [14]. This information can help to identify needs for age-specific services in this special population.

The mean length of stay for birth hospitalizations in this study (17.2 days) was slightly higher than the 15.1 days previously reported using AHRQ HCUP 2003 KID data [2]. The difference could reflect differences in the ascertainment methods of infants with birth defects [113]. Differences in number of hospitalized days from previous research findings may also be the result of different methods for addressing the issue of inter-hospital transfers and the effect of transfers on calculation of the number of hospitalized days [2, 177]. The findings for length of stay during infancy were almost double that of a previous study (10.5 day versus 5.9 days) [14]. The previous study was based on a small sample size (n=13 infants) and may not be comparable.

No direct comparisons were available for length of stay for ages one to four years. One previous study found an average length of stay of 5.5 days per hospitalization for children with SB ages 1-17 years [14]. The findings of this study (4.9 days) were slightly shorter.

Finally, in response to research question one, children who were self-pay or under-insured had fewer and shorter hospitalizations during infancy compared with

children with some health care coverage. Among CSHCN, the most critical determinant of access to care is adequate health insurance coverage [127]. Families who must pay for health care out-of-pocket or who have inadequate insurance may need to limit the health care they seek for their infant with SB. It would be useful for health care providers to make families aware of insurance options, including support provided by the Title V Maternal and Child Health Block Grant funds or through the Affordable Care Act [194]. It important to acknowledge that the "multiple payers" category was primarily composed of infants who were self-pay or under-insured at birth and then obtained insurance coverage for a subsequent infancy admission (results not shown). Numbers in the self-pay or under-insured category were small, so findings for infants and children in these categories should be viewed with caution.

In response to my second research question, I observed several predisposing, enabling, and need characteristics that influenced hospital use and costs. Four need characteristics were consistently associated with higher hospital use and costs for children with SB: the presence of hydrocephalus, non-isolated SB, prematurity, and a higher level of hospital nursery care at birth. I also observed that certain predisposing characteristics (maternal race/ethnicity, sex of child) and enabling characteristics (rural residency and payer type) influenced hospital use and costs.

Among predisposing characteristics, infants born to mothers of minority racial or ethnic groups had significantly fewer hospital admissions during infancy and during ages one to four years than children born to non-Hispanic White mothers. Mothers of minority racial or ethnic groups may experience barriers to accessing care for their children. These include: personal barriers (health care beliefs, perceptions of need, or

language and other social and cultural influences); financial barriers (insufficient monetary resources or lack of health care insurance); and or structural barriers (transportation and geographic location of services). While most predisposing factors are not mutable, a better understanding of the effects of certain predisposing factors, such as maternal ethnicity, on use of health care resources and costs may be useful. For example, educational programs that promote appropriate use of hospital resources can reach out to mothers with unique needs or risks. Similarly, programs and policies that focus on selected groups who would benefit from additional support to access care, including information on financial and other family support, would be helpful.

The most consistent statistically significant associations across all outcomes were among the need characteristics. Examining need characteristics, in addition to those already reported for hydrocephalus and isolated vs. non-isolated SB, infants born preterm were hospitalized more often and incurred higher total costs during the first year of life compared with those born full-term. Both prematurity and the presence of another major birth defect in addition to SB may present further medical challenges that result in increased hospital use and costs. These findings are consistent with previous general research that found comorbidities can add to an individual's use of health services [17, 158, 192, 193]. In addition, infants with SB born at a hospital with a lower level of nursery care had fewer hospitalizations and incurred lower costs than infants born in a hospital with a high level of nursery care. If an infant with SB is diagnosed prenatally, mothers may pre-select a delivery hospital that has a higher level of hospital nursery care, especially if the SB condition is severe or the infant has other major birth defects. Infants treated in a hospital with a lower level of nursery care may have fewer medical needs

than those treated at a birth hospital with high level of nursery care. Findings also may be associated with different fees charged by hospitals with different levels of nursery care.

Examining enabling characteristics, rural maternal residence was associated with fewer hospitalizations and lower costs for children during ages one to four years. This expected finding was consistent with previous research that found infants in rural counties experienced fewer hospitalized days compared with those living in urban areas [195]. Increased travel time and distance, high transportation costs, and limited transportation resources may all be barriers to accessing health care [120, 121]. Families living in rural areas may experience more of these accessibility barriers than families living in urban areas [128, 136]. However, rural residence was not associated with number of hospitalizations during infancy. This finding was inconsistent with previous research that found infants in rural counties experienced fewer hospitalizations compared with infants living in urban areas [195]. The complex nature of SB and perceived *need* for care may motivate mothers to seek care for their infants with SB, despite barriers associated with rural residence.

The results described in the previous paragraphs suggest that need characteristics may have the most influence on hospital use and costs among children in this study sample. This finding is consistent with a previous study found that children with multiple medical conditions (an indication of need), were eight times as likely to be high users of physician services as those without multiple conditions [99]. Another study reported that among CSHCN, those with more than one chronic condition were 27% more likely to be hospitalized in the course of a year and those with poor or fair perceived health were over

twice as likely to be hospitalized than CSHCN with only one chronic condition or perceived good health [100].

In summary, hydrocephalus and the isolated vs. non-isolated SB were associated with substantially higher hospital resource use and costs during infancy. While still notable, the effects of hydrocephalus and isolated vs. non-isolated SB decreased during ages one to four years. Birth hospitalizations were the greatest contributor to all infancy costs. The patterns of hospital use during post-birth hospitalizations were more similar to those of hospitalizations during ages one to four years than to birth hospitalizations. Children with isolated SB with hydrocephalus were more like children with non-isolated SB with hydrocephalus in their use of hospital resources than they were like children with isolated SB without hydrocephalus. Among predisposing, enabling, and need characteristics, need characteristics appeared to have the most influence on hospital resource use and costs for this study sample. This dissertation extended research by quantifying the differences in hospital resource use and costs for infants and children with hydrocephalus and isolated vs. non-isolated SB.

3.8 Strengths and Limitations

3.8.1 Innovation and Strengths in the Research Topic

This study examined differences in hospital use and costs in a population-based, statewide study of unduplicated Florida children with SB insured by different health care payer types. The study followed children for the first four years of life, which provided new opportunities to examine hospital use and costs and associated predisposing, enabling, and need characteristics over time. The comparison of health care use, and cost findings in the presence and absence of hydrocephalus was also unique. With the

exception of one study, which explored hospital expenditures for North Carolina children with SB and SB with hydrocephalus born 1995-2002 and continuously enrolled in Medicaid [12], no similar work related to comorbidities and SB existed. Thus, each of these topics represents new or expanded areas of research.

3.8.2 Innovation and Strengths in the Methodology

The study population for this research was an important strength in that it represented a diverse group of children. The state of Florida was the fastest growing and fourth most populous state according to the 2000 U.S. Census [22, 23]. Florida was fourth in number of annual live births, second in number of live births to non-Hispanic Black women, and third in number of live births to Hispanic women, nationwide during the study period of 1998-2008 [22-24]. Florida also supports a statewide, population-based birth defects registry and a statewide agency for the collection of hospital discharge data that provided information for this project. This statewide, population-based study sample used linked, longitudinal data from the FBDR and the Florida AHCA, which provided a robust source of information for this project.

Additionally, this research incorporated several methods that are not frequently used in health services research for birth defects, and thus are both strengths and innovations. First, the unit of study was the individual child, rather than the more typical observation level of hospital admission or other aggregate data level. Second, the dataset provided access to hospital discharge data for children from multiple payer sources, rather than the more commonly researched single payer source. This feature allowed for a more complete picture of hospital use across multiple payers. This feature also provided the opportunity to observe changes in payer type through infancy. Third, the

linked, longitudinal data provided the opportunity to follow each child through early childhood to give unique insights into the differences in hospital use and costs over time. Fourth, I converted the total hospital charges to estimated costs using the AHRQ Healthcare Cost and Utilization Project cost-to-charge ratio files, which are based on accounting reports from Centers for Medicare and Medicaid Services [178]. Multiplication of the hospital charge by the cost-to-charge ratio results in an estimated hospital cost for those charges [178] and is a useful tool for making comparisons across cost and charge data. In addition, I adjusted the costs to the current dollar value amount.

A final strength of this methodology is its ability to be replicated for other birth defects. Health services researchers and public health researchers can use these methods to examine different types of birth defects in collaboration with other birth defect registries and state and federal agencies.

Regarding generalizability, the demographic findings of this study are similar to the characteristics of all Florida-born infants during with study period with a few exceptions. Infants and children in the sample were significantly more likely to have been born preterm compared to all live-born infants in Florida born during the study period (27% preterm births compared to 11% in Florida, 2007) [27]. This finding is consistent with previous findings related to birth defects and prematurity [172, 196, 197]. The study sample also included a slightly lower proportion of Hispanic mothers than found statewide in Florida (29% statewide compared to 25% in the study sample). In addition, about 50% of infants with SB had only public insurance for all hospitalizations during infancy; about 43% of all births in Florida during the study period were insured by Medicaid [27]. These differences in the study sample versus the population

characteristics of Florida were expected or minor. Thus, the results may be generalizable to the state.

3.8.3 Limitations Inherent in the Data Sources

This research faced several limitations based on the data used. Infants identified for this study were based on the passive surveillance methodologies for identifying infants with birth defects using ICD-9-CM codes. Some birth defects surveillance systems actively identify birth defect diagnoses using modifications of the British Pediatric Association (BPA) *Classification of Diseases* [198, 199]. In contrast, passive birth defects surveillance systems, while widely used, do not actively verify the birth defect diagnosis by review of medical records, hospital charts, or nursery logs. Passive surveillance techniques may lead to under-reporting or miss reporting of infants with birth defects or a specific defect type [27-29, 200, 201]. However, the FBDR's overall completeness of ascertainment of birth defects has been estimated at 87%, with case ascertainment variation noted by specific defect [28, 29]. Because SB is relatively easy to detect, a passive surveillance system may be less of a limitation than with other birth defects that are more difficult to detect. In the FBDR data, case ascertainment of infants with SB without anencephaly was 88.0% [29], a relatively high completeness of ascertainment of SB. The presence of hydrocephalus and the presence of other major birth defects used in identifying children with isolated or non-isolated SB were also based on coded data rather than clinically verified data. Finally, limited information on prenatal diagnosis is available in passive surveillance systems [27]. For Florida, there is no access to data on prenatal diagnosis for birth defects through the FBDR. Further, because this

analysis used data from the FBDR, it is a state-specific study, which may limit generalizability to other states or regions of the country.

The nature of the study sample also presented a limitation because the sample size was not constant over the ten-year study period. While the Florida AHCA provided ten years of hospital discharge data, the full ten years only applied to the children born in the first year of the study. Each subsequent birth cohort had one less year of data, ending with the birth cohort of 2007, which had only one year of data. The smaller sample size for each cohort decreased statistical power, thus reducing the opportunity to observe effects that may exist in the study sample. To reduce this risk, I limited the analyses to the first four years of life.

Additionally, the principal payer source variable that was used in analyses of birth payer and payer types across the four years was an expected principal payer source. It is not known if this was the actual payer source. Furthermore, some infants may have dual payer sources (e.g., private and public payer) for a single hospitalization. Such information is not generally reported with hospital discharge data.

Another limitation was the fact that the data were based on Florida hospital administrative data. Administrative data may be at risk for error or inconsistent coding that could incorrectly code maternal residential addresses and hospital facility codes or introduce error in diagnostic coding. This data did not include information on families that sought care out-of-state for their child. Additionally, while approximately 290 Florida hospitals report data to the Florida AHCA, not all are required to report, including one Shriner's Hospital that provides care at no cost to patients, as well as long and short-term psychiatric hospitals, inpatient residential treatment and rehabilitation facilities, and

military hospitals [202]. Thus, I was not able to capture access to care data on all the children within the Florida SB population under study. However, because data from 108 different Florida hospitals were represented in the data set [140] and most of the non-reporting hospitals do not provide newborn care, the amount of data lost was likely limited. Thus, the findings of this research may be generalizable at least to the state of Florida.

Administrative datasets may also be limited by missing observations. Among the all covariables observed in this study, no covariable was missing more than 10% of its observations. The covariable with the largest number of missing values was "adequacy of prenatal care", that had 5.6% missing observations. While deletion of missing data may reduce sample size, lower statistical power, and potentially introduce bias if the data is not missing at random [203], the numbers missing and deleted from analysis in this dataset were small and likely had no effect on the outcomes.

Lastly, the use of administrative data does not capture all aspects of an individual's inclination to use health services resources. Characteristics such as travel time and distance, family resources, employment status, and health beliefs and health literacy are not available in this administrative dataset; however, these are characteristics that may influence the use of hospital services.

3.8.4 Limitations Inherent in Research Design

This research faced additional limitations resulting from the study design. I could not directly compare hospital cost estimates from my dissertation with previous estimates for children with SB for several reasons. First, costs are not equivalent to the charges or expenditures reported in previous research. Second, some previous studies used a single

payer source, such as private health insurance or Medicaid, which can have different reimbursement rates for services. Third, some previous studies did not adjust costs for inflation. Fourth, some previous studies did not adjust costs for inflation or used different case ascertainment methods. While acknowledging differences in charges, costs and expenditures, I addressed this limitation by converting charges to estimated hospital costs based on Florida's average hospital cost-to-charge ratio using the most recent cost-to-charge ratios from AHRQ Healthcare Cost and Utilization Project. In addition, I adjusted costs to the most current dollar amount using hospital industry data from the Producer Price Index, U.S. Department of Labor to provide timely findings.

Further, I did not examine the reason for the hospitalization. The hospitalization may have been directly related to a child's SB diagnosis or could have been unrelated (such as an illness or accident). This is limitation of study could result in overstated or underestimated SB costs.

Finally, I noted that total estimated hospital costs represent only one component of health care costs. Thus, this research did not capture the full health care and societal cost burden associated with the care of SB during the first four years of childhood. To better estimate the total cost of care for infants with SB, information on other cost components, such as outpatient costs and prescription drug costs, would be needed. These costs were included in the dataset; however, the data were not as complete as the inpatient charges data and thus were not used. Professional fees, including physician charges, were also absent from the analysis because they are not commonly included in hospital discharge data and were not available in the Florida AHCA data set. Inclusion of indirect costs, such as the value of care provided by the family within the home or the

value of lost parental work time, would also have contributed to a more complete understanding of the financial burden of this condition. Because these types of data were not available, the findings of this study underestimate the total health care costs for children with SB.

3.9 Implications for Public Health Practice and Research

This study suggests several points for consideration in the areas of public health and access to health care services. First, this research suggests that the use of birth defects surveillance data combined with hospital discharge data can provide vital information about patterns and predictors of hospital use and costs for children with SB. An understanding of patterns and predictors of hospital use and costs may be important to inform health care planning by governmental providers such as federal, state, and local agencies, and particularly those serving CSHCN. For example, cost information for children covered by public payers can help state-level planners develop economic forecasts for state health care programs serving CSHCN. In addition, information about health resource use may help policy makers to assess new or changing needs for services among CSHCN.

Second, collaborative multi-state, population-based studies linking multiple birth defect registries and linked, longitudinal data would be useful to further examine hospital use and costs for SB and other birth defects. Collaborative research projects would increase study sizes, thus increasing study power and potentially the ability to observe effects over time. Continued support and expansion of the *National Spina Bifida Patient Registry* [204], as well as continued funding for the *National Birth Defects Prevention Network, National Birth Defects Prevention Study*, and state birth defects surveillance

systems to advance the knowledge of healthcare utilization among individuals with SB will be important for these efforts.

Third, the observed differences in health care use based on *need* were expected and do not necessarily indicate the disparities or inequities in care. Primary prevention of SB continues to be the best way to reduce need. Continued support of the mandatory folic acid fortification of the U.S. cereal grain supply is important to this goal [51, 52, 116]. The enactment of new policies that support the fortification of corn masa flour with folic acid may help prevent further cases of SB, especially among the Hispanic population [205].

The observed differences in hospital use and costs based on maternal race/ethnicity, maternal nativity, and education, rural residency and payer type may be associated with different policies and reimbursement rates of the payers or may indicate disparities in access to and quality of care. Each of these presents opportunity for further exploration.

Additional opportunities for future research include examination of hospital use and costs by other comorbidities associated with SB and across the lifespan. Among CSHCN, adequate health insurance coverage is one of the most critical determinants of access to care. Thus, further research to explore the different types of payer changes that occur during infancy and childhood would be important (e.g., identifying percentages and reasons for change from public to private payer or from private to public payer). A better understanding of the predisposing, enabling, and need characteristics associated with changes in payer type and the effects of those changes on health care resource utilization and health outcomes would also be important to explore.

In conclusion, my dissertation research provided estimates of health care resource utilization from birth to age four years for children born with SB in Florida between 1998 and 2007. My research also quantified differences in hospital use and cost by the presence of hydrocephalus and isolated vs. non-isolated SB and by birth and post-birth hospitalizations during infancy, for all infancy and for ages one to four years. The use of quantified, summary measures of hospital use and costs may offer new opportunities to identify the impact of the commonly occurring comorbidities such as SB and hydrocephalus, on health care utilization and costs. This information can assist in health service planning and financing for children with SB and other birth defects and associated comorbidities.

Finally, the findings of this study can help to inform research for other birth defects. This dissertation research demonstrated that hospital discharge data and data collected by birth defects surveillance programs can be used to analyze differences in costs and payer status by selected diagnoses and sociodemographic information. Health service researchers and other state birth defects surveillance programs may collaborate to conduct similar analyses and determine any patterns and differences in results. A more complete understanding of the patterns of hospital use and costs associated with SB and other birth defects can inform program planning and policy development, which may ultimately contribute to reduced health costs, improved health care delivery and quality of care, and improved long-term quality of life for families and children born with SB or other similar birth defects.

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Table 3.1 Selected characteristics of Florida-born children with spina bifida, with and without hydrocephalus, 1998-2007

Characteristics	All infants (n=614)		*Without* hydrocephalus (n=265)		*With* hydrocephalus (n=349)		p-value
	n	(%)	n	(%)	n	(%)	
Exposures of interest							
Hydrocephalus							
Yes	349	(56.8)					
No	265	(43.2)					
Spina Bifida [1]							
Isolated	458	(74.6)	198	(74.7)	260	(74.5)	0.9509
Non-isolated	156	(25.4)	67	(25.3)	89	(25.5)	
Spina Bifida and Hydrocephalus							
Isolated SB without hydrocephalus	198	(32.3)					
Non-isolated SB without hydrocephalus	67	(10.9)					
Isolated SB with hydrocephalus	260	(42.4)					
Non-isolated SB with hydrocephalus	89	(14.5)					
Predisposing characteristics							
Maternal age (in years)							
≤24	224	(36.5)	78	(29.4)	146	(41.8)	**0.0062**
25-29	164	(26.7)	79	(29.8)	85	(24.4)	
≥30	225	(36.6)	108	(40.8)	117	(33.5)	
Missing	1	(0.2)					
Maternal race/ethnicity							
Non-Hispanic White	323	(52.6)	136	(51.3)	187	(53.6)	0.2988
Hispanic	153	(24.9)	73	(27.5)	80	(22.9)	
Non-Hispanic Black	128	(20.9)	54	(20.4)	74	(21.2)	
Other	10	(1.6)	2	(0.8)	8	(2.3)	
Maternal nativity							
Born in U.S.	466	(75.9)	198	(75.0)	268	(77.0)	0.5631
Foreign-born	146	(23.8)	66	(25.0)	80	(23.0)	
Missing	2	(0.3)					
Maternal marital status							
Married	368	(59.9)	162	(61.1)	206	(59.0)	0.5978
Not married	246	(40.1)	103	(38.9)	143	(41.0)	
Maternal parity							
First child	237	(38.7)	99	(37.4)	138	(39.5)	0.6073
Second or subsequent child	376	(61.3)	165	(62.5)	211	(60.5)	

Table 3.1 (continued)

Characteristics	All infants		*Without* hydrocephalus		*With* hydrocephalus		p-value
	n	(%)	n	(%)	n	(%)	
Maternal education							
High school diploma or more	468	(76.2)	214	(81.7)	254	(73.8)	**0.0226**
No high school diploma	138	(22.5)	48	(18.3)	90	(26.2)	
Missing	8	(0.01)					
Sex of infant							
Female	317	(51.6)	142	(53.6)	175	(50.1)	0.3980
Male	297	(48.4)	123	(46.4)	174	(49.9)	
Enabling characteristics							
Prenatal care [2]							
Adequate prenatal care	447	(72.8)	194	(77.9)	253	(76.4)	0.6754
Inadequate prenatal care	133	(21.7)	55	(22.1)	78	(23.6)	
Missing	34	(5.5)					
Residential rurality [3]							
Urban /urban cluster	525	(85.5)	226	(85.3)	299	(86.2)	0.7563
Rural	87	(14.2)	39	(14.7)	48	(13.8)	
Missing	2	(0.3)					
Payer for birth hospitalization (n=569) [4]							
Public payer	292	(47.6)	119	(44.9)	173	(49.6)	0.1912
Private payer	253	(4.2)	116	(43.8)	137	(39.3)	
Self or uninsured	24	(3.9)	14	(5.3)	10	(2.9)	
No birth hospitalization	45	(7.3)	16	(6.0)	29	(8.3)	
Payer across all infancy [4]							
Public payer only	306	(49.8)	123	(46.4)	183	(52.4)	0.1013
Private payer only	236	(38.4)	114	(43.0)	122	(35.0)	
Self or uninsured only	8	(1.3)	5	(1.9)	3	(0.9)	
Multiple payers	64	(10.4)	23	(8.7)	41	(11.8)	
Need characteristics							
Preterm birth (< 37 weeks gestation)							
No	448	(73.0)	205	(77.9)	243	(69.8)	**0.0247**
Yes	163	(26.5)	58	(22.1)	105	(30.2)	
Missing	3	(0.5)					
Low birth weight (< 2500 grams)							
No	492	(80.1)	206	(77.7)	286	(82.2)	0.1705
Yes	121	(19.7)	59	(22.3)	62	(17.8)	
Missing	1	(0.2)					

Table 3.1 (continued)

Characteristics	All infants		*Without* hydrocephalus		*With* hydrocephalus		p-value
	n	(%)	n	(%)	n	(%)	
Plurality							
Singleton birth	593	(96.6)	252	(95.1)	341	(97.7)	0.0776
Multiple birth	21	(3.4)	13	(4.9)	8	(2.3)	
Nursery level of birth hospital [5]							
Level III	511	(83.2)	201	(76.1)	310	(89.3)	**<.0001**
Level I or II	100	(16.3)	63	(23.9)	37	(10.7)	
Missing	3	(0.5)					
Inter-hospital transfer [6]							
No inter-hospital transfer	468	(76.2)	205	(77.4)	263	(75.4)	0.5642
Inter-hospital transfer	146	(23.8)	60	(22.6)	86	(24.6)	
Death [7]							
No death	561	(91.4)	239	(90.2)	322	(92.3)	0.2972
Died during neonatal period	19	(3.1)	12	(4.5)	7	(2.0)	
Died during infancy	22	(3.6)	10	(3.8)	12	(3.4)	
Died after infancy	12	(2.0)	4	(1.5)	8	(2.3)	

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point. Columns may not add to 100% because of missing or unknown values. P-values in bold are considered statistically significant at <0.05.

[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.

[2] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered "adequate prenatal care".

[3] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.

[4] Payers are expected, but not confirmed, health care payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no insurance, no third party coverage, or less than 30%. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.

[5] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.

[6] Inter-hospital transfers occurred during birth hospitalization. Transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a one-day difference existed between a discharge from one hospital and an admission to another hospital and records indicated an accompanying transfer.

[7] All deaths occurred during study period, prior to December 31, 2008. Neonatal deaths are deaths at < 28 days. Infancy deaths < 365 days.

Table 3.2 Selected characteristics of Florida-born children with spina bifida, with and without hydrocephalus and stratified by isolated or non-isolated spina bifida, 1998-2007

Characteristics	All infants (n=614)		Hydrocephalus				No hydrocephalus				p-value
			Isolated SB (n=260)		Non-isolated SB (n=89)		Isolated SB (n=198)		Non-isolated SB (n=67)		
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	
Predisposing characteristics											
Maternal age (in years)											
≤24	224	(36.5)	104	(40.2)	42	(47.2)	54	(27.3)	24	(35.8)	**0.0207**
25-29	164	(26.7)	68	(26.3)	17	(19.1)	58	(29.3)	21	(31.3)	
≥30	225	(36.6)	87	(33.6)	30	(33.7)	86	(43.4)	22	(32.8)	
Missing	1	(0.2)									
Maternal race/ethnicity											
Non-Hispanic White	323	(52.6)	131	(50.4)	56	(62.9)	109	(55.0)	27	(40.0)	0.0733
Hispanic	153	(24.9)	61	(23.5)	13	(14.6)	53	(26.8)	20	(29.9)	
Non-Hispanic Black	128	(20.9)	61	(23.5)	19	(21.5)	34	(17.0)	20	(29.9)	
Other	10	(1.6)	7	(2.7)	1	(1.1)	2	(1.0)	0	(0)	
Maternal nativity											
Born in U.S.	466	(75.9)	197	(76.1)	71	(79.8)	150	(75.8)	48	(72.7)	0.7801
Foreign-born	146	(23.8)	62	(23.9)	18	(20.2)	48	(24.2)	18	(27.3)	
Missing	2	(0.3)									
Maternal marital status											
Married	368	(59.9)	157	(60.4)		()	132	(66.7)	30	(44.8)	**0.0114**
Not married	246	(40.1)	103	(39.6)		()	66	(33.3)	37	(55.2)	
Maternal parity											
First child	237	(38.7)	94	(36.2)	44	(49.4)	81	(40.9)	18	(27.3)	**0.0283**
Second or subsequent child	376	(61.3)	166	(63.8)	45	(50.6)	117	(59.1)	48	(72.7)	

Table 3.2 (continued)

Characteristics	All infants		Hydrocephalus				No hydrocephalus				p-value
			Isolated SB		Non-isolated SB		Isolated SB		Non-isolated SB		
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	
Maternal education											
High school diploma or more	468	(76.2)	187	(72.8)	67	(77.0)	165	(83.8)	49	(75.4)	0.0500
No high school diploma	138	(22.5)	70	(27.2)	20	(23.0)	32	(16.2)	16	(24.6)	
Missing	8	(0.01)									
Sex of infant											
Female	317	(51.6)	127	(48.9)	48	(53.9)	103	(52.0)	39	(58.2)	0.5381
Male	297	(48.4)	133	(51.1)	41	(46.1)	95	(48.0)	28	(41.8)	
Enabling characteristics											
Prenatal care [2]											
Adequate prenatal care	447	(72.8)	190	(76.6)	63	(75.9)	150	(80.2)	44	(71.0)	0.4852
Inadequate prenatal care	133	(21.7)	58	(23.4)	20	(24.1)	37	(19.8)	18	(29.0)	
Missing	34	(5.5)									
Residential rurality [3]											
Urban /urban cluster	525	(85.5)	225	(87.2)	74	(83.1)	170	(85.9)	56	(83.6)	0.7518
Rural	87	(14.2)	33	(12.8)	15	(16.9)	28	(14.1)	11	(16.4)	
Missing	2	(0.3)									
Payer for birth hospitalization [4]											
Public payer	292	(47.6)	128	(49.2)	45	(50.5)	84	(42.4)	35	(52.2)	0.1675
Private payer	253	(4.2)	104	(40.0)	33	(37.1)	90	(45.5)	26	(38.8)	
Self or uninsured	24	(3.9)	10	(3.8)	0	(0)	10	(5.0)	4	(6.0)	
No birth hospitalization	45	(7.3)	18	(6.9)	11	(12.4)	14	(7.1)	2	(3.0)	

Table 3.2 (continued)

Characteristics	All infants		Hydrocephalus				No hydrocephalus				p-value
			Isolated SB		Non-isolated SB		Isolated SB		Non-isolated SB		
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	
Payer across all infancy [4]											
Public payer only	306	(49.8)	130	(50.0)	53	(59.6)	89	(44.9)	34	(50.7)	**0.0486**
Private payer only	236	(38.4)	93	(35.8)	29	(32.6)	91	(46.0)	23	(34.3)	
Self or uninsured only	8	(1.3)	3	(1.2)	0	(0)	2	(1.0)	3	(4.5)	
Multiple payers	64	(10.4)	34	(13.1)	7	(7.9)	16	(8.1)	7	(10.5)	
Need characteristics											
Preterm birth (< 37 weeks gestation)											
No	448	(73.0)	172	(66.4)	71	(79.8)	152	(77.2)	53	(80.3)	**0.0100**
Yes	163	(26.5)	87	(33.6)	18	(20.2)	45	(22.8)	13	(19.7)	
Missing	3	(0.5)									
Low birth weight (≤ 2500 grams)											
No	492	(80.1)	218	(84.2)	68	(76.4)	160	(80.8)	46	(68.7)	**0.0284**
Yes	121	(19.7)	41	(15.8)	21	(23.6)	38	(19.2)	21	(31.3)	
Missing	1	(0.2)									
Plurality											
Singleton birth	593	(96.6)	253	(97.3)	88	(98.9)	187	(94.4)	65	(97.0)	0.2027
Multiple birth	21	(3.4)	7	(2.7)	1	(1.1)	11	(5.6)	2	(3.0)	
Nursery level of birth hospital [5]											
Level III	511	(83.2)	235	(90.7)	75	(85.2)	146	(74.1)	55	(82.1)	**<.0001**
Level I or II	100	(16.3)	24	(9.3)	13	(14.8)	51	(25.9)	12	(17.9)	
Missing	3	(0.5)									

Table 3.2 (continued)

Characteristics	All infants		Hydrocephalus				No hydrocephalus				p-value
			Isolated SB		Non-isolated SB		Isolated SB		Non-isolated SB		
	n	(%)	n	(%)	n	(%)	n	(%)	n	(%)	
Inter-hospital transfer [6]											
No inter-hospital transfer	468	(76.2)	208	(80.0)	55	(61.8)	156	(78.8)	49	(73.1)	**0.0040**
Inter-hospital transfer	146	(23.8)	52	(20.0)	34	(38.2)	42	(21.2)	18	(26.9)	
Death [7]											
No death	561	(91.4)	245	(94.2)	77	()86.5	188	(95.0)	51	(76.1)	**<.0001**
Died during neonatal period	19	(3.1)	2	(0.8)	5	(5.6)	5	(2.5)	7	(10.5)	
Died during infancy	22	(3.6)	8	(3.1)	4	(4.5)	3	(1.5)	7	(10.5)	
Died after infancy	12	(2.0)	5	(1.9)	3	(3.4)	2	(1.0)	2	(3.0)	

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point. Columns may not add to 100% because of missing or unknown values. P-values in bold are considered statistically significant at <0.05.

[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.

[2] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered "adequate prenatal care".

[3] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.

[4] Payers are expected, but not confirmed, health care payers. Public insurance included Medicare, Medicaid, KidCare, Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.

[5] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.

[6] Inter-hospital transfers occurred during birth hospitalization. Transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a one-day difference existed between a discharge from one hospital and an admission to another hospital.

[7] All deaths occurred during study period, prior to December 31, 2008. Neonatal deaths are deaths at < 28 days. Infancy deaths < 365 days.

Table 3.3 Number of hospitalizations [1], number of hospitalized days [2], and estimated total costs [3] for Florida-born children with spina bifida with and without hydrocephalus by age category, 1998-2007

Hospitalizations	All children	*Without* hydrocephalus	*With* hydrocephalus	p-value
Birth hospitalizations [4]				
Number of children with hospitalizations	569	249	320	
Total number of hospitalizations	569	249	320	
Mean (SD) total number hospitalized days	17.2 (23.2)	13.2 (25.7)	20.2 (20.6)	**<0.0001**
Median (IQR) total number hospitalized days	10.0 (17.0)	5.0 (8.0)	15.0 (16.0)	
Range for total number hospitalized days	1-221	1-221	1-149	
Mean (SD) total inpatient costs ($)	30,557 (52,148)	23,711 (64,967)	35,884 (38,675)	**<0.0001**
Median (IQR) total inpatient costs ($)	18,789 (29.411)	6,615 (19,105)	27,491 (25,185)	
Range for total inpatient costs ($)	124-706,793	124-706,793	202-309,432	
Post-birth hospitalizations during infancy				
Number of children with hospitalizations	406	153	253	
Total number of hospitalizations	884	264	620	
Mean (SD) number of hospitalizations	2.2 (1.7)	1.7 (1.4)	2.5 (1.9)	**<.0001**
Median (IQR) number of hospitalizations	2.0 (2.0)	1.0 (1.0)	2.0 (2.0)	
Range for number of hospitalizations	1-11	1-11	1-11	
Mean (SD) total number hospitalized days	14.2 (24.7)	11.5 (26.1)	15.9 (23.6)	**0.0044**
Median (IQR) total number hospitalized days	5.0 (11.0)	4.0 (6.0)	7.0 (16.0)	
Range for total number hospitalized days	1-255	1-255	1-206	
Mean (SD) total inpatient costs ($)	29,592 (64,931)	25,050 (77,901)	32,338 (55,636)	**0.0006**
Median (IQR) total inpatient costs ($)	11,286 (19,758)	8,775 (11,908)	13,787 (27,546)	
Range for total inpatient costs ($)	720-883,476	817-883,476	720-487,037	
All infancy hospitalizations (< 1 year)				
Number of children with hospitalizations	614	265	349	
Total number of hospitalizations	1453	513	940	
Mean (SD) number of hospitalizations	2.4 (1.7)	1.9 (1.4)	2.7 (1.9)	**<.0001**
Median (IQR) number of hospitalizations	2.0 (2.0)	2.0 (1.0)	2.0 (2.0)	
Range for number of hospitalizations	1-12	1-12	1-12	
Mean (SD) total number hospitalized days	25.3 (34.4)	19.0 (38.6)	30.0 (30.1)	**<.0001**
Median (IQR) total number hospitalized days	14.0 (24.0)	8.0 (12.0)	20.0 (26.0)	
Range for total number hospitalized days	1-476	1-476	1-216	
Mean (SD) total inpatient costs ($)	47,884 (86,934)	36,742 (110,248)	56,345 (62,655)	**<.0001**
Median (IQR) total inpatient costs ($)	26,825 (39,039)	14,838 (21,203)	38,253 (37,778)	
Range for total inpatient costs ($)	124-1,590,268	124-1,590,268	445-500,210	

Table 3.3 (continued)

Hospitalizations	All children	*Without* hydrocephalus	*With* hydrocephalus	p-value
Hospitalizations during years 1-4				
Number of children with hospitalizations	251	61	190	
Total number of hospitalizations	763	146	617	
Mean (SD) number of hospitalizations	3.0 (3.0)	2.4 (2.8)	3.2 (3.1)	**0.0023**
Median (IQR) number of hospitalizations	2.0 (3.0)	1.0 (1.0)	2.0 (3.0)	
Range for number of hospitalizations	1-19	1-15	1-19	
Mean (SD) total number hospitalized days	14.8 (26.8)	15.6 (34.8)	14.5 (23.8)	0.1601
Median (IQR) total number hospitalized days	6.0 (12.0)	5.0 (8.0)	6.0 (12.0)	
Range for total number hospitalized days	1-206	1-206	1-175	
Mean (SD) total inpatient costs ($)	30,483 (57,427)	29,177 (64,877)	30,902 (55,004)	**0.0181**
Median (IQR) total inpatient costs ($)	11,593 (22,733)	9,223 (21,535)	13,095 (25,194)	
Range for total inpatient costs ($)	304-505,528	812-361,726	304-505,528	

SD =standard deviation, IQR =interquartile range Note; Statistical significance at $p < 0.05$ (p-value based on Wilcoxon Rank Sum test

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point.

[1] Hospitalizations refer to hospitalizations initiated, but not necessarily completed in age category. Hospitalizations were assessed as continuous episodes of hospital care, regardless of whether a transfer took place. Multiple admission records were merged into one if an infant was admitted to a hospital on the same day as a discharge from a previous admission, or if the infant was admitted to a hospital on the day after a previous discharge with an accompanying "transfer" code.

[2] Hospitalized days refers to the number of days that an infant spent in the hospital for all admissions initiated within the specified age category.

[3] Estimated costs in 2012 U.S. dollars. Estimated costs calculated as total charges adjusted to Florida's statewide average hospital cost-to-charge ratio (Agency for Healthcare Research and Quality, Health Care Utilization Project, 2012). Inpatient charges include all hospital facility charges (excludes professional fees): pharmacy, medical and surgical supplies, laboratory, radiology and other imaging, cardiology, operating room, anesthesia, recovery room, emergency room (if an inpatient hospital admission originated in the emergency room), treatment or observation room (if a visit resulted in an inpatient hospital admission) charges (Agency for Health Care Administration, 2011).

[4] Birth hospitalizations defined as a first hospitalization with age at admission of 0 days or a first hospitalization with an age at admission of 1 day with an accompanying indication of hospital transfer.

Table 3.4 Number of hospitalizations [1], number of hospitalized days [2], and estimated total costs [3] for Florida-born children with spina bifida with and without hydrocephalus, stratified by isolated or non-isolated spina bifida by age category, 1998-2007

Hospitalizations	Hydrocephalus		No hydrocephalus		p-value
	Isolated SB (n=242)	Non-isolated SB (n=78)	Isolated SB (n=184)	Non-isolated SB (n=65)	
Birth hospitalizations [4]					
Number of children with hospitalizations	242	78	184	65	
Total number of hospitalizations	242	78	184	65	
Mean (SD) total number hospitalized days	18.9 (20.0)	24.1 (21.9)	9.1 (18.4)	24.9 (37.5)	**<0.0001**
Median (IQR) total number hospitalized days	14.0 (14.0)	16.5 (21.0)	3.0 (6.0)	10.0 (26.0)	
Range for total number hospitalized days	1-149	1-125	1-134	1-221	
Mean (SD) total inpatient costs ($)	33,021 (35,633)	44,767 (46,012)	13,495 (29,741)	52,628 (112,591)	**<0.0001**
Median (IQR) total inpatient costs ($)	25,699 (23,033)	32,482 (31,166)	3,405 (13,998)	16,194 (31,078)	
Range for total inpatient costs ($)	202-275,297	587-309,431	124-269,876	134-706,793	
Post-birth hospitalizations during infancy					
Number of children with hospitalizations	191	62	111	42	
Total number of hospitalizations	431	189	146	118	
Mean (SD) number of hospitalizations	2.2 (1.6)	3.0 (2.5)	1.3 (0.7)	2.8 (2.1)	**<.0001**
Median (IQR) number of hospitalizations	2.0 (2.0)	2.0 (3.0)	1.0 (0)	2.0 (3.0)	
Range for number of hospitalizations	1-11	1-11	1-5	1-11	
Mean (SD) total number hospitalized days	13.7 (22.1)	22.8 (26.8)	5.3 (6.2)	27.8 (45.4)	**<.0001**
Median (IQR) total number hospitalized days	6.0 (13.0)	10.5 (27.0)	4.0 (3.0)	10.0 (27.0)	
Range for total number hospitalized days	1-206	1-115	1-39	1-255	
Mean (SD) total inpatient costs ($)	27,452 (48,637)	47,389 (71,514)	10,873 (10,513)	62,517 (142,195)	**<.0001**
Median (IQR) total inpatient costs ($)	13,246 (20,922)	19,442 (43,723)	8,012 (7,280)	20,943 (43,347)	
Range for total inpatient costs ($)	720-487,037	1,324-400,854	817-70,320	1,254-883,475	

Table 3.4 (continued)

Hospitalizations	Hydrocephalus		No hydrocephalus		p-value
	Isolated SB	Non-isolated SB	Isolated SB	Non-isolated SB	
All infancy hospitalizations (< 1 year)					
Number of children with hospitalizations	260	89	198	67	
Total number of hospitalizations	673	267	330	183	
Mean (SD) number of hospitalizations	2.6 (1.7)	3.0 (2.5)	1.7 (0.8)	2.7 (2.1)	**<.0001**
Median (IQR) number of hospitalizations	2.0 (2.0)	2.0 (3.0)	1.5 (1.0)	2.0 (3.0)	
Range for number of hospitalizations	1-12	1-12	1-6	1-12	
Mean (SD) total number hospitalized days	27.7 (30.0)	37.0 (30.0)	11.5 (18.6)	41.5 (65.1)	**<.0001**
Median (IQR) total number hospitalized days	19.0 (23.0)	26.0 (40.0)	7.0 (7.0)	23.0 (47.0)	
Range for total number hospitalized days	1-216	1-125	1-138	1-476	
Mean (SD) total inpatient costs ($)	50,901 (59,062)	72,247 (70,100)	18,637 (29,419)	90,247 (205,294)	**<.0001**
Median (IQR) total inpatient costs ($)	36,041 (36,286)	48,275 (66,890)	11,974 (15,934)	36,787 (66,269)	
Range for total inpatient costs ($)	445-500,211	1,934-400,855	124-276,261	134-1,590,268	
Hospitalizations during years 1-4					
Number of children with hospitalizations	143	47	37	24	
Total number of hospitalizations	429	188	62	84	
Mean (SD) number of hospitalizations	3.0 (3.1)	4.0 (3.1)	1.7 (1.9)	3.5 (3.5)	**<.0001**
Median (IQR) number of hospitalizations	2.0 (3.0)	3.0 (4.0)	1.0 (1.0)	2.0 (3.5)	
Range for number of hospitalizations	1-19	1-13	1-12	1-15	
Mean (SD) total number hospitalized days	13.2 (23.1)	18.9 (25.9)	10.1 (26.4)	24.3 (44.0)	**0.0038**
Median (IQR) total number hospitalized days	6.0 (12.0)	11.0 (17.0)	4.0 (5.0)	10.0 (23.0)	
Range for total number hospitalized days	1-175	1-142	1-155	1-206	
Mean (SD) total inpatient costs ($)	29,556 (56,075)	34,998 (51,972)	20,584 (60,637)	42,423 (70,164)	**0.0010**
Median (IQR) total inpatient costs ($)	11,356 (21,884)	20,234 (29,844)	6,832 (11,463)	19,120 (24,195)	
Range for total inpatient costs ($)	304-505,528	2,198-290,398	812-361,726	1,077-263,345	

SD =standard deviation, IQR =interquartile range. Note; Statistical significance at $p < 0.05$ (p-value based on Wilcoxon Rank Sum test

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point.

[1] Hospitalizations refer to hospitalizations initiated, but not necessarily completed in age category. Hospitalizations were assessed as continuous episodes of hospital care, regardless of whether a transfer took place. Multiple admission records were merged into one if an infant was admitted to a hospital on the same day as a discharge from a previous admission, or if the infant was admitted to a hospital on the day after a previous discharge with an accompanying "transfer" code.

[2] Hospitalized days refers to the number of days that an infant spent in the hospital for all admissions initiated within the specified age category.

[3] Estimated costs in 2012 U.S. dollars. Estimated costs calculated as total charges adjusted to Florida's statewide average hospital cost-to-charge ratio (Agency for Healthcare Research and Quality, Health Care Utilization Project, 2012). Inpatient charges include all hospital facility charges (excludes professional fees): pharmacy, medical and surgical supplies, laboratory, radiology and other imaging, cardiology, operating room, anesthesia, recovery room, emergency room (if an inpatient hospital admission originated in the emergency room), treatment or observation room (if a visit resulted in an inpatient hospital admission) charges (Agency for Health Care Administration, 2011).

[4] Birth hospitalizations defined as a first hospitalization with age at admission of 0 days or a first hospitalization with an age at admission of 1 day with an accompanying indication of hospital transfer.

Table 3.5 Number of hospitalizations [1], number of hospitalized days [2], and estimated total costs [3] for Florida-born infants with spina bifida by expected health care payer [4] and age category, 1998-2007

Hospitalizations	All payers	Public payer only	Private payer only	Self or under-insured	Multiple payers	p-value
Birth hospitalizations [6]						
Number of children with hospitalizations	569	292 (51.3%)	253 (44.5%)	24 (4.2%)	0	na
Total number of hospitalizations	569	292	253	24		
Mean (SD) total number hospitalized days	17.1(23.2)	19.1(23.4)	14.9(22.5)	16.3(27.1)		**0.0058**
Median (IQR) total number hospitalized days	10.0(17.0)	11.5(18.0)	8.0(14.0)	7.5(19.0)		
Range for total number hospitalized days	0-221	0-150	0-221	1-134		
Mean (SD) total inpatient costs ($)	30,557(52,148)	31,282(39,265)	29,712(63,761)	30,635(55,624)		**0.0151**
Median (IQR) total inpatient costs ($)	18,789(29,411)	23,341(28,989)	16,243(25,107)	14,503(29,928)		
Range for total inpatient costs ($)	124-706,793	124-309,431	166-706,793	172-269,876		
Post-birth hospitalizations						
Number of children with hospitalizations	406	207 (51.0%)	143 (35.2%)	0	56 (13.8%)	
Total number of hospitalizations	884	461	279		144	
Mean (SD) number of hospitalizations	2.2(1.7)	2.2(1.8)	2.0(1.5)		2.6(1.9)	**0.0226**
Median (IQR) number of hospitalizations	2.0(2.0)	2.0(2.0)	1.0(1.0)		2.0(2.0)	
Range for number of hospitalizations	1-11	1-11	1-11		1-11	
Mean (SD) total number hospitalized days	14.2(24.7)	15.4(25.0)	10.4(15.9)		19.8(37.6)	0.0662
Median (IQR) total number hospitalized days	5.0(11.0)	5.0(15.0)	4.0(8.0)		7.0(18.5)	
Range for total number hospitalized days	0-255	0-206	1-85		0-255	
Mean (SD) total inpatient costs ($)	29,592(64,931)	29,970(58,836)	22,279(32,353)		46,867(122,205)	0.6069
Median (IQR) total inpatient costs ($)	11,286(19,758)	10,986(22,789)	10,864(15,431)		15,176(30,503)	
Range for total inpatient costs ($)	720-883,476	720-487,037	817-176,184		1,971-883,476	

Table 3.5 (continued)

Hospitalizations	All payers	Public payer only	Private payer only	Self or under-insured	Multiple payers	p-value
All infancy hospitalizations (< 1 year)						
Number of children with hospitalizations	614	306(49.8%)	236(38.4)%	8(1.3%)	64(10.4%)	
Total number of hospitalizations	1453	738	500	8	207	
Mean (SD) number of hospitalizations	2.4(1.7)	2.4(1.8)	2.1(1.5)	1.0(0)	3.2(2.0)	**<.0001**
Median (IQR) number of hospitalizations	2.0(2.0)	2.0(2.0)	2.0(2.0)	1.0(0)	3.0(2.0)	
Range for number of hospitalizations	1-12	1-12	1-12	1	1-12	
Mean (SD) total number hospitalized days	25.3(34.4)	28.0(32.7)	19.4(24.2)	10.4(10.5)	11.8(21.5)	**<.0001**
Median (IQR) total number hospitalized days	14.0(24.0)	16.5(30.0)	12.0(16.5)	6.5(11.5)	6.5(5.8)	
Range for total number hospitalized days	0-476	0-216	0-168	2-31		
Mean (SD) total inpatient costs ($)	47,884(86,934)	48,947(64,036)	39,049(57,194)	23,618(31,693)	25,179(66,180)	**0.0007**
Median (IQR) total inpatient costs ($)	26,825(39,039)	28,516(40,509)	22,559(33,411)	13,342(27,437)	12,532 (12,460)	
Range for total inpatient costs ($)	1241,590,268	124-500,210	166-454,909	695-96,520	1,441-530,089	

SD =standard deviation, IQR =interquartile range· Statistical significance at $p < 0.05$ (p-value based on Wilcoxon Rank Sum test
Note: Not all children are represented in each time point.
[1] Hospitalizations refer to hospitalizations initiated, but not necessarily completed in age category. Hospitalizations were assessed as continuous episodes of hospital care, regardless of whether a transfer took place. Multiple admission records were merged into one if an infant was admitted to a hospital on the same day as a discharge from a previous admission, or if the infant was admitted to a hospital on the day after a previous discharge with an accompanying "transfer" code.
[2] Length of stay or number of hospitalized days refers to the number of days that an infant spent in the hospital for all admissions initiated within the specified age category.
[3] Estimated costs in 2012 U.S. dollars. Estimated costs calculated as total charges adjusted to Florida's statewide average hospital cost-to-charge ratio (Agency for Healthcare Research and Quality, Health Care Utilization Project, 2012). Inpatient charges include all hospital facility charges (excludes professional fees): pharmacy, medical and surgical supplies, laboratory, radiology and other imaging, cardiology, operating room, anesthesia, recovery room, emergency room (if an inpatient hospital admission originated in the emergency room), treatment or observation room (if a visit resulted in an inpatient hospital admission) charges (Agency for Health Care Administration, 2011).
[4] Payers are expected, but not confirmed, health care payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no insurance, no third party coverage, or less than 30%. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[5] Birth hospitalizations defined as a first hospitalization with age at admission of 0 days or a first hospitalization with an age at admission of 1 day with an accompanying indication of hospital transfer.

Table 3.6 Adjusted multivariable regression results (adjusted prevalence ratios, aPR, and 95% confidence intervals, CI) for the association between selected characteristics and total number of hospitalizations[1] during post-birth hospitalizations, during infancy, and ages one to four years for Florida-born children with spina bifida, 1998-2007

Characteristics	Number of Hospitalizations *Post-birth infancy* (n=406)		Number of Hospitalizations *All infancy* (n=614)		Number of Hospitalizations *Ages 1-4 years* (n=251)	
	aPR	(95% CI)	aPR	(95% CI)	aPR	(95% CI)
Exposure of interest						
Spina Bifida[2] and Hydrocephalus						
Isolated SB without hydrocephalus	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated SB without hydrocephalus	**2.01**	**(1.57-2.56)**	**1.72**	**(1.44-2.06)**	**2.11**	**(1.35-3.31**
Isolated SB with hydrocephalus	**1.63**	**(1.35-1.97)**	**1.53**	**(1.35-1.74)**	**1.82**	**(1.27-2.62)**
Non-isolated SB with hydrocephalus	**2.33**	**(1.87-2.89)**	**1.79**	**(1.52-2.10)**	**2.40**	**(1.62-3.55)**
Predisposing characteristics						
Maternal age (in years)						
<25	1.01	(0.85-1.21)	0.97	(0.85-1.11)	**1.36**	**(1.02-1.80)**
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	0.94	(0.79-1.11)	0.94	(0.83-1.07)	0.97	(0.74-1.28)
Maternal race/ethnicity						
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	**0.78**	**(0.64-0.96)**	0.90	(0.77-1.05)	0.78	(0.56-1.09)
Non-Hispanic Black	0.88	(0.73-1.06)	**0.86**	**(0.75-0.99)**	**0.75**	**(0.58-0.98)**
Other	nr[7]		nr[7]		nr[7]	
Maternal nativity						
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	1.07	(0.88-1.30)	0.93	(0.79-1.08)	0.88	(0.63-1.22)
Maternal marital status						
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	1.10	(0.93-1.29)	1.02	(0.90-1.16)	0.87	(0.70-1.09)

Table 3.6 (continued)

Characteristics	Number of Hospitalizations *Post-birth infancy*		Number of Hospitalizations *All infancy*		Number of Hospitalizations *Ages 1-4 years*	
	aPR	(95% CI)	aPR	(95% CI)	aPR	(95% CI)
Maternal education						
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	0.99	(0.83-1.17)	1.04	(0.91-1.19)	0.81	(0.63-1.05)
Sex of child						
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.94	(0.82-1.07)	0.92	(0.83-1.02)	**0.76**	**(0.62-0.93)**
Enabling characteristics						
Adequate prenatal care [3]						
No	1.00	(1.00)	1.00	(1.00)		
Yes	0.96	(0.82-1.13)	1.03	(0.91-1.16)		
Residential rurality [4]						
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	0.94	(0.78-1.14)	0.96	(0.83-1.11)	**0.54**	**(0.37-0.80)**
Payer type for infancy [5]						
Public payer only	1.06	(0.89-1.26)	1.09	(0.96-1.24)		
Private payer only	1.00	(1.00)	1.00	(1.00)		
Self or under-insured	nr[7]		nr[7]			
Multiple payers	**1.25**	**(1.01-1.54)**	**1.49**	**(1.27-1.76)**		
Need characteristics						
Preterm Birth (< 37 weeks)						
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.16**	**(1.00-1.34)**	1.03	(0.92-1.16)	1.05	(0.84-1.30)

Table 3.6 (continued)

Characteristics	Number of Hospitalizations *Post-birth infancy*		Number of Hospitalizations *All infancy*		Number of Hospitalizations *Ages 1-4 years*	
	aPR	(95% CI)	aPR	(95% CI)	aPR	(95% CI)
Death						
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**0.86**	**(0.66-1.13)**	1.21	(0.99-1.47)	**0.46**	**(0.30-0.70)**

Values in bold are statistically significant. Models are adjusted for all covariables listed.

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point.

[1] Hospitalizations refer to hospitalizations initiated, but not necessarily completed in age category. Hospitalizations were assessed as continuous episodes of hospital care, regardless of whether a transfer took place. Multiple admission records were merged into one if an infant was admitted to a hospital on the same day as a discharge from a previous admission, or if the infant was admitted to a hospital on the day after a previous discharge with an accompanying "transfer" code.

[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.

[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.

[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.

[5] Payer is expected payer, not confirmed payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans benefits. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations

Table 3.7 Adjusted multivariable regression results (adjusted exp(β), and 95% CI) for the association between selected characteristics and total inpatient hospital costs[1] during birth and post-birth hospitalizations, during infancy, and ages one to four years for Florida-born children with spina bifida, 1998-2007

Characteristics	Costs *Birth* (n=569)		Costs *Post-Birth* (n=406)		Costs *All Infancy* (n=614)		Costs *Ages 1-4 years* (n=251)	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Exposure of interest								
Spina Bifida and Hydrocephalus								
Isolated SB without hydrocephalus	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated SB without hydrocephalus	**2.75**	**(1.68-4.51)**	**1.45**	**(1.02-2.05)**	**2.48**	**(1.67-3.68)**	1.30	(0.84-2.01)
Isolated SB with hydrocephalus	**3.70**	**(2.69-5.09)**	1.16	(0.97-1.37)	**2.57**	**(2.06-3.22)**	1.28	(0.91-1.78)
Non-isolated SB with hydrocephalus	**5.59**	**(3.74-8.35)**	**1.43**	**(1.09-1.89)**	**3.61**	**(2.73-4.77)**	1.44	(1.00-2.08)
Predisposing characteristics								
Maternal age (in years)								
<25	0.95	(0.70-1.29)	0.96	(0.78-1.18)	0.98	(0.77-1.24)	0.76	(0.58-1.00)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	1.05	(0.79-1.38)	1.03	(0.85-1.26)	1.06	(0.86-1.31)	0.80	(0.60-1.07)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	0.97	(0.67-1.39)	0.98	(0.79-1.22)	0.95	(0.72-1.24)	1.12	(0.83-1.50)
Non-Hispanic Black	1.29	(0.93-1.80)	1.28	(0.98-1.68)	1.29	(0.98-1.71)	**1.31**	**(1.01-1.70)**
Other	nr[7]		nr[7]		nr[7]		nr[7]	
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	1.08	(0.75-1.55)	1.14	(0.91-1.44)	0.98	(0.74-1.30)	0.91	(0.66-1.26)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	0.98	(0.74-1.29)	0.95	(0.77-1.17)	0.93	(0.75-1.16)	0.98	(0.81-1.18)

Table 3.7 (continued)

Characteristics	Costs *Birth*		Costs *Post-Birth*		Costs *All Infancy*		Costs *Ages 1-4 year*	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	1.18	(0.87-1.62)	0.92	(0.75-1.13)	1.00	(0.78-1.27)	1.10	(0.88-1.36)
Sex of child								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.98	(0.78-1.23)	0.94	(0.81-1.11)	0.96	(0.80-1.15)	0.95	(0.78-1.15)
Enabling characteristics								
Adequate prenatal care [3]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)		
Yes	0.89	(0.65-1.21)	0.83	(0.69-1.01)	0.92	(0.75-1.13)		
Residential rurality [4]								
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	0.78	(0.56-1.07)	0.90	(0.72-1.12)	0.79	(0.61-1.02)	**0.46**	**(0.34-0.61)**
Payer for birth hospitalization [5]								
Public payer	1.21	(0.94-1.57)						
Private payer	1.00	(1.00)						
Self or uninsured	0.81	(0.41-1.62)						
Payer type for infancy [5]								
Public payer only			0.99	(0.80-1.23)	1.14	(0.91-1.43)		
Private payer only			1.00	(1.00)	1.00	(1.00)		
Self or uninsured			nr[7]		nr[7]			
Multiple payers			1.06	(0.82-1.38)	1.15	(0.87-1.53)		

Table 3.7 (continued)

Characteristics	Costs *Birth*		Costs *Post-Birth*		Costs *All Infancy*		Costs *Ages 1-4 year*	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Need characteristics								
Preterm Birth (< 37 weeks)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.34**	**(1.03-1.74)**	0.92	(0.75-1.11)	1.10	(0.89-1.37)	0.88	(0.71-1.10)
Level of nursery care [6]								
Level III	1.00	(1.00)						
Level I or II	**0.40**	**(0.29-0.55)**						
Death [7]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	1.08	(0.61-1.90)	**1.79**	**(1.02-3.15)**	1.12	(0.66-1.90)	**3.81**	**(1.93-7.53)**

Exp(β)=exponent of beta, 95% CI=95% confidence intervals. Values in bold are statistically significant. Models are adjusted for all covariables listed.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point.
[1] Estimated costs in 2012 U.S. dollars. Estimated costs calculated as total charges adjusted to Florida's statewide average hospital cost-to-charge ratio (Agency for Healthcare Research and Quality, Health Care Utilization Project, 2012). Inpatient charges include all hospital facility charges (excludes professional fees): pharmacy, medical and surgical supplies, laboratory, radiology and other imaging, cardiology, operating room, anesthesia, recovery room, emergency room (if an inpatient hospital admission originated in the emergency room), treatment or observation room (if a visit resulted in an inpatient hospital admission) charges (Agency for Health Care Administration, 2011).
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Payer is expected payer, not confirmed payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans benefits. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations
[6] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.
[7] Deaths were those that occurred during the study period, prior to December 31, 2008. Neonatal deaths are deaths < 28 days; infancy deaths are deaths <365 days.

Table 3.8 Adjusted multivariable regression results (adjusted exp(β) and 95% CI) for the association between selected characteristics and total length of stay (or number of hospitalized days) during birth and post-birth hospitalizations, during infancy, and ages one to four years for Florida-born children with spina bifida, 1998-2007

Characteristics	Hospitalized days *Birth* (n=569)		Hospitalized days *Post-Birth* (n=406)		Hospitalized days *All Infancy* (n=614)		Hospitalized days *Ages 1-4 years* (n=251)	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Exposures of interest								
Spina Bifida and Hydrocephalus								
Isolated SB without hydrocephalus	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated SB without hydrocephalus	**2.20**	**(1.77-2.73)**	**3.81**	**(2.60-5.60)**	**3.11**	**(2.22-4.37)**	2.40	(0.99-5.78)
Isolated SB with hydrocephalus	**2.01**	**(1.40-2.88)**	**2.17**	**(1.60-2.95)**	**2.64**	**(2.18-3.19)**	1.38	(0.74-2.56)
Non-isolated SB with hydrocephalus	**3.03**	**(2.22-4.12)**	**4.36**	**(2.93-6.48)**	**3.61**	**(2.74-4.76)**	**2.19**	**(1.13-4.25)**
Predisposing characteristics								
Maternal age (in years)								
<25	1.03	(0.82-1.31)	1.09	(0.75-1.58)	1.04	(0.82-1.31)	1.09	(0.60-1.98)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	1.00	(0.82-1.22)	0.99	(0.73-1.36)	0.98	(0.81-1.20)	0.99	(0.59-1.65)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	0.86	(0.66-1.12)	**0.65**	**(0.45-0.93)**	0.84	(0.66-1.07)	0.72	(0.42-1.23)
Non-Hispanic Black	1.21	(0.93-1.56)	1.44	(0.99-2.10)	1.13	(0.88-1.47)	0.91	(0.52-1.61)
Other	nr[7]		nr[7]		nr[7]		nr[7]	
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	1.23	(0.94-1.61)	1.34	(0.97-1.87)	0.97	(0.75-1.26)	0.78	(0.42-1.45)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	0.95	(0.77-1.16)	1.06	(0.78-1.44)	0.90	(0.73-1.12)	0.93	(0.62-1.42)

Table 3.8 (continued)

Characteristics	Hospitalized days *Birth*		Hospitalized days *Post-Birth*		Hospitalized days *All Infancy*)		Hospitalized days *Ages 1-4 years*	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	1.15	(0.91-1.45)	0.82	(0.58-1.17)	1.03	(0.82-1.31)	0.99	(0.58-1.68)
Sex of child								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.88	(0.74-1.04)	0.81	(0.61-1.06)	**0.84**	**(0.72-0.99)**	**0.57**	**(0.38-0.84)**
Enabling characteristics								
Adequate prenatal care [2]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)		
Yes	0.88	(0.70-1.10)	0.84	(0.60-1.17)	0.94	(0.76-1.15)		
Residential rurality [3]								
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	0.91	(0.72-1.14)	0.81	(0.54-1.20)	0.85	(0.68-1.06)	**0.28**	**(0.13-0.61)**
Payer for birth hospitalization [4]								
Public payer	**1.24**	**(1.03-1.49)**						
Private payer	1.00	(1.00)						
Self or uninsured	0.95	(0.59-1.52)						
Payer type for infancy [4]								
Public payer only			1.32	(0.95-1.83)	**1.31**	**(1.08-1.59)**		
Private payer only			1.00	(1.00)	1.00	(1.00)		
Self or uninsured			nr[7]		nr[7]			
Multiple payers			1.49	(1.00-2.22)	**1.53**	**(1.14-2.06)**		

Table 3.8 (continued)

Characteristics	Hospitalized days *Birth*		Hospitalized days *Post-Birth*		Hospitalized days *All Infancy*)		Hospitalized days *Ages 1-4 years*	
	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)	exp(β)	(95% CI)
Need characteristics								
Preterm Birth (< 37 weeks)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.32**	**(1.07-1.63)**	1.26	(0.91-1.73)	1.18	(0.97-1.45)	0.95	(0.62-1.43)
Level of nursery care [5]								
Level III	1.00	(1.00)						
Level I or II	**0.54**	**(0.44-0.68)**						
Death [6]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	1.04	(0.67-1.62)	**2.42**	**(1.59-3.67)**	1.00	(0.62-1.61)	**5.18**	**(2.37-11.35)**

Note: Exp(β)=exponent of beta, 95% CI= 95% confidence intervals. Values in bold are statistically significant. Models are adjusted for all covariables listed.
Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified. Not all children are represented in each time point.
[1] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[2] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[3] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[4] Payer is expected payer, not confirmed payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations
[5] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.
[6] Deaths were those that occurred during the study period, prior to December 31, 2008. Neonatal deaths are deaths <28 days. Infancy deaths are deaths <365 days
[7] Results are unreliable due to small cell size (<10) and are thus not reported (nr).

CHAPTER 4: FACTORS ASSOCIATED WITH TIMELINESS OF SURGICAL REPAIR OF SPINA BIFIDA

4.1 Introduction

The Institute of Medicine considers adherence to standard clinical guidelines, including timeliness of care, a core component of its framework of quality care [102]. Timely care is important to reduce comorbidities among children with birth defects and can contribute to improved quality of life and lower health care use and costs [16, 17, 105, 206, 207]. Understanding factors associated with timely care among infants with birth defects is useful because birth defects are a leading contributor of disability and pediatric hospitalizations in the U.S. and account for more than $2.6 billion in annual hospital costs [2, 6, 8, 208].

This research focuses on one type of major birth defect, spina bifida (SB), and reports the proportion of Florida-born infants who had timely surgical repair of this birth defect. This study also examines predisposing, enabling, and need characteristics associated with timely surgical repair. Findings from this study can contribute to our understanding of adherence to recommended standards of care for timely surgical repair of SB. Results of this research also add to our understanding of factors that may result in differences in timing of surgical repair and help to inform health education, program planning and development, and management and treatment protocols.

4.2 Literature Review

4.2.1 Epidemiology of Neural Tube Defects and Spina Bifida

Spina bifida (SB), one type of major birth defect, is a neural tube defect that results from a failure of the caudal neural tube to fuse early in embryonic development. Spina bifida occurs when the caudal neural tube does not close completely at some point along the spine between the cervical and sacral regions [10, 11]. Spina bifida occulta ("closed") occurs when a small gap in the spine exists, but no opening occurs on the back, thus the spinal cord and nerves remain essentially intact [10, 11, 41]. This type of SB presents few health care problems and may not be diagnosed at birth [43].

Meningoceles are an "open" type of SB in which a sac of cerebral fluid is exposed through an opening on the infant's back, but no part of the spinal cord is herniated into the sac [11]. This form of SB results in little nerve damage and is typically associated with only minor disabilities [11, 209].

Meningomyeloceles are also "open" SB defects; however, they involve the herniation of both the meninges (the membranes that surround the central nervous system) and the spinal cord into a sac outside the vertebral column [10, 43, 209]. Meningomyeloceles are the most serious and most common presentation of SB and often result in significant disability [11, 41]. A 2012 National Birth Defects Prevention Study used population-based birth defects surveillance data from a ten-state area to examine infants with various presentations of SB [42]. The researchers found that 85.6% of infants with SB had the subtypes of SB that included myelomeningocele, meningocele, and myelocele [42]. Other research reports as many as 90% of children with SB have the subtype myelomeningocele [11, 41].

Spina bifida may present as a single condition in a newborn or SB may be accompanied by other conditions diagnosed at birth or later in life. Isolated SB is SB with the single SB malformation or SB with commonly associated malformations, such as hydrocephalus, orthopedic problems, including scoliosis and lower limb issues, or defects of the urinary system [12, 46, 79, 80, 94]. Isolated SB also includes SB with other minor anomalies, such as low set ears, skin tags, or curved fingers (clinodactyly) [12, 80]. Non-isolated SB is defined as SB with another major, unrelated malformation and without a syndromic diagnosis [12, 79, 80]. Approximately 15% to 25% of infants with SB have non-isolated SB [81, 82]. Children with SB and other birth defects most commonly have orofacial clefts, cardiac defects, and renal or abdominal wall anomalies or have defects that occur as a part of a recognized genetic syndrome [81, 82]

Spina bifida affects approximately 1,500 live-born U.S. infants each year [1] and typically requires life-long, multidisciplinary health care. According to the Florida Birth Defects Registry (FBDR) data, an average of 70 infants with SB were born each year in the state of Florida between 1998 and 2007 [27].

4.2.2 Management and Treatment of Spina Bifida

Fifty years ago, infants born with SB received only palliative care because of a lack of viable options for clinical treatment [46, 60-62]. New medical and surgical interventions now provide hope for both survival and improved quality of life and health outcomes for children born with SB [49, 63, 64].

The Spina Bifida Association's publication *Guidelines for Spina Bifida Health Care Services throughout the Lifespan* comes closest to providing standardized guidelines for the care of individuals with SB [65]. Postnatal surgical closure of the

defect within the first 48 hours of life is the current recommended standard of treatment for SB, particularly for infants with myelomeningocele [65]. Surgical closure includes closing the opening in the spinal column as well as restoring skin and muscle tissues that cover the site [66]. Prompt closure of the site is important because it prevents infection from developing in the exposed nerves and tissues [66]. Prompt surgical repair also protects the exposed nerves and structures from additional trauma [66, 67]. Surgical closure, however, does not restore function lost because of damage to exposed neural tissue.

A growing body of research suggests an alternate prenatal surgical repair of the SB defect [68-73]. The Management of Myelomeningocele Study (MOMS) conducted a randomized control trial in 2003 to compare the safety and efficacy of prenatal surgical repair of the SB defect with that of postnatal repair [68, 74]. Prenatal surgical repair was associated with a reduced need for shunt placement for treatment of hydrocephalus and improved mobility in early childhood [56, 68]. After recruiting 183 of the projected 200 participants, researchers halted the trial because of the efficacy of the prenatal repair [68]. Related to my dissertation research, no Florida hospitals participated in the Management of Myelomeningocele Study clinical trials [68, 74].

Prenatal surgical repair of SB, however, may pose risks to both the mother and fetus. Risks include abruption of the placenta and uterine scarring among mothers, and preterm births among infants [56, 68]. Additionally, the number of facilities in the United States that can perform the surgery is limited [75]. Thus, the adoption of prenatal surgical repair of SB is not yet widespread [56, 76, 77]. The Spina Bifida Association recommends caution in the acceptance of a new standard of care based on a single study

[75] with a sample size that is small and not representative of the U.S. population. The post-natal surgical repair of SB therefore remains the primary method of initial treatment.

4.2.3 Timeliness of Surgical Repair for Infants with Spina Bifida

Experts convened by the Centers for Disease Control and Prevention (CDC) named timeliness of services a public health research priority for selected birth defects, including orofacial clefs, craniosynostosis, congenital heart defects, and Down syndrome [33-36]. In addition to being a core component of the Institute of Medicine's quality of care framework, timely care is important in the reduction of comorbidities among children with birth defects and can contribute to improved quality of life and lower health care use and costs [12, 16, 17, 102-105].

Timely post-natal surgical repair of SB reduces the risk of injury to the exposed neural tissues and reduces the risk of central nervous system or other infections [56]. Timely repair of SB has also been associated with a reduction in the risk of comorbidities, including infection of a ventriculoperitoneal shunt [18], neurogenic bladder [16], and neurodevelopmental delays [19]. In addition, mortality rates appear lower if SB repair occurs in the first 36-48 hours of life [18, 20, 21].

However, timely care among children with birth defects remains an understudied area. To my knowledge, no peer-reviewed research exists on adherence to recommended standards of care of timely surgical repair of SB among infants with SB. Further, no studies report factors associated with the timing of the initial surgical repair for infants with SB. An understanding of timely surgical repair of SB is useful because early surgical repair may reduce mortality and also decrease the likelihood of certain

comorbidities associated with SB, such as neurogenic bladder, neurodevelopmental delays, and ventriculoperitoneal shunt infections [16, 18-21, 107].

4.3 Study Objectives and Hypotheses

The purpose of this study was to examine the timeliness of initial surgical repair and factors associated with surgical repair of the SB lesion among infants with SB. This study reported the proportion of infants with SB who had a surgical repair within the recommended 48 hours of birth and those who had a repair later, more than 48 hours after birth through the end of the first year of life. This research also examined predisposing, enabling, and need characteristics associated with timely repair. The research questions were:

1. What proportion of infants (birth through one year) with spina bifida, who had surgical repair, had a timely surgical repair of the SB lesion (within 48 hours of birth)?
2. What predisposing, enabling, and need characteristics were associated with timely surgical repair of SB?

Based on previous research, I hypothesized that differences in timely surgical repair would vary based on predisposing characteristics (maternal nativity [169, 170], maternal race/ethnicity [168, 169, 171]), enabling characteristics (adequacy of prenatal care [132] and payer type [108, 125, 127, 134]), and need characteristics (preterm birth, nursery level of care at the birth hospital, and presence of comorbidities, such as hydrocephalus [17, 157]).

4.4 Conceptual Framework

The Aday and Andersen *Framework for the Study of Access to Medical Care* provided the conceptual basis for this project [152, 156, 175, 176]. I adapted the Aday and Andersen model to the research questions presented above and included the specific predisposing, enabling, and need variables used in each component of the model (the model is shown in Chapter 2, Figure 2.3, page 37).

4.5 Study Design and Methods

4.5.1 Study Design

This study was a retrospective, statewide, population-based cohort analysis of timeliness of surgical repair for infants with SB born in Florida between January 1, 1998, and December 31, 2007.

4.5.2 Data Acquisition and Study Sample

Data for this study were obtained from linked, longitudinal datasets provided by the Florida Birth Defects Registry (FBDR) within the Florida Department of Health (FDOH), Florida Bureau of Vital Statistics within the FDOH, and the Florida Agency for Health Care Administration (AHCA), which provided the hospital discharge data. Infants with SB without anencephaly born in Florida between 1998 and 2007 were identified using the *International Classification of Disease, 9th revision; Clinical Modification* (ICD-9-CM) codes 741.00-741.93. Hospital discharge data from January 1, 1998 through December 31, 2008, were used to allow for at least one year of hospitalizations for each infant with SB.

Infants who were adopted or prospectively adopted or who were born out of state were excluded by the FBDR. Included infants were live-born in Florida to a mother who

was a Florida resident at the time of delivery and matched with an inpatient hospital discharge record during the first year of life. Infants had to have a birth hospitalization to be included in the study to determine whether or not the infant had SB surgical repair during the first 48 hours of birth. Infants without a recorded birth hospitalization were excluded to reduce error in the analysis because repairs may have occurred at a hospital that did not report hospital discharge data. A total of 290 Florida hospitals reported data to the Florida AHCA during the study period; children with SB were hospitalized in 108 of those reporting hospitals [140]. Non-reporting hospitals included long and short-term psychiatric hospitals, inpatient residential treatment and rehabilitation facilities, and military hospitals in Florida, as well as one non-profit Shriner's Hospital that provided care at no cost to patients [202].

Infants who died during the neonatal period (the first 28 days of life) were excluded from the bivariate and multivariable analyses. Infants who died during the neonatal period likely had more severe or complex medical conditions than infants who survived the neonatal period. Thus, their experience of surgical repair may not be typical of infants with SB. Infants who died later in infancy or during childhood were retained in the bivariate and multivariable analysis to capture the full extent of factors associated with timely surgical repair among infants with SB who had a repair. Figure 1.1 in Chapter 1 (page 9) shows the process for identification of infants for the final study sample.

4.5.3 Primary Outcome of Interest: Timeliness of Surgical Repair

The primary outcome of interest was timely surgical repair of the infant's SB defect among those infants who had a repair recorded in the hospital discharge data at

any point during the first year of life. The ICD-9-CM procedural codes 0351 (meningocele repair) and 0352 (myelomeningocele repair) were used to identify surgical repair of SB. Discussions with several expert clinical consultants from the CDC's National Center on Birth Defects and Developmental Disabilities (NCBDDD) informed the selection of these ICD-9-CM procedural codes.

Using the Spina Bifida Association's recommended guideline of surgical repair within 48 hours of birth [65], infants were considered to have timely repair if the hospital discharge data time-to-procedure code for the repair was on day 0, 1, or 2 of an infant's life (i.e., within first 48 hours of life). Surgical repair on day three or later was considered untimely repair. I coded each infant with a binary variable as having had timely or untimely surgical repair.

4.5.4 Primary Exposures of Interest: Isolated or Non-Isolated Spina Bifida and Hydrocephalus

The primary exposure of interest was a need characteristic, isolated or non-isolated SB. I reported SB for each infant as a dichotomous variable, isolated or non-isolated SB. Infants were classified as having isolated SB if they met any of the following criteria: 1) had only the SB birth defect; 2) had the SB defect and another minor birth defect, such as low set ears or skin tags; or 3) had the SB defect accompanied by a documented sequence of defects related to SB and no additional unrelated major defects [79, 80]. Classification of each infant as having isolated or non-isolated SB was informed by discussions with expert clinical consultants from the CDC's NCBDDD, as well as previous research [12, 79]. Expert clinical consultants from the CDC's NCBDDD manually reviewed approximately 15% of the study sample that required

additional consideration because of multiple conditions. For example, patent ductus arteriosus (PDA) is a heart condition common among premature infants. If an infant with SB had a PDA, the infant was considered to have isolated SB if the infant was premature, but *non-isolated* SB if the infant was born at term. These and other similar situations required a case-by-case review. I referenced surveillance guidelines from the National Birth Defects Prevention Network (NBDPN) for ICD-9-CM diagnostic codes for major birth defects [83]. Appendix A lists ICD-9-CM diagnostic codes considered as major birth defects by the NBDPN for its surveillance and research purposes.

A secondary exposure of interest was the presence of hydrocephalus, also a *need* characteristic. Hydrocephalus was identified by ICD-9-CM codes 741.01- 741.03. Discussions with several expert clinical consultants from the CDC's NCBDDD informed the selection of these ICD-9-CM codes. I reported the presence of hydrocephalus as a dichotomous variable, yes or no. Hydrocephalus cannot be used as a direct predictor of timely repair because an infant may be born with hydrocephalus or may develop hydrocephalus following surgical repair or at a later date [66]. However, the presence of hydrocephalus may serve as a proxy for the more severe forms of SB, based on the high percent of children with myelomeningoceles who have hydrocephalus [11, 41, 210, 211]. Previous research has found that 80% to 90% of infants with myelomeningocele have a chance of developing hydrocephalus that requires placement of a ventriculoperitoneal shunt at birth or sometime thereafter [11, 41, 42, 210, 211].

4.5.5. Covariables Measuring Predisposing, Enabling, and Need Characteristics

I categorized additional covariables as predisposing, enabling, and need characteristics, corresponding to components of the Aday and Andersen conceptual

model. Consistent with the Aday and Andersen conceptual model, I considered the following predisposing characteristics: maternal age, maternal race/ethnicity, maternal nativity, parity, marital status, maternal education, and infant's sex. Enabling characteristics included the variables that measured adequacy of prenatal care, residential rurality, and health insurance payer. Need characteristics included preterm birth, level of nursery care at the birth hospital, an inter-hospital transfer during the birth hospitalization, and the presence or absence of hydrocephalus, and isolated or non-isolated SB. I describe these variables and their coding in detail in the paragraphs that follow.

4.5.5.1 Predisposing Characteristics of Mothers and Infants

Predisposing characteristics of mothers included maternal race/ethnicity, age, education and nativity and marital status. This information was obtained from the FBDR and Florida vital statistics data. I calculated maternal parity by adding the number of live-born infants still living and those live-born now deceased as reported in the FBDR data.

Predisposing characteristics of infants were sex and age. The sex of the infant was obtained from the FBDR data. I calculated the age of the infant using the time to admission variable in the Florida AHCA (hospital discharge) data, which was reported in days.

4.5.5.2 Enabling Characteristics of Mothers and Infants

Enabling characteristics included adequacy of prenatal care and rurality of maternal residential address at delivery, using data obtained from the FBDR and Florida statistics data, and the expected health insurance payer, which was obtained from the

Florida AHCA (hospital discharge) data. Adequacy of prenatal care was determined using the Kotelchuck Index. The Kotelchuck Index creates a ratio comparing the month in which prenatal care is initiated with the total number of prenatal visits prior to delivery to calculate four categories of prenatal care: inadequate (less than 50% of expected visits), intermediate (50-79%), adequate (80-109%), and adequate plus (110% or more) [181, 182]. The Kotelchuck scoring system considers scores less than 80% to be inadequate care [181, 182]. Based on an examination of these data and to ensure adequate cell sizes for meaningful results, I reported adequacy of prenatal care as a binary variable. I used the Kotelchuck cut point of 80% to classify "adequate" and "adequate plus" care as "adequate" prenatal care, and "intermediate" and "inadequate" care as "inadequate" prenatal care.

I identified maternal residential rurality by comparing the geocoded maternal residential addresses reported at birth with the 2000 U.S. Census data that reported rurality by block group level. In the 2000 U.S. Census, the Census Bureau defined "urban" as all territory, population, and housing units located within an urbanized area or in an urban cluster [183]. Urban areas and urban clusters were described by the U.S. Census Bureau as densely settled areas consisting of core census block groups or blocks that had a population density of at least 1,000 people per square mile and surrounding census blocks that had an overall density of at least 500 people per square mile [183]. The U.S. Census Bureau defined all territory, population, and housing units located outside of urban areas or clusters as "rural" [183]. The U.S. Census Bureau assigned a designation to each census block group identifying the geographic area as urban, an urban cluster area, or as rural. Following consultation with spatial research experts at UNC

Charlotte Department of Geography and Earth Sciences and examination of the data, I collapsed urban and urban cluster designations into a single "urban" category. I reported maternal residential "rurality" as a dichotomous variable, "urban" or "rural", to ensure adequate cell sizes for meaningful results.

Health insurance payer was the expected payer source for the birth hospitalization. I defined a birth hospitalization as a first hospitalization with an age at admission of zero days or a first hospitalization with an age at admission of one day with an indication of hospital transfer [177]. I classified health insurance payers for the birth hospitalization as: 1) public, 2) private, or 3) self-insured, under-insured, or no insurance. Public payer sources included Medicare, Medicaid, KidCare (Florida's state children's health insurance program), and Veteran's Administration insurance. Private payer sources included private or employer-based insurance, including military coverage (CHAMPUS or TriCare). The self-pay, no insurance or under-insured category was defined by the Florida AHCA as either no third party coverage or less than 30% estimated insurance coverage [184].

4.5.5.3 Need Characteristics of Mothers and Infants

Need characteristics included preterm birth (less than 37 weeks gestation), low birth weight (less than 2500 grams), and plurality, all obtained from Florida vital statistics data. I reported low birth weight (1500-2499 grams) and very low birth weight (125-1499 grams) separately in the descriptive findings, but collapsed the two categories into one low birth weight category in subsequent analyses because of small cell sizes.

I also considered the level of nursery care at the birth hospital as a *need* characteristic. The American Academy of Pediatrics classifies level of nursery care at the

birth hospital as Level I, II, or III [185, 186]. Level III nursery care provides the most sophisticated care for complex cases [185, 186]. I reported the level of hospital nursery care for the *birth* hospitalization, even if an infant was transferred at birth to a hospital with a higher level of nursery care, in order to most accurately represent an infant's initial hospital experience. I defined a *birth* hospitalization as a first hospitalization with age at admission of zero days or a first hospitalization with an age at admission of one day with an accompanying indication of hospital transfer [177].

Inter-hospital transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a one-day difference existed between a discharge from one hospital and an admission to another hospital [177]. Only inter-hospital transfers that occurred during the birth hospitalization were observed. Inter-hospital transfers were coded as no transfer, transfer by day three of life, or transfer after day 3 of life.

Finally, I reported the death of infants in the study sample, categorized as no death, infancy death ($\leq$ 365 days), or death following infancy (>365 days and before December 31, 2008). Because of small cell sizes, I did not include death as a covariable in bivariate or multivariable analyses.

4.5.6 Statistical Analyses

Descriptive analyses were conducted for the predisposing, enabling, and need characteristics. I reported the mean, median, and range in days for time-to-repair among the infants who had a surgical repair during the first year of life.

Bivariate analyses examined repair in less than or equal to 2 days versus greater than two days by predisposing, enabling, and need characteristics. I conducted chi-

square analyses to determine significance level using a p-value of <0.05. Where appropriate, I used Fisher's exact test to account for small cell sizes. I also compared mean time-to-repair by isolated or non-isolated SB, the presence of hydrocephalus, and the experience of an inter-hospital transfer. I used the Wilcoxon rank sum test to report p-values because of the skewed nature of the continuous outcome.

For the multivariable analysis, I used a modified log-linear Poisson regression with a robust variance estimate to examine how selected predisposing, enabling, and need characteristics were associated with timely surgical repair of SB. Poisson regression is a form of the generalized linear model that provides directly interpretable results in analysis of dichotomous variables, especially when the outcome of interest is not rare [187, 212, 213]. I computed effect estimates, unadjusted prevalence ratios (uPR) and adjusted prevalence ratios (aPR), and corresponding 95% confidence intervals for each covariable to assess the magnitude and precision of the effect estimates.

For the multivariable analysis, I constructed models for: 1) all infants with a recorded surgical repair, 2) infants with a recorded surgical repair and with isolated SB, and 3) infants with a recorded surgical repair and non-isolated SB. I also constructed models comparing timely surgical repair for infants 4) with hydrocephalus and 5) without hydrocephalus. The multiple models allowed me to observe the influence of the two exposures of interest independently and to confirm the results of the model containing all the covariables.

The goal of modeling was to arrive at models that were theory-based, informed by previous research, and parsimonious given the relatively small sample size; thus, selected predisposing, enabling, and need covariables were included in the final regression model.

I excluded parity because no theory or previous research supported its inclusion. I excluded low birth weight because of its close correlation with preterm birth. I excluded plurality because too few infants were part of multiple births to contribute meaningfully to these results. Finally, I excluded transfers because of their correlation with level of nursery care in the birth hospital. Because I excluded infants who died during the neonatal period, I did not control for death in the multivariable analysis. I also ran models separately for each of the categories of theory-based characteristics (predisposing, enabling, and need).

I conducted sensitivity analyses to observe for differences in selected characteristics among the study sample. First, I examined for differences between infants who had a recorded birth hospitalization and those who did not. Second, I examined differences in characteristics between infants who experienced an inter-hospital transfer as part of their birth hospitalization and infants who did not. Infants with no birth hospitalization or infants who were transferred may have been different in terms of their clinical experiences or severity of medical conditions compared to other infants in the study population. I did not examine differences between infants with and without a surgical repair because so many factors that could not be measured in the data set may have contributed to the absence of a recorded repair, including fetal repair or repair at an out-of-state hospital or at a Florida hospital that did not report data to the Florida AHCA.

I assessed individual variables used in the multivariable analysis for multicollinearity. There was no evidence of notable multicollinearity based on recommended maximum levels of the variance inflation factor [189, 190].

Analyses were conducted using SAS 9.2 statistical software (SAS Institute, Inc., Cary, NC). This study was approved by the Institutional Review Boards at the University of North Carolina Charlotte, the FDOH, and at the CDC's NCBDDD.

4.6 Results

4.6.1 Study Population

The FBDR data identified 914 Florida-resident infants who were born between January 1, 1998, and December 31, 2007, with an ICD-9-CM code indicating a neural tube defect. Of these 914 infants, 668 had ICD-9-CM codes associated with the neural tube defect, SB without anencephaly. Of the 668 infants with SB without anencephaly, 614 linked to at least one inpatient hospital discharge record.

Infants in the FBDR who did not have a linked hospital record were significantly more likely to be born to mothers who were less educated ($p=0.0011$), foreign-born ($p<.0001$), and of Hispanic ethnicity ($p= 0.0044$) than infants who had a linked hospital record (results not shown). There were no significant differences in maternal age, marital status, infant's sex and birth weight, or death between infants with or without a linked hospital record (results not shown).

Of the 614 infants with a linked hospital record, 569 infants had a first hospitalization recorded in the Florida AHCA dataset that was also the infant's birth hospitalization. Infants in the FBDR who did not have a linked birth hospitalization were more likely to be born to younger mothers ($p=0.0034$), mothers who were rural residents ($p=0.0206$), and mothers were not born in the United States ($p=0.0031$) (results not shown). I found no significant differences in maternal race/ethnicity, maternal education,

marital status, and infant's sex and birth weight between infants with and without a birth hospitalization (results not shown).

Among the 569 infants with a birth hospitalization, 299 had a recorded SB repair and no death in the neonatal period. This group of 299 comprised the final sample for analysis.

4.6.2 Descriptive Results

4.6.2.1 Descriptive Characteristics of the Mothers and Infants

Table 4.1 shows the descriptive characteristics of the mothers and infants in this study. Among all infants with a birth hospitalization (n=569), 301 (52.9%) had a recorded surgical repair of the primary defect and two died during the neonatal period, leaving a study sample of 299 infants. About 61% (n=181/299) of mothers were married. Most mothers were born in the United States (76.6%, n=229/299), had at least a high school diploma (74.9%, n=224/299), and received adequate prenatal care (75.9%, n=227/299).

About 54% (n=161/299) of infants were born to non-Hispanic White mothers. About 11% (n=32/299) of infants were born low birth weight (1500-2499 grams) and 2.7% were born very low birth weight (n=8/299); 25.4% (n=76/299) of infants were born preterm. About 80% (n=240/299) of infants had hydrocephalus and 20.4% (n=61/299) had non-isolated SB.

Among the 299 infants, 27.8% (n=83) had an inter-hospital transfer during their birth hospitalization. About 96% of those transfers (n=80/83) occurred by day three of the infant's life. Infants who had an inter-hospital transfer were more likely to have *non-isolated* SB (p=0.0037) and to have private insurance (p=0.0368) than infants who were

not transferred (results not shown). Infants born at a hospital with Level III nursery care were less likely to be transferred at birth ($p<0.0001$) (results not shown). There were no differences between infants who were transferred and those who were not, based on maternal race/ethnicity, maternal age and education, maternal nativity or marital status, or sex or gestational age of the infant (results not shown).

4.6.2.2 Descriptive Results for Time-to-Repair for Spina Bifida

Table 4.2 shows the descriptive results for time-to-repair. Mean time-to-repair for all infants was 11.3 days (standard deviation, SD, 37.0 days) and median time-to-repair was 1.0 day (interquartile range, IQR, 3.0 days). While mean time-to-repair varied, infants with isolated SB, non-isolated SB, and hydrocephalus all had a median time-to-repair of 1.0 day. The range for time-to-repair was 0 to 305 days.

Table 4.3 shows time-to repair by categories. Of the 299 infants with a repair, 68.6% (n=205) of infants had the surgical repair by day two of life. About 15% (n=45/299) of infants had a surgical repair between days three and seven. About 16% (n=49/299) of infants had a surgical repair after day seven, but before the end of their first year of life.

4.6.3 Bivariate Results

4.6.3.1 Bivariate Results for the Primary Exposures of Interest: Isolated or Non-Isolated SB and Hydrocephalus

Table 4.1 presents the results of bivariate analysis for the dichotomous outcomes of timely (<48 hours after birth, day 0-2) or untimely repair (after day 2). Examining the primary outcome of interest, the variable isolated or non-isolated SB was not

significantly associated with time of repair (p=0.2373). However, the presence of hydrocephalus was significantly associated with having a timely repair (p<0.0001).

Table 4.2 presents the results for time-to-repair, stratified by isolated or non-isolated SB and by the presence of hydrocephalus. The variable isolated or non-isolated SB did not significantly affect the timeliness of SB repair; however, there was a significant difference in average time-to-repair comparing infants with and without hydrocephalus (p<0.0001). Infants with hydrocephalus experienced a mean time-to-repair of 5.3 days (SD: 21.3); infants without hydrocephalus experienced a mean time-to-repair of 35.5 days (SD: 66.4 days). Median values were 1.0 day (IQR, 1.5 days) for infants with SB with hydrocephalus and 5.0 days (IQR, 36.0 days) for infants with SB without hydrocephalus.

4.6.3.2 Bivariate Results for Predisposing, Enabling, and Need Characteristics

Table 4.1 also presents the results of bivariate analysis comparing predisposing, enabling, and need characteristics between infants with and without a timely repair. I observed no associations between the timing of repair and the predisposing or enabling characteristics. Among need-related characteristics, infants who were born preterm (p=0.0486) and born in a hospital with Level III nursery care (p=0.0012) had a greater likelihood of a timely SB repair. From Table 4.1, the rate of timely repair among infants who were born preterm was 77.6% (number of preterm births with timely repair/total number of preterm births; n=59/76) compared to slightly lower percentage of 65.5% for full-term infants (number of term births with timely repair/total number of term births; n=146/223). Also calculated from Table 4.1, infants who were born in a hospital with Level III nursery care had a much higher percent of infants with a timely repair

(178/245=72.7%) compared with infants born in a hospital with a lower level of nursery care (27/54= 50%).

4.6.4 Multivariable Results

4.6.4.1 Multivariable Results for the Primary Exposures of Interest: Isolated or Non-Isolated SB and Presence of Hydrocephalus, for All Infants

Table 4.4 shows the unadjusted (uPR) and adjusted (aPR) prevalence ratios (PR) and 95% confidence intervals (CI) for models that include the exposures of interest (hydrocephalus and isolated or non-isolated SB) and all other selected predisposing, enabling, and need covariables. The variable isolated or non-isolated SB was not statistically associated with a timely repair in either the unadjusted or the adjusted models (aPR=0.99, 95% CI: 0.80-1.21). Infants with SB and hydrocephalus, however, was associated with about an 80% increased likelihood for timely repair in both adjusted and unadjusted models [(uPR=1.82, 95% CI: 1.33-2.50) and (aPR=1.80, 95% CI: 1.31-2.48)].

4.6.4.2 Multivariable Results for Predisposing, Enabling, and Need Characteristics for All Infants

Table 4.4 further shows the unadjusted and adjusted PR and 95% CI for the selected predisposing, enabling, and need characteristics for all infants, including the need characteristics and exposures of interest, isolated or non-isolated SB and hydrocephalus. When the model was adjusted for all predisposing, enabling, and need characteristics, the only characteristic associated with timely repair was health payer status (other than the exposure of interest hydrocephalus that was reported in section 4.6.4.1). Infants who were self-pay or underinsured were more likely to have a timely

repair (aPR: 1.39, 95% CI: 1.07-1.82). However, this finding was based on only 13 observations and should be interpreted with caution.

4.6.4.3 Multivariable Results for Infants Stratified by the Presence of Hydrocephalus

Table 4.4 also shows the unadjusted and adjusted PR and 95% CI for the variable isolated or non-isolated SB and for selected covariables, comparing models of infants with and without hydrocephalus. The primary exposure of interest, isolated or non-isolated SB, was not associated with timely repair among infants with or without hydrocephalus.

Examining predisposing, enabling, and need characteristics, infants with or without hydrocephalus who were self-pay or under-insured were more likely to undergo timely repair (with hydrocephalus: aPR=1.37, 95% CI: 1.14-1.65; without hydrocephalus: aPR=4.20, 95% CI: 1.46-12.1). However, these results are based on very small sample sizes and should be interpreted with caution. There were three differences between the adjusted models for infants with and without hydrocephalus. Among infants without hydrocephalus, younger maternal age (aPR=2.46, 95% CI: 1.10-5.48) and inadequate prenatal care (aPR=3.93, 95% CI: 1.36-11.3) were associated with timely repair, compared with mothers who were 25 and older or who had adequate prenatal care. Infants without hydrocephalus born to mothers of a racial or ethnic minority group were less likely to undergo a timely repair (Hispanic: aPR=0.34, 95% CI: 0.13-0.87; non-Hispanic Black: aPR=0.22, 95% CI: 0.06-0.82) compared with infants born to non-Hispanic White mothers (referent group). No predisposing, enabling, or need characteristics were associated with timely repair among infants with hydrocephalus, except for the self-pay status, which was based on a small cell size.

4.6.4.4 Multivariable Results for Infants Stratified by Isolated or Non-Isolated SB

Table 4.5 shows the unadjusted and adjusted PR and 95% CI for the exposure of interest hydrocephalus and selected covariables, comparing models of infants by isolated or non-isolated SB. In the adjusted models, infants with isolated SB and hydrocephalus were 85% more likely to have a timely repair (aPR=1.85, 95% CI: 1.27-2.70) compared with infants with isolated SB without hydrocephalus. In contrast, there was no association between hydrocephalus and timely repair among infants with non-isolated SB.

Table 4.5 also shows the unadjusted and adjusted PR and 95% CI for the selected predisposing, enabling, and need characteristics, comparing infants with isolated or non-isolated SB. Infants with isolated or non-isolated SB who were self-pay or under-insured were more likely to have timely repair (isolated SB: aPR=1.40, 95% CI: 1.05-1.85; non-isolated SB: aPR=3.01, 95% CI: 1.05-8.66) compared with infants who had a private payer; however, these findings are based on very small sample sizes and should be interpreted with caution. Infants born preterm with isolated SB had a marginally greater likelihood of timely repair (aPR=1.17, 95% CI: 0.99-1.38) compared with infants born at full-term. Infants who were born preterm with non-isolated SB had a lower likelihood of a timely repair (aPR=0.38, 95% CI: 0.15-0.97) compared with infants born at full-term. Among infants with non-isolated SB, infants born to unmarried mothers were much more likely to have a timely repair (aPR=2.29, 95% CI: 1.36-3.86) compared to mothers who were married.

4.7 Discussion

4.7.1 Discussion of Timeliness of SB Repair

This study examined the timeliness of surgical repair among infants with SB and factors associated with timely repair of SB. I found that among infants who had a surgical repair, 68.6% had their repair within the recommended 48 hours of birth. The mean time-to-repair was 11.3 days, while the median time-to-repair was 1.0 days, suggesting that a small number of infants had substantially longer times to their surgical repair.

Previous studies that examined time-to-repair for SB are sparse, and these studies were conducted with infants born in the 1970s or 1980s [18, 20, 214]. Thus, findings from these studies may be outdated. One study of long-term outcomes for 101 infants born with meningomyelocele born between 1971 and 1981 observed that 56.4% of infants in the study had a surgical repair within 48 hours of life; 66.3% had a repair within the first week of life [20]. A hospital-based study of 110 infants born with myelomeningocele between 1978 and 1982 found that 47.3% of the infants had repair within 48 hours of birth, an additional 29.1% by the first week of life, and 10.9% between one week and ten months of life [214]. Researchers conducting a study of hydrocephalus that included 31 infants with myelomeningoceles treated between 1984 and 1987 found that surgical closure of meningomyeloceles occurred on average by 2.4 days [18].

The studies just described used small convenience samples. Further, these studies focused on infants with myelomeningocele, so exact comparisons cannot be made to this study. In contrast, I used ICD-9-CM codes that do not distinguish between diagnoses of myelomeningocele, meningocele, and myelocele. Thus, my study sample included infants who were more severely affected by SB as well as infants less severely affected. Findings from my research suggest that a higher percent of infants had surgical repair of

SB within the first two days of life (68.6%) than in previous research, which indicated an improvement in time to repair among infants with SB [20, 214]. Because infants with less severe forms of SB (meningoceles) may not require immediate surgical repair, the 68.6% of repairs in two days or less may underestimate the percentage of timely repairs for infants with more severe forms of SB (myelomeningoceles). This finding may indicate an even greater improvement in timely repair among infants with the most serious presentations of SB.

4.7.2 Discussion of Effects of Hydrocephalus and Isolated or Non-Isolated SB on Timely Surgical Repair of Spina Bifida

I expected that non-isolated SB might delay the surgical repair of SB because of the increased complexity of medical care an additional major birth defect might require. However, the variable isolated or non-isolated SB was not associated with the timing of the SB repair. This finding was unexpected. The additional diagnoses associated with non-isolated SB may not require the type of care that would influence surgical repair of SB. For example, certain heart conditions may be treated with drug therapy, or certain chromosomal defects or heart conditions may be monitored until further management and treatment becomes necessary after several months of life. Other conditions, such as orofacial clefts (cleft lip with or without cleft palate and cleft palate alone), may require surgical intervention, but not necessarily in the first few days of life and usually are not life threatening. The broad categories of isolated and non-isolated SB may not provide adequate detail to observe for the effects of comorbid conditions on the timely repair of SB.

In contrast, infants who had hydrocephalus were consistently more likely to have a timely repair than infants who did not have hydrocephalus. It is unlikely that hydrocephalus predicted timely repair. Instead, hydrocephalus, which appears in 80% to 90% of infants with myelomeningocele [11, 41, 42, 56, 66], may serve as a proxy for the myelomeningocele presentation of SB. As previously described, myelomeningocele is the most serious form of SB and typically requires prompt surgical repair; thus, this finding of timely repair among infants with hydrocephalus is consistent with the degree of severity of SB and recommended treatment [11, 41, 42, 56, 66].

4.7.3 Discussion of Effects of Other Covariables on Timely Surgical Repair of Spina Bifida

In the bivariate analysis, I found that infants born preterm or born in a hospital with Level III nursery care were more likely to have a timely surgical repair. The rate of timely repair among preterm infants (a need characteristic) was substantially higher than among full term infants (77.6% versus 65.5%). Infants who were born in a hospital with Level III nursery care (a need characteristic) had a much higher rate of timely repair than infants who were born in a hospital with a lower level of nursery care (72.7% versus 50%). The higher rate of timely repair among preterm infants may be associated with delivery at hospitals with higher levels of nursery care and the availability of more sophisticated treatment options [173, 174].

In adjusted multivariable analysis, few predisposing, enabling, or need characteristics were associated with timely surgical repair. First, somewhat unexpectedly, infants who were self-pay or under-insured were more likely to have a timely surgical repair compared with infants with private insurance (an enabling

characteristic). I expected under-insured infants to have a delay in timely repair, based on previous research that found that adequate insurance was the most critical determinant of access to care among children with special health care needs [127]. This finding should be interpreted with caution, however, as the number of infants who were self-pay or under-insured was very small (n=13). This finding highlights the need for additional population-based research among children who have different health care payers across multiple hospitalizations, to examine the influence of changes in payer status on health resource use and health outcomes.

In the stratified models, the presence of hydrocephalus and non-isolated SB may indicate that these infants have more severe or complex medical conditions. Among infants with hydrocephalus or non-isolated SB, there were few notable associations among predisposing and enabling characteristics and timeliness of SB repair. While, maternal marital status and payer status had statistically significant associations, the cell sizes for each were small and should be interpreted with caution. However, among infants with isolated SB or without hydrocephalus (possibly less severe or less complex medical conditions), several predisposing and enabling characteristics were associated with the timeliness of surgical repair including maternal age, maternal race/ethnicity, and adequacy of prenatal care. In this study, the infants with the most complex presentations of SB had fewer factors that influenced time-to-repair, while infants with less complex presentations had more factors that influenced time-to-repair. These findings may suggest that in the face of increased clinical need, the effects of predisposing and enabling factors may be limited. In this study sample, timely SB repair may be critical to an infant's survival and thus becomes an important predictor of timing of the repair. The

fact that the infants in this study were already hospitalized, and thus had access to care, may also reduce the effect of predisposing and enabling characteristics.

In summary, findings of this study suggest that the clinical need for repair, indicated by the presence of hydrocephalus, is the most important factor in predicting timely surgical repair of SB. Premature birth and the level of nursery care at the birth hospital may also influence timely repair. While most infants had a timely surgical repair, the difference between mean and median time-to-repair among all infants suggests that a small group of infants experienced a notably delayed repair.

4.8 Strengths and Limitations

4.8.1 Innovation and Strengths in the Research Topic

This study examined differences in the timely surgical repair of SB in a population-based, statewide study of unduplicated Florida infants insured by different health care payer types. The study reported the proportion of infants who had repair within the recommended 48 hours following birth. This study also examined predisposing, enabling, and need characteristics associated with timely repairs. No previous studies have examined adherence to the recommended guidelines for surgical repair of SB and its associated factors. Therefore, these topics represent new or expanded areas of research.

4.8.2 Innovation and Strengths in Methodology

The study population for this research was an important strength in that it represented a diverse group of infants. The state of Florida was the fastest growing and fourth most populous state according to the 2000 U.S. Census [22, 23]. Florida was fourth in number of annual live births, second in number of live births to non-Hispanic

Black women, and third in number of live births to Hispanic women, nationwide during the study period of 1998-2008 [22-24]. The state of Florida also supports a statewide, population-based birth defects registry and a statewide agency for the collection of hospital discharge data that provided information for this project. The statewide, population-based study population used in this study included linked, longitudinal data from the FBDR and the Florida AHCA, and thus provided a robust, diverse source of information for this project.

Additionally, this research incorporated several methods that are not frequently used in health services research for birth defects and thus are both strengths and innovations of the project. First, because of the nature of the data, the unit of analysis for this project was the individual infant, rather than the more typical observation of aggregate level data, such as the hospital visit. Secondly, the dataset provided access to hospital discharge data for infants from multiple payer sources, rather than the more commonly researched single payer source. This allowed for a more complete picture of timeliness of surgical repair across multiple payers.

A final strength of this methodology is its ability to be replicated by health services researchers and public health researchers examining different types of birth defects in collaboration with other birth defect registries and state and federal agencies. This research could also be replicated using hospital discharge or paid claims data, such as Medicaid, from other states and federal and state public health agencies. These methods underscore the value of collaboration between local, state and federal public health agencies, academic universities, and public health and health services researchers.

Regarding generalizability, the demographic findings are similar for all Florida-born infants during the study period with a few exceptions. Compared to all infants born in Florida 1998-2007, the study population included a slightly lower proportion of Hispanic mothers (29% statewide compared to 24% in the study sample), a higher proportion of preterm births (26% compared to 11%) and a slightly higher proportion of infants with public payer sources (51% compared to 43%)[27]. The higher percentage of preterm births was consistent with previous research that has reported an association between low birth weight and prematurity among infants with birth defects [167, 172, 196].

4.8.3 Limitations

Several limitations were due to the inherent nature of administrative data. The FBDR identified infants for this study using passive surveillance methodologies for identifying infants with birth defects. While widely used, passive birth defects surveillance systems do not actively verify the birth defect diagnosis by review of medical records, hospital charts, or nursery logs. In addition, limited information on prenatal diagnosis is available in passive birth defects surveillance systems [27]. While passive surveillance techniques may lead to under-reporting or miss reporting of infants with birth defects or a specific defect type [27-29, 200, 201], the FBDR's overall completeness of ascertainment of birth defects has been estimated at 86.6%, with case ascertainment variation noted by specific defect [28, 29]. Because SB is relatively easy to detect, passive surveillance may be less of a limitation than with other birth defects that are more difficult to detect. In the FBDR data, case ascertainment for SB without anencephaly was 88.0% [29]. In addition, because this analysis used data from the

FBDR, it is a state-specific study, which may limit generalizability to other states or regions of the country.

Another limitation was the use of hospital administrative data. Administrative data may be at risk for error or inconsistent coding that could incorrectly code hospital facility codes or introduce error in diagnostic or procedural coding. These data did not include information on families that sought care out-of-state for their infant. Additionally, while approximately 290 Florida hospitals report data to the Florida AHCA, not all are required to report, including one non-profit Shriner's Hospital that provides care at no cost to patients, as well as long and short-term psychiatric hospitals, inpatient residential treatment and rehabilitation facilities, and military hospitals in Florida [202]. The lack of reporting to the Florida AHCA meant I was not able to capture data on all the infants within the Florida SB population under study. However, because data from 108 different Florida hospitals are represented in the data set [140] and because most of the non-reporting hospitals do not provide newborn care, the amount of data lost was likely very limited and thus the findings of this research may be generalizable at least to the state of Florida.

Another limitation in the use of administrative data was its lack of additional information that could have contributed further insights into the timely repair of SB. For example, I was unable to determine if an infant was prenatally diagnosed with SB. I also did not know from the data if an infant had prenatal surgical repair of SB. While information on prenatal surgical repair of SB was not available from the dataset, I believe that no prenatal repairs occurred within the study sample in Florida after February 2003. For the duration of the Management of Myelomeningocele clinical trial (February 2003

through December 2010), all cases of fetal repair of myelomeningocele were referred to one of the three research centers: The Children's Hospital of Philadelphia in Philadelphia, PA; Vanderbilt University in Nashville, Tennessee; and the University of California San Francisco in San Francisco, CA [68]. All other pediatric surgery centers in the United States not participating in the study agreed not to perform fetal surgery for SB for the duration of the clinical trial (February 2003 through December 2010) [68]. The Management of Myelomeningocele clinical trial overlapped the January 1, 1998 – December 31, 2008 study period of this research and thus limited the number of infants that might have had prenatal SB repair in Florida. The effects of this concurrent research on the outcomes of this study are unknown.

In addition to having no information on prenatal diagnosis and prenatal surgical repair, the administrative data used in this study did not provide information on other factors, such as possible medical contraindications to the repair or insights into the family's decision-making processes related to the care of their infant. Personal family decisions could potentially affect where and when the surgery took place. A family, for example, might choose care in a location with family nearby or might request a referral to a specific pediatric hospital.

Finally, ICD-9-CM codes differentiate between SB with and without hydrocephalus and include indicators for the spinal level of the SB lesion. However, ICD-9-CM codes do not specifically differentiate between myelomeningocele, meningocele, and myelocele cases. While myelomeningoceles typically require immediate surgical repair, other presentations, such as meningoceles, may not need urgent repair. This lack of differential diagnoses limited the exploration of SB repair by

type and severity, and thus limits interpretation of the findings. The new ICD-10-CM codes that will be implemented October 1, 2014, do not provide any updates in differentiation of SB by type [215]. Future research that uses active birth defects surveillance systems that can provide additional clinical information and differential diagnoses on larger study populations, may contribute to a better understanding of factors associated with timely repair of SB.

4.9 Implications for Public Health Practice and Research

The findings of this study have several implications for public health policy and practice. First, the results reinforce the value and importance of having prenatal care and prenatal diagnosis of SB. Delivery of an infant with SB in a hospital with Level III nursery care was associated with timely repair among infants who had a repair. This suggests that access to an appropriate level of health care may improve the opportunity for a timely repair among infants with SB and potentially reduce the risk of long-term comorbidities [16, 18-21, 56]. Health care providers should clearly communicate the value of maternal serum alpha-fetoprotein screening for SB, which typically is offered to all pregnant mothers during their second-trimester [216]. Prenatal diagnosis of SB can facilitate appropriate counseling [217, 218] and possible planning for the infant's birth location and experience. Programs that advocate for SB awareness, such as the Spina Bifida Association of Central Florida's 2012 campaign: *Redefining Spina Bifida* [219], are important for increasing public awareness of SB and perhaps reducing barriers to prenatal screening.

The study of the timeliness of surgical repair of SB warrants further research on several fronts. First, an analysis of a national hospital use dataset with individual-level

data is needed to provide nationwide information on the proportion of infants with SB who undergo surgical repair. MarketScan® Commercial and Medicaid databases provide de-identified, patient-level data across both private and public payer sources [220], thus making them possible resources for nation-wide research. Research that uses data from the Agency for Healthcare Research and Quality's (AHRQ) Healthcare Cost and Utilization Project (HCUP), including the Kids' Inpatient Database (KID), would not provide individual-level data, but could provide further insights to SB repair [221]. Knowledge of the proportion of infants who had surgical repair of SB can provide a baseline comparison for later population-based studies. Additional population-based research that examines timeliness of care, the different presentations of SB, and associations with later health outcomes could provide valuable information that informs both clinical practice and standards of care. An understanding of patterns and predictors of timely care, including timely repair of SB, are important to inform efforts by public health practitioners, health care planners and coordination of delivery of services by governmental providers, such as federal, state, and local agencies, particularly those serving CSHCN [13].

Second, collaborative multi-state, population-based studies linking multiple birth defect registries and linked, longitudinal data would be useful to examine both proportion and timeliness of surgical repair of SB and related factors. Collaborative research projects would increase study sizes, thus increasing study power and potentially the ability to observe effects over time. Continued support and expansion of the National Spina Bifida Patient Registry [204], as well as continued funding for the National Birth Defects Prevention Network, National Birth Defects Prevention Study, and state birth

defects surveillance systems to advance the knowledge of health care utilization among individuals with SB will be extremely important for these efforts.

Third, qualitative research could be important to gaining a deeper understanding of family or parental attitudes and opinions that may influence decisions and timing related to surgical repair of SB. A better understanding of qualitative findings, such as feelings, attitudes, pressures and fears, prenatal experiences, and reasons for SB repair or reasons for selecting a particular hospital for SB repair, could inform methods for better communication during this difficult time of decision-making.

Finally, the findings of study can help to inform research for other birth defects, particularly those that require surgical repair. Birth defects, such as gastroschisis (a defect of the abdominal wall) and orofacial clefts, typically require surgical repairs, and like SB, may have multiple presentations and factors that influence their surgery and its timing. A better understanding of current practices and factors related to the repair of SB and other major birth defects requiring surgical repair can improve access to care and adherence to standards of timeliness of care. Increased access to appropriate and timely care may improve mortality and reduce associated morbidities for this population of children, thus reducing health costs and increasing long-term quality of life and health outcomes.

Table 4.1 Selected characteristics for Florida-born infants with spina bifida by timing of primary surgical repair of spina bifida, 1998-2007

Characteristics	All infants [1] (n=299)		Timing of surgical repair				p-value
			Timely Repair Day 0-2 (n=205, 68.6%)		*Untimely Repair* After Day 2 (n=94, 31.4%)		
	n	(%)	n	(%)	n	(%)	
Exposures of interest							
Spina bifida [2]							
Isolated	238	(79.6)	167	(81.5)	71	(75.5)	0.2373
Non-isolated	61	(20.4)	38	(18.5)	23	(24.5)	
Hydrocephalus							
Yes	240	(80.3)	180	(87.8)	60	(63.8)	**<0.0001**
No	59	(19.7)	25	(12.2)	34	(36.2)	
Predisposing characteristics							
Maternal age (in years)							
<25	116	(38.8)	83	(40.5)	33	(35.1)	0.6712
25-29	86	(28.8)	57	(27.8)	29	(30.9)	
≥30	97	(32.4)	65	(31.7)	32	(34.0)	
Maternal race/ethnicity							
Non-Hispanic White	161	(53.8)	113	(55.1)	48	(51.1)	0.8684
Hispanic	73	(24.4)	50	(24.4)	23	(24.5)	
Non-Hispanic Black	60	(20.1)	39	(19.0)	21	(22.3)	
Other	5	(1.7)	3	(1.5)	2	(2.1)	
Maternal nativity							
Born in U.S.	229	(76.6)	161	(78.5)	68	(72.3)	0.2401
Foreign-born	70	(23.4)	44	(21.5)	26	(27.7)	
Maternal marital status							
Married	181	(60.5)	120	(58.5)	61	(64.9)	0.2964
Not married	118	(39.5)	85	(41.5)	33	(35.1)	
Maternal parity							
First child	114	(38.1)	73	(35.6)	41	(43.6)	0.1857
Second or subsequent child	185	(61.9)	132	(64.4)	53	(56.4)	
Maternal education							
High school diploma or more	224	(74.9)	154	(76.2)	70	(74.4)	0.8565
No high school diploma	71	(23.7)	48	(23.8)	23	(24.5)	
Sex of infant							
Female	155	(51.8)	109	(53.2)	46	(48.9)	0.4963
Male	144	(48.2)	96	(46.8)	48	(51.1)	

Table 4.1 (continued)

Characteristics	All infants [1]		Timing of surgical repair				p-value
			Timely Repair Day 0-2		*Untimely Repair* After Day 2		
	n	(%)	n	(%)	n	(%)	
Enabling characteristics							
Prenatal care [3]							
Adequate prenatal care	227	(75.9)	153	(74.6)	74	(78.7)	0.4636
Inadequate prenatal care	58	(19.4)	42	(20.5)	16	(17.0)	
Missing	14	(4.7)	10	(4.9)	4	(4.3)	
Residential rurality [4]							
Urban /urban cluster	254	(85.0)	171	(83.4)	83	(88.3)	0.2729
Rural	45	(15.0)	34	(16.6)	11	(11.7)	
Payer for birth hospitalization [5]							
Public payer	158	(52.8)	111	(54.2)	47	(50.0)	0.2746
Private payer	128	(42.8)	83	(40.5)	45	(47.9)	
Self or uninsured	13	(4.4)	11	(5.4)	2	(2.1)	
Need characteristics							
Preterm Birth (< 37 weeks gestation)							
Yes	76	(25.4)	59	(28.8)	17	(18.1)	**0.0486**
No	223	(74.6)	146	(71.2)	77	(81.9)	
Birth Weight (in grams)							
Normal weight (≥2500)	259	(86.6)	178	(86.8)	81	(86.2)	0.4855
Low birth weight (1500-2499)	32	(10.7)	23	(11.2)	9	(9.6)	
Very low birth weight (<1500)	8	(2.7)	4	(20.)	4	(4.3)	
Singleton birth							
Yes	293	(98.0)	201	(98.1)	92	(97.9)	0.9195
No	6	(2.0)	4	(1.9)	2	(2.1)	
Level of nursery care at birth hospital [6]							
Level III	245	(81.9)	178	(86.8)	67	(71.3)	**0.0012**
Level I or II	54	(18.1)	27	(13.2)	27	(28.7)	
Inter-hospital transfer [7]							
No inter-hospital transfer	216	(72.2)	154	(75.1)	62	(66.0)	0.1466
Transfer within 3 days of birth	80	(26.8)	50	(24.4)	30	(31.9)	
Transfer after 3 days	3	(0.01)	2	(2.1)	1	(0.5)	
Death [8]							
No death	286	(95.6)	195	(95.1)	91	(96.8)	0.7880
Death during infancy	5	(1.7)	4	(2.0)	1	(1.1)	
Death during ages 1-4 years	8	(2.7)	6	(2.9)	2	(2.1)	

Note: Columns may not add to 100% because of missing or unknown values. P-values in bold are considered statistically significant at <0.05. Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.

[1] Excludes infants with death during neonatal period (≤ 28 days).

[2] Isolated spina bifida is defined as SB with no additional major coded defects, other than the sequence of defects related to SB.

[3]Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care".
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no insurance or no third party coverage or less than 30%.
[6] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.
[7] Inter-hospital transfers were identified when hospital discharge records showed that an infant was admitted to a hospital on the same day the infant was discharged from another hospital or if a one-day difference existed between a discharge from one hospital and an admission to another hospital.
[8] Infants who died during the neonatal period ($\leq$ 28 days) were excluded from this analysis. All deaths occurred during study period, January 1, 1998, through December 31, 2008.

Table 4.2 Time to primary surgical repair of spina bifida for Florida-born infants by isolated or non-isolated SB and presence of hydrocephalus, 1998-2007

Characteristics	N	Time-to-repair (days)					p-value
		Mean	(SD)	Median	(IQR)	Range	
All infants	299	11.3	(37.0)	1.0	(3.0)	0-305	
Spina bifida [1]							
Isolated	238	12.1	(40.2)	1.0	(3.0)	0-305	0.3249
Non-isolated	61	7.9	(19.8)	1.0	(4.0)	0-129	
Hydrocephalus							
Yes	240	5.3	(21.3)	1.0	(1.5)	0-212	**<0.0001**
No	59	35.5	(66.4)	5.0	(36.0)	0-305	
Hydrocephalus and SB							
No hydrocephalus with isolated SB	47	42.0	(72.7)	5.0	(48.0)	0-305	**<0.0001**
No hydrocephalus with non-isolated SB	12	9.6	(15.2)	2.0	(11.0)	0-44	
Hydrocephalus with isolated SB	191	4.7	(21.4)	1.0	(2.0)	0-212	
Hydrocephalus with non-isolated SB	49	7.5	(20.9)	1.0	(3.0)	0-129	

N, number. SD, standard deviation. IQR, interquartile range. P-value significant at 0.05 using Wilcoxon rank sum test.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Isolated spina bifida was defined as SB with no additional coded major defects, other than the sequence of defects related to SB.

Table 4.3 Spina bifida repair for Florida-born infants by day of repair and by isolated or non-isolated SB and presence of hydrocephalus, 1998-2007 (n=299)

Characteristics	Day of SB repair						p-value
	Day 0-2		Day 3-7		After Day 7		
	n	(%)	n	(%)	n	(%)	
All infants with SB	205	(68.6)	45	(15.1)	49	(16.4)	
Spina bifida [1]							
Isolated	167	(70.2)	33	(13.9)	38	(16.0)	0.4375
Non-isolated	38	(62.3)	12	(19.7)	11	(18.0)	
Hydrocephalus							
Yes	180	(75.0)	31	(12.9	29	(12.1)	**<0.0001**
No	25	(42.4)	14	(23.7)	20	(33.9)	
Hydrocephalus and SB							
No hydrocephalus with isolated SB	18	(38.3)	12	(25.5)	17	(36.2)	**<0.0001**
No hydrocephalus with non-isolated SB	7	(58.3)	2	(16.7)	3	(25.0)	
Hydrocephalus with isolated SB	149	(78.0)	21	(11.0)	21	(11.0)	
Hydrocephalus with non-isolated SB	31	(63.3)	10	(20.4)	8	(16.3)	

N, number. P-value significant at 0.05 using chi-square test.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Isolated spina bifida was defined as SB with no additional coded major defects, other than the sequence of defects related to SB.

Table 4.4 Unadjusted and adjusted modified Poisson regression results for the association of selected characteristics with timely repair[1] of spina bifida among Florida-born infants by hydrocephalus, 1998-2007 (n=299)

Characteristics	All infants with SB (n=299)				*With* hydrocephalus (n=240)				*Without* hydrocephalus (n=59)			
	Unadjusted model		Adjusted model		Unadjusted model		Adjusted model		Unadjusted model		Adjusted model	
	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)
Exposures of interest												
Spina Bifida [2]												
Isolated	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	0.97	(0.79-1.19)	0.99	(0.80-1.21)	0.91	(0.73-1.12)	0.96	(0.77-1.19)	1.42	(0.74-2.73)	1.09	(0.53-2.23)
Hydrocephalus												
No	1.00	(1.00)	1.00	(1.00)								
Yes	**1.82**	**(1.33-2.50)**	**1.80**	**(1.31-2.48)**								
Predisposing characteristics												
Maternal age (in years)												
<25	1.06	(0.90-1.24)	0.98	(0.80-1.20)	0.91	(0.77-1.06)	0.87	(0.71-1.06)	**2.22**	**(1.27-3.89)**	**2.46**	**(1.10-5.48)**
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	0.96	(0.81-1.14)	0.99	(0.82-1.20)	1.01	(0.86.1.19)	0.95	(0.79-1.13)	0.82	(0.42-1.59)	1.95	(0.79-4.83)
Maternal race/ethnicity												
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	1.00	(0.82-1.21)	1.09	(0.87-1.36)	1.06	(0.90-1.26)	1.18	(0.96-1.46)	0.83	(0.42-1.65)	**0.34**	**(0.13-0.87)**
Non-Hispanic Black	0.95	(0.77-1.18)	0.91	(0.72-1.15)	0.97	(0.79-1.18)	1.01	(0.80-1.27)	0.53	(0.15-1.87)	**0.22**	**(0.06-0.82)**
Other	0.87	(0.42-1.79)	0.87	(0.43-1.75)	0.80	(0.39-1.64)	0.92	(0.47-1.82)				
Maternal nativity												
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	0.93	(0.76-1.13)	0.92	(0.74-1.14)	0.98	(0.81-1.17)	0.94	(0.76-1.15)	0.75	(0.34-1.66)	0.79	(0.27-2.29)
Maternal marital status												

Table 4.4 (continued)

Characteristics	All infants with SB				*With* hydrocephalus				*Without* hydrocephalus			
	Unadjusted model		Adjusted model		Unadjusted model		Adjusted model		Unadjusted model		Adjusted model	
	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)
Maternal marital status												
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	1.10	(0.94-1.29)	1.13	(0.92-1.38)	1.03	(0.89-1.20)	1.10	(0.90-1.34)	1.64	(0.90-2.98)	1.24	(0.67-2.67)
Maternal education												
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	1.00	(0.82-1.20)	0.90	(0.73-1.12)	0.94	(0.78-1.13)	0.91	(0.74-1.13)	1.09	(0.49-2.43)	0.49	(0.18-1.37)
Sex of infant												
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.94	(0.80-1.10)	0.94	(0.80-1.09)	0.99	(0.86-1.15)	1.00	(0.86-1.17)	0.51	(0.25-1.03)	0.81	(0.36-1.84)
Enabling characteristics												
Prenatal care [3]												
Adequate prenatal care	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Inadequate prenatal care	1.06	(0.88-1.28)	1.01	(0.83-1.24)	0.94	(0.78-1.14)	0.97	(0.79-1.19)	**1.92**	**(1.06-3.45)**	**3.93**	**(1.36-11.3)**
Residential rurality [4]												
Urban/urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	1.10	(0.91-1.34)	1.09	(0.89-1.32)	1.09	(0.91-1.30)	1.12	(0.93-1.35)	0.79	(0.24-2.55)	0.56	(0.17-1.91)
Payer for birth hospitalization [5]												
Public payer	1.08	(0.92-1.27)	0.98	(0.82-1.18)	0.97	(0.83-1.13)	0.97	(0.81-1.16)	1.36	(0.74-2.48)	1.47	(0.68-3.18)
Private payer	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Self or uninsured	1.25	(0.98-1.60)	**1.39**	**(1.07-1.82)**	**1.34**	**(1.24-1.45)**	**1.37**	**(1.14-1.65)**	1.51	(0.69-3.33)	**4.20**	**(1.46-12.1)**
Need characteristics												
Preterm Birth (< 37 weeks)												
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.17**	**(1.00-1.37)**	1.09	(0.92-1.28)	1.14	(0.99-1.32)	1.10	(0.94-1.28)	0.78	(0.29-2.07)	0.97	(0.32-2.94)

Table 4.4 (continued)

Characteristics	All infants with SB				*With* hydrocephalus				*Without* hydrocephalus			
	Unadjusted model		Adjusted model		Unadjusted model		Adjusted model		Unadjusted model		Adjusted model	
	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)
Nursery care in birth hospital [6]												
Level III	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Level I or II	**0.68**	**(0.50-0.91)**	0.75	(0.57-1.01)	0.74	(0.55-1.01)	0.75	(0.55-1.01)	0.74	(0.35-1.55)	0.64	(0.26-1.59)

PR=prevalence ratio, 95% CI= 95% confidence interval. Values in bold are statistically significant. Adjusted model is adjusted for all covariates.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Timely repair defined as repair of SB on day 0, 1, or 2 of life.
[2] Isolated spina bifida is SB with no additional coded major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence was identified using geocoded maternal residence and 2000 U.S. Census data.
[5] All payers were expected payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage.
[6] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.

Table 4.5 Unadjusted and adjusted modified Poisson regression results for the association of selected characteristics with a timely repair[1] of spina bifida among Florida-born infants by isolated or non-isolated SB, 1998-2007

Characteristics	Isolated SB [2] (n=238)				Non-isolated SB (n=61)			
	Unadjusted model		Adjusted model		Unadjusted model		Adjusted model	
	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)
Exposures of interest								
Hydrocephalus								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**2.01**	**(1.38-2.91)**	**1.85**	**(1.27-2.70)**	1.28	(0.72-2.27)	1.50	(0.86-2.59)
Predisposing characteristics								
Maternal age (in years)								
<25	1.09	(0.91-1.30)	1.04	(0.83-1.31)	0.93	(0.62-1.37)	0.83	(0.49-1.40)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	0.97	(0.80-1.18)	1.02	(0.83-1.26)	0.92	(0.61-1.39)	0.99	(0.66-1.47)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	1.01	(0.82-1.25)	1.07	(0.84-1.36)	0.95	(0.63-1.45)	1.77	(0.84-3.74)
Non-Hispanic Black	1.02	(0.81-1.27)	0.95	(0.74-1.21)	0.59	(0.24-1.42)	0.66	(0.33-1.33)
Other	1.10	(0.62-1.96)	1.14	(0.65-1.98)				
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	0.97	(0.78-1.20)	0.91	(0.73-1.13)	0.76	(0.44-1.31)	0.86	(0.35-2.14)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	1.06	(0.89-1.26)	1.01	(0.81-1.24)	1.30	(0.90-1.88)	**2.29**	**(1.36-3.86)**
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	1.00	(0.82-1.23)	0.91	(0.72-1.14)	0.94	(0.58-1.54)	0.91	(0.56-1.50)
Sex of infant								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.90	(0.75-1.07)	0.92	(0.78-1.08)	1.12	(0.77-1.63)	1.08	(0.74-1.57)
Enabling characteristics								
Prenatal care [3]								
Adequate prenatal care	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Inadequate prenatal care	1.16	(0.96-1.40)	1.07	(0.87-1.32)	0.76	(0.44-1.31)	0.65	(0.34-1.24)
Residential rurality [4]								
Urban/urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	1.12	(0.90-1.39)	1.11	(0.89-1.39)	1.06	(0.40-1.49)	0.99	(0.66-1.51)

Table 4.5 (continued)

Characteristics	Isolated SB [2]				Non-isolated SB			
	Unadjusted model		Adjusted model		Unadjusted model		Adjusted model	
	uPR	(95% CI)	aPR	(95% CI)	uPR	(95% CI)	aPR	(95% CI)
Payer for birth hospitalization [5]								
Public payer	1.14	(0.96-1.36)	1.05	(0.87-1.29)	0.86	(0.59-1.25)	0.65	(0.37-1.13)
Private payer	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Self or uninsured	1.22	(0.94-1.60)	**1.40**	**(1.05-1.85)**	**1.51**	**(1.25-1.84)**	**3.01**	**(1.05-8.66)**
Need characteristics								
Preterm Birth (< 37 weeks gestation)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.27**	**(1.08-1.50)**	1.17	(0.99-1.38)	0.63	(0.29-1.33)	**0.38**	**(0.15-0.97)**
Level of birth hospital nursery care [6]								
Level III	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Level I or II	**0.65**	**(0.46-0.92)**	0.76	(0.55-1.06)	0.78	(0.44-1.39)	0.77	(0.40-1.49)

PR=prevalence ratio, 95% CI= 95% confidence interval. SB=spina bifida. Values in bold are statistically significant. Adjusted model is adjusted for all covariates.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Timely repair defined as repair of SB on day 0, 1, or 2 of life.
[2] Isolated spina bifida was SB with no additional coded major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus were considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence was identified using geocoded maternal residence and 2000 U.S. Census data.
[5] All payers were expected payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage.
[6] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.

CHAPTER 5: FACTORS ASSOCIATED WITH TRAVEL TIME AND DISTANCE TO ACCESS HOSPITAL CARE AMONG CHILDREN WITH SPINA BIFIDA

5.1 Introduction

The Institute of Medicine's quality of care framework includes equity in access to care as a fundamental measure of quality health care [102]. Lack of access or inequity in access, based on inadequate health insurance coverage, poverty or on such characteristics as race/ethnicity, age, education, disability, or location of residence may result in increased morbidity and increased mortality among individuals with health care needs [102, 118, 141, 222]. In particular, children with special health care needs (CSHCN), a category that includes children with birth defects, face significant barriers to accessing health care compared to children without special health care needs [13, 128, 157, 223-227].

This research focuses on one type of major birth defect, spina bifida (SB), to examine travel time and distance to access inpatient hospital care. This research also explores predisposing, enabling, and need factors associated with travel time and distance to access hospital care. Findings from this study can help identify geographic barriers and suggest ways to improve access to hospital care for children with SB and other special needs.

5.2 Literature Review

5.2.1 Access to Health Care

The *Healthy People 2020* program overseen by the U.S. Department of Health and Human Services included access to care in their series of Maternal-Infant-Child Health (MICH) goals [31]. MICH Goal 30.2 states that *Healthy People 2020* programs will "increase the proportion of children with special health care needs who have access to a medical home" [31]. MICH Goal 31 states that another objective of *Healthy People 2020* is to "increase the proportion of children with special health care needs who receive their care in family-centered, comprehensive, coordinated systems" [31]. Similarly, the Spina Bifida Association's 2012 Congressional Policy Agenda calls for ensured access to care for individuals with SB, especially through provision of adequate insurance [32].

Previous literature has described five interdependent dimensions of access to care [119-121]. Those dimensions include availability, accessibility, accommodation, affordability, and acceptability [119-121]. Availability is associated with the adequacy of health care personnel, facilities, and special services [120, 121]. Accommodation describes the relationship the health care providers' organizational plans to accept clients, and clients' perceptions of the plans' suitability and appropriateness [120, 121]. Affordability addresses health care costs, clients' abilities to pay those costs, as well as clients' perceptions of the value of the costs [120, 121]. Acceptability is the relationship between attitudes of clients and providers about personal and practical characteristics that influence both seeking and providing care [120, 121]. The final dimension of access to care is accessibility. Accessibility describes the relation between the location of the health care service or provider and the potential health care client, and examines

measures such as transportation resources, travel time and distance, and travel costs [120, 121]. The dimension of accessibility is the focus of this research.

A number of studies have explored barriers to accessing health care for children [122-128]. Barriers to care can fall into the categories of personal barriers, financial barriers, or organizational (structural) barriers [129], generally paralleling the dimensions of access to care [121]. Personal barriers result from individual perceptions of health care need or personal health beliefs. Personal barriers may also include cultural and social influences, such as language barriers and expectations of care [127, 129]. Financial barriers occur when an individual has insufficient monetary resources or health insurance coverage to access care adequate health care [108, 127, 129-131]. Organizational or structural barriers are factors related to the health care system and include such characteristics as capacity, transportation, and geographic location of resources [129, 132]. Factors in each of these three categories (personal, financial, or organizational and structural) can influence the ability of an individual to access health care. The inability to access health care can result in missed or delayed opportunities for health services and can ultimately result in poorer health outcomes with higher health care costs [127].

Personal and financial barriers are commonly reported barriers to accessing care for children with or without special health care needs and include low income, minority status, and lack of insurance [125, 130, 133, 134]. A 2001 study of Latino children found that children of immigrant parents were less likely to have insurance and less likely to access routine health care than Latino children of US-born parents [135]. Studies have found that CSHCN are particularly likely to face barriers with accessing health care [122,

124-128, 130]. Among CSHCN, adequate insurance has been reported to be the most critical determinant of access to care [127].

Less research exists on organizational or structural barriers to accessing health care, and specifically for children with birth defects, a subset of CSHCN. Using the 2001 *National Household Travel Survey*, researchers found that the average distance traveled to access medical or dental care in the United States was 10.2 road miles or 22.0 minutes [136]. This study also found that rural residents traveled 31.4% longer time to access care than residents of urban areas [136]. Specifically examining access to care for CSHCN, researchers used 2000-2002 data from the *National Survey of Children with Special Health Care Needs* and found that CSHCN who lived in rural areas were less likely to be seen by a pediatrician than children living in urban areas [128]. Another study using data from the 2005-2006 *National Survey of Children with Special Health Care Needs* found that geographic disparities existed for CSHCN in the western and northeastern regions of the United States [137].

While a number of studies have used survey data to examine access to care for CSHCN and specifically for children with birth defects, a subset of CSHCN [123, 128, 137], fewer studies have used GIS methods to examine access to care. Using a statewide, population-based birth defects registry data and geographic information system (GIS) methods, researchers found geographic disparities in access to pediatric genetic clinics among children born with major structural or chromosomal anomalies in Texas between 1999 and 2004 [138]. Using a statewide, population-based birth defects registry and survey data in North Carolina, a qualitative study of perceived barriers to care among mothers of children with orofacial clefts born between 2001 and 2004 found multiple

perceived barriers to accessing care, including location of services and lack of transportation [124]. Using data from the same North Carolina survey, researchers found that approximately 48% of mothers who responded traveled more than one hour to access care for their children with clefts [123]. In another study, researchers used a statewide, population-based study using Texas birth defects registry data and GIS methods to examine mortality among infants with congenital heart disease born in Texas between 1996 and 2003. These researchers found no association between increased mortality rates and home distance to a cardiac center [139]. Using linked Florida birth defects registry and hospital discharge data from 1998-2007, researchers calculated one-way travel time and distance to access hospital care for infants with spina bifida [140]. Researchers found that 56.4% of infants traveled less than 30 minutes to access hospital care, while 22.4% traveled more than 60 minutes to access hospital care [140]. Collectively, these studies contribute to our understanding of the role that structural barriers play in accessing health care, especially for CSHCN, including children with birth defects. The findings of these studies suggest that geographic location is associated with the use of health care services. These studies also suggest that CSHCN may travel longer times and distances than individuals without these conditions in the general population to access health care.

Notwithstanding previous research, the role of geography and health remains an important and under-researched component to understanding heath [141]. Few studies have examined the role of geography and access to health care among children with birth defects. Siffel et al. examined the role of GIS methods in birth defects surveillance [143]. These researchers indicated that “place” is the least researched of the three

epidemiological components (person, place, and time) [142] because of the challenges inherent in geographic research methods. These challenges include standardizing and defining spatial features and maintaining individual confidentiality [143]. Siffel et al. (2006), however, recommended an expanded and wide-ranging use of GIS in collaborative birth defects research to better understand the role of place in birth defect interventions [143]. Similarly, Kirby (2006) noted that the evaluation of the spatial component of disease occurrence, specifically intellectual and developmental disabilities, could address previously unanswered issues related to spatial distribution of incidence, prevalence, or of distribution of appropriate health care providers or health services use [144].

One recent study used GIS-methods to examine the geographic distribution of low birth weight prevalence at both the county and census tract level for all singleton infants live-born in 2000 in the state of Georgia [228]. A similar study used GIS-methods to investigate trends and spatial clusters of low birth weight rates over 11 years at the county level in the state of Georgia [229]. To date, researchers have used GIS methods to examine risk factors or geographic distribution of birth defects [145-148, 150, 228-230]; however, limited research has used GIS methods to examine access to care for children with birth defects [138-140].

5.2.2 Description of Spina Bifida

Spina bifida (SB), one type of major birth defect, is a neural tube defect that results from a failure of the caudal neural tube to fuse early in embryonic development. SB has been called one of the most severe birth defects that remains compatible with life [231]. The severity of impairment caused by SB is directly related to the size and

position of the defect along the spinal column, which thus affects a child's mobility and ability to maintain bowel and bladder control [44-46, 82, 97]. A child with SB is also at risk for comorbidities associated with SB, such as hydrocephalus, neurogenic bladder and decreased renal function, orthopedic problems including scoliosis and lower limb issues, and obesity [46, 62]. A child with SB may also face challenges with educational, social, and psychological development [47] and typically requires life-long, multidisciplinary health care. However, CSHCN (a category that includes children with SB) face substantial barriers to accessing heath care services compared with children with no special health care needs [13, 128, 157, 223-227].

5.2.3 Spina Bifida: Isolated and Non-isolated

Spina bifida may present as a single condition in a newborn or may be accompanied by other conditions diagnosed at birth or later in life. Isolated SB is SB with the single SB malformation or SB with sequential or associated malformations, such as hydrocephalus, hip dislocation, or defects of the urinary system [12, 79, 80]. Isolated SB may also include SB with other minor anomalies, such as low set ears, skin tags, or abnormally bent or curved fingers (clinodactyly) [12, 80]. Non-isolated SB is defined as SB with another major, unrelated birth defect and without a syndromic diagnosis [12, 79, 80]. Children with non-isolated SB most commonly have orofacial clefts, cardiac defects, and renal or abdominal wall anomalies [81, 82]. Approximately 15% to 25% of cases of SB are non-isolated and occur with another anomaly not related to the central nervous system or its associated defects [81, 82, 160-162].

5.2.4 Spina Bifida with Hydrocephalus

One of the most common comorbidities associated with SB is hydrocephalus [56, 66]. Hydrocephalus is an abnormal accumulation of cerebrospinal fluid in the ventricles of the brain that may cause swelling and increased intracranial pressure [37, 49, 50, 56, 66, 85]. This increased pressure can cause multiple central nervous system-related symptoms and may result in seizures, brain damage, and if untreated, death [56, 84]. Even when treated, hydrocephalus may be associated with chronic conditions, including cognitive and developmental disabilities [47, 85-87]. Hydrocephalus is diagnosed in approximately 80 to 90% of children with SB whose defect type is a meningomyelocele [43, 66] and is the leading cause of death among children with SB [21]. Among children with spina bifida with hydrocephalus, approximately 15% undergo immediate ventriculoperitoneal (VP) shunt placement in conjunction with the repair of their SB defect, and as many as 80-90% eventually undergo surgical placement of the VP shunt [56].

In general, individuals who have multiple comorbidities have poorer health outcomes and higher health care costs than those without comorbidities [17]. Health resource use by individuals with multiple conditions may appear different when the conditions are examined separately as compared with examining the conditions together [17]. Thus, a concurrent examination of comorbidities is important to understanding health resource use among individuals with more than one health condition.

5.3 Study Objectives and Hypotheses

The purpose of this study is to examine the effect of hydrocephalus and other selected predisposing, enabling, and need characteristics on travel time and distance to access hospital care among children with SB. The research questions are:

1. What are the differences in average time and distance traveled to access hospital care for children with SB by the presence of hydrocephalus, payer type, and age group?

2. What predisposing, enabling, and need characteristics are associated with average time and distance traveled to access hospital care for children with SB by the presence of hydrocephalus?

Based on previous research, I hypothesize that differences in average time and distance travelled to access hospital care will vary based on predisposing characteristics (e.g., maternal race/ethnicity and educational level), enabling characteristics (e.g., health payer and rural residence), and need factors (e.g., premature birth and presence of hydrocephalus) [125-128, 132, 133, 135, 137, 157].

5.4 Conceptual Framework

The Aday and Andersen *Framework for the Study of Access to Medical Care* provided the conceptual basis for this project [152, 156, 175, 176]. The Aday and Andersen *Framework for the Study of Access to Medical Care* is shown in Chapter 2 (Figure 2.2, page 36). I adapted the Aday and Andersen model to the research questions presented above and included the specific predisposing, enabling, and need variables used in each component of the model (this model is shown in Chapter 2, Figure 2.3, page 37).

5.5 Study Design and Methods

5.5.1 Study Design

This study was a retrospective, statewide, population-based cohort analysis of inpatient hospital use for children with SB ages birth to four years born in Florida between January 1, 1998, and December 31, 2007.

5.5.2 Data Acquisition and Study Sample

Data for this study were obtained from linked, longitudinal datasets provided by the Florida Birth Defects Registry (FBDR) within the Florida Department of Health FDOH), Florida Bureau of Vital Statistics, also within the FDOH, and the Florida Agency for Healthcare Administration (AHCA), which provided the hospital discharge data. Infants with SB without anencephaly born in Florida between 1998 and 2007 were identified using the *International Classification of Disease, 9th revision; Clinical Modification* (ICD-9-CM) codes 741.00-741.93. Hospital discharge data from January 1, 1998, through December 31, 2008, were used to allow for at least one year of data for hospitalizations for each infant.

Included infants were live-born in Florida to a mother who was a Florida resident at the time of delivery and who matched with an inpatient hospital discharge record during the first year of life. Infants who were adopted or prospectively adopted or who were born out of state were excluded by the FBDR. Figure 1.1 (Chapter 1, page 9) shows the process for identifying infants and children for the final study sample.

5.5.3 Primary Outcomes of Interest: Travel Time and Distance

The FDOH first geocoded the maternal residential address at the time of the infant's birth, using information from the birth certificate and hospital addresses from the

AHCA data. The FDOH successfully geocoded 90.7% of maternal addresses [140]. The University of North Carolina at Charlotte (UNC Charlotte) Department of Geography and Earth Sciences further geocoded addresses at the street and ZIP-code level to successfully geocode a total of 99.7% of maternal residential addresses [140]. The UNC Charlotte Department of Geography and Earth Sciences also geocoded at the street level all Florida hospitals where children with SB were hospitalized, 1998 through 2008 [140].

The Geography and Earth Sciences Department at UNC Charlotte used GIS methods to calculate the primary outcomes of interest, travel time and distance, to access hospital care. Using 2007 data from the Florida Department of Transportation, colleagues in the UNC Charlotte Department of Geography and Earth Sciences created a road network that incorporated different road types, including interstate highways, state, county and local roads. Topological modeling incorporated turn restrictions to enhance the validity of travel time and distance measures. Travel time for each road segment was computed as the length of that road segment divided by the maximum allowable driving speed on that segment of road in 2007.

The one-way shortest and fastest route from maternal residential address at infant's birth (origin) to the hospital where care was received (destination) was estimated using the Dijkstra algorithm [140, 232-234]. The Dijkstra algorithm is included in the software package of ArcGIS (ESRI, Redlands, CA). ArcGIS is a commercial software system used for GIS analysis.

Hospitalizations were defined as single admissions that involved no inter-hospital transfers. Some researchers merge data from two hospitalizations that are linked by an inter-hospital transfer. The combined admissions are then reported as a single hospital

experience [177]. However, I did not merge hospital data based on transfers. I examined hospitalizations separately to capture information each episode of travel, and thus reduced error and more accurately measured the travel time and distance associated with accessing care for each hospitalization.

I measured one-way travel time and distances in minutes and miles, respectively. Mean, medians, ranges, and total travel time and distance were reported for children during infancy and for ages one to four years old and by the presence of hydrocephalus and payer type for infancy.

Based on my examination of the data and informed by categories used by previous relevant studies of time and distance for birth defects or other medical conditions [123, 138, 140, 148, 235-237], and to maintain adequate cell sizes for meaningful results and to be able to directly compare with previous research, I collapsed categories for time and distance. I reported one-way travel time in four categories: 0-30 minutes, 31-60 minutes, 61 to 90 minutes, and more than 90 minutes. Similarly, I reported one-way travel distance in categories of 0-30 miles, 31-60 miles, 61 to 90 miles, and more than 90 miles. For the multivariable analysis, I dichotomized travel time and distance into categories of greater than 30 minutes and less than 30 minutes (and greater or less than 30 miles) per one-way trip. I based this decision on the sample size, the observed median travel time and distance for these data, previous literature, and in consultation with GIS and birth defects experts.

5.5.4 Exposures of Interest: Hydrocephalus and Isolated or Non-Isolated Spina Bifida

Hydrocephalus, a need characteristic, was the primary exposure of interest. I identified hydrocephalus by the ICD-9-CM codes 741.01- 741.03 from the FBDR dataset.

The selection of these ICD-9-CM codes was informed by discussions with several expert clinicians from the Centers for Disease Control and Prevention's (CDC) National Center on Birth Defects and Developmental Disabilities (NCBDDD). I reported the presence of hydrocephalus as a dichotomous variable.

The variable isolated or non-isolated SB, also a need characteristic, was another exposure of interest. Spina bifida was reported for each infant as a dichotomous variable, isolated or non-isolated SB. Infants were classified as having isolated *SB* if they met any of the following criteria: 1) only the SB birth defect; or 2) had only the SB defect and another minor birth defect associated or not associated with the SB, such as low set ears or skin tags; or 3) had only the SB defect accompanied by a documented sequence of related defects and no additional unrelated major defects [79, 80], as verified by a clinical expert at the CDC's NCBDDD. Classification of having isolated or non-isolated SB was informed by discussions with expert clinicians from the CDC's NCBDDD and by previous research [12, 79]. The classification of isolated or non-isolated SB referenced ICD-9-CM codes for major birth defects listed in the surveillance guidelines by the National Birth Defects Prevention Network (NBDPN) [83]. Expert clinical consultants from the CDC's NCBDDD manually reviewed approximately 15% of the study sample that required additional consideration because of multiple conditions. For example, patent ductus arteriosus (PDA) is heart condition common among premature infants. If an infant with SB had a PDA, the infant was considered to have isolated SB if the infant was premature, but non-isolated SB if the infant was born at term. These and other similar situations required a case-by-case review. I referenced surveillance guidelines from the National Birth Defects Prevention Network (NBDPN) for ICD-9-CM diagnostic

codes for major birth defects [83]. Table A in the appendix lists ICD-9-CM diagnostic codes considered as major birth defects by the NBDPN for its surveillance and research purposes.

5.5.5 Covariables Measuring Predisposing, Enabling, and Need Characteristics

I categorized additional covariables as predisposing, enabling, and need characteristics. These characteristics corresponded to the components of the Aday and Andersen conceptual model. Consistent with the Aday and Andersen conceptual model, the following characteristics were considered predisposing characteristics: maternal age, maternal race/ethnicity, maternal nativity, parity, marital status, maternal education, and child's sex. Enabling characteristics included the variables that measured adequacy of prenatal care, residential rurality, and health insurance payer. In addition to the primary exposures of interest, hydrocephalus and isolated or non-isolated SB, need characteristics also included preterm birth and level of nursery care at the birth hospital. I describe these variables and their coding in detail in the paragraphs that follow.

5.5.5.1 Predisposing Characteristics of Mothers and Children

Predisposing characteristics of mothers included maternal race/ethnicity, maternal age and education, maternal nativity, and marital status. This information was obtained from the FBDR and Florida statistics data. Maternal parity was calculated by adding the number of live born children still living and those live born now deceased as reported in the FBDR data.

Predisposing characteristics of the child were sex and age. The sex of the child was obtained from the FBDR data. The child's age was calculated in years using the

time to admission variable in the Florida AHCA (hospital discharge) data, which was reported in days.

5.5.5.2 Enabling Characteristics of Mothers and Children

Enabling characteristics included adequacy of prenatal care, obtained from Florida vital statistics data, rurality of maternal residential address, also obtained from Florida vital statistics data and using information from the U.S. Census Bureau, and health insurance payer, which was obtained from the Florida AHCA (hospital discharge) data. Adequacy of prenatal care was determined using the Kotelchuck Index. The Kotelchuck Index creates a ratio comparing the month in which prenatal care was initiated with the total number of prenatal visits prior to delivery to calculate four categories of prenatal care: inadequate (less than 50% of expected visits), intermediate (50-79%), adequate (80-109%), and adequate plus (110% or more) [181, 182]. The Kotelchuck scoring system considers scores less than 80% to be inadequate care [181, 182]. For the purpose of this research, based on an examination of the data, and to ensure adequate cell sizes for meaningful results, I reported adequacy of prenatal care as a dichotomous variable. I used the Kotelchuck cut point of 80% to classify adequate and adequate plus care as "adequate prenatal care", and intermediate and inadequate care as "inadequate prenatal care."

I identified maternal residential rurality by comparing the geocoded maternal residential addresses reported at birth with the 2000 U.S. Census data that reported rurality by block group level. All maternal residential addresses that had been geocoded could be identified as urban, urban clusters, or rural [140]. In the 2000 U.S. Census, the U.S. Census Bureau defined "urban" as all territory, population, and housing units

located within an urbanized area or in an urban cluster [183]. "Urban areas" and "urban clusters" were described by the U.S. Census Bureau as densely settled areas consisting of core census block groups or blocks that had a population density of at least 1,000 people per square mile and surrounding census blocks that had an overall density of at least 500 people per square mile [183]. The U.S. Census Bureau defined all territory, population, and housing units located outside of urban areas or clusters as "rural" [183]. The U.S. Census Bureau assigned a designation to each census block group identifying the geographic area as urban, an urban cluster area, or as rural. Following consultation with spatial research experts at the UNC Charlotte Department of Geography and Earth Sciences and upon examination of the data, I collapsed urban and urban cluster designations into a single "urban" category. I then reported maternal residential "rurality" as a dichotomous variable, "urban" or "rural", to ensure adequate cell sizes for meaningful results.

Health insurance payers were classified in two ways using hospital discharge data from the Florida AHCA. First, payers for the birth hospitalization were classified as: 1) public, 2) private, or 3) self-insured, under-insured, or no insurance. Second, payer type was reported across infancy and across ages one to four years. Payer type was classified as: 1) public payers only for all hospitalizations, 2) private payers only for all hospitalizations, 3) self-insured, under-insured, or no insurance only for all hospitalizations, or 4) multiple payer type. Multiple payer type indicated that a child had different types of health insurance coverage types across more than one hospitalization; for example, one hospitalization was covered by a private health insurance payer and a subsequent hospitalization was covered by a public payer. Separate payer types were

reported for hospitalizations during infancy and for hospitalizations during ages one to four years. Public payer sources included Medicare, Medicaid, KidCare (Florida's state children's health insurance program), and Veteran's Administration insurance. Private payer sources included private or employer-based insurance, including military coverage (CHAMPUS or TriCare). The self-pay, no insurance or under-insured category was defined by the Florida AHCA as either no third party coverage or less than 30% estimated insurance coverage [184].

5.5.5.3 Need Characteristics of Mothers and Children

In addition to the exposures of interest, hydrocephalus and isolated or non-isolated SB, the need characteristics also included preterm birth (less than 37 weeks gestation), low birth weight (less than or equal to 2500 grams), and plurality, which were all obtained from Florida vital statistics data.

I also considered the level of nursery care at the birth hospital as a *need* characteristic. The American Academy of Pediatrics classifies level of nursery care at the birth hospital as Level I, II, or III [185, 186]. Level III nursery care provides the most sophisticated care for complex cases [185, 186]. I reported the level of hospital nursery care for the birth hospitalization, even if an infant was transferred at birth to a hospital with a higher level of nursery care. I defined a birth hospitalization as a first hospitalization with age at admission of zero days or a first hospitalization with an age at admission of one day with an accompanying indication of hospital transfer [177]. I used the level of nursery care only in analyses that examined birth hospitalizations.

Finally, I reported the child's death, which was categorized as no death as of December 31, 2008, death during infancy (birth through one year), or death between ages one to four years.

5.5.6 Statistical Analyses

I conducted descriptive analyses for the predisposing, enabling, and need characteristics of the study population and health system. I reported the means, median, and ranges in minutes and miles for one-way travel time and distance. I reported the means, medians, and ranges separately for birth hospitalizations, post-birth hospitalizations during infancy, all infancy hospitalizations, and hospitalizations during ages one to four years. I collapsed data from children ages one to four years into a single category to ensure adequate sample sizes for meaningful results.

Bivariate analyses examined one-way travel time and distance of more or less than or equal to 30 minutes or 30 miles, respectively, by selected predisposing, enabling, and need characteristics. Among infants, one-way travel time and distance for birth and post-birth hospitalizations were reported separately from all infancy hospitalizations, because experiences of travel to birth hospitalizations maybe unique from subsequent hospitalizations. Chi-square analyses were conducted on the categorical variables to determine significance level using a p-value of <0.05. Where appropriate, I used Fisher's exact test to account for small cell sizes, using a p-value of <0.05 to determine statistical significance. Because of the skewness of the continuous measures of travel time and distance, I conducted Wilcoxon Rank Sums tests to determine significance level using a p-value of <0.05.

For the multivariable analyses, logistic regression models were used to calculate unadjusted odds ratios (uOR), adjusted odds ratios (aOR), and corresponding 95% confidence intervals (CI) to determine if the selected *predisposing*, *enabling*, and *need* factors were associated with one-way travel time and distance of more or less than or equal to 30 minutes or 30 miles, respectively, to access hospital care. I used logistic regression because of the dichotomous nature of the outcomes: travel time of less than or equal to 30 minutes (reference) vs. greater than 30 minutes, and travel distance of less than or equal to 30 miles (reference) vs. greater than 30 miles.

The goal of the multivariable analyses was to arrive at models that were theory-based, informed by previous literature, and parsimonious given the relatively small sample size; thus, selected *predisposing*, *enabling*, and *need* covariables were included in the final regression model. Low birth weight was excluded because of its close correlation with preterm birth. Plurality was excluded because too few infants were part of multiple births to contribute meaningfully to the results. The following variables were included in the final regression models: hydrocephalus, isolated or non-isolated SB, maternal age, maternal race/ethnicity, and maternal nativity, marital status, parity and education, adequacy of prenatal care, residential rurality, health care payer, child's sex, preterm birth, and child's death. Logistic regression models were created for each of the categories of theory-based factors (predisposing, enabling, and need) and with all variables included except for low birth weight and plurality as previously mentioned. Analysis was conducted both including and excluding the children who died during the study period (prior to December 31, 2008). I included all children to capture as much information as possible on one-way travel time and distance, but also conducted an

analysis excluding children who died to reduce error in measurement of time and distance. For models that included children who died, death was included as a dichotomous covariable.

Individual variables used in the multivariable analysis were assessed for multicollinearity [189, 190]. I conducted sensitivity tests to examine differences in children with and without hydrocephalus and among infants who had or did not have a *birth* hospitalization. I also used the likelihood ratio test to examine for differences in model fit with and without the variable hydrocephalus.

All analyses were conducted using SAS 9.2 statistical software (SAS Institute, Inc., Cary, NC). This study was approved by the Institutional Review Boards at UNC Charlotte, the FDOH, and the CDC's NCBDDD.

5.6 Results

I conducted several initial statistical tests to examine the data. There was no evidence of notable multicollinearity based on recommended maximum levels of the variance inflation factor [189, 190]. Results of the likelihood ratio test comparing the full model for infancy with and without the variable for hydrocephalus showed no significant difference in the models ($p=0.0771$). However, my final model was informed by theory and previous research. Thus, the final model included the variable for hydrocephalus.

Among the covariables, some covariables were missing no observations; no covariable was missing more than 10% of its observations. The covariable with the largest number of missing values was "adequacy of prenatal care", an enabling characteristic, which had 5.6% missing observations.

5.6.1 Descriptive Results

5.6.1.1 Selection of Study Sample

The FBDR data identified 914 Florida-resident infants who were born between January 1, 1998 and December 31, 2007, with an ICD-9-CM code indicating a neural tube defect. Of these 914 infants, 668 had ICD-9-CM codes for SB without anencephaly. Of the 668 infants with SB, 614 linked to at least one hospital discharge record. Infants who did not have a linked hospital discharge record and were in the FBDR were more likely to be born to younger mothers of Hispanic origin and foreign-born and were more likely to have lower educational levels than the infants who matched with a hospital discharge record (data not shown). No significant differences were found for maternal age, marital status, infant's sex and birth weight between the infants who matched and did match to hospital discharge records (data not shown).

From the time of the infant's delivery, 612 infants had a maternal residential address that could be geocoded. These infants comprised the sample for analysis. Figure 1.1 (Chapter 1, page 9) shows the process for selecting the study sample. Figures 5.1 and 5.2 are Florida maps that show the distribution of the geomasked maternal residential addresses of the infants at the time of delivery. Geomasking is a GIS method that alters a geocoded location in such a way that maintains individuality confidentiality, while preserving the relationship between geocoded locations [238, 239]. The maps show a greater number of children in urban areas, and fewer children in the rural panhandle section of northwest Florida and in the south central Everglades area, as expected. Figure 5.3 shows the locations of the hospitals used by infants and children with SB during the study period.

One-way travel time and distance to access a birth hospital were reported separately for the 569 infants who had a birth hospitalization. Infants without a birth hospitalization reported in the AHCA data were more likely to be born to younger mothers who were rural residents and not born in the United States (data not shown). No significant differences were found in maternal race/ethnicity, maternal education, marital status, and infant's sex and birth weight between children with and without a *birth* hospitalization (results not shown).

5.6.1.2 Descriptive Characteristics of the Mothers and Children

Table 5.1 shows the descriptive characteristics of the mothers and infants in this study (n=612). About 53% (n=321) of infants were born to non-Hispanic White mothers. About 60% (n=366) of mothers were married. Most mothers were native-born in the United States (75.8%, n=464), had at least a high school diploma (76.1%, n=466), and had received "adequate" prenatal care (72.7%, n=445). Approximately 86% (n=525) of mothers lived in urban or urban cluster areas, and 14.2% (n=87) of mothers lived in rural areas. About 20% (n=120) of infants were born low birth weight and 26.5% (n=162) were born preterm. About 26% of infants (n=156) had non-isolated SB. Approximately 57% (n=347) of infants had hydrocephalus.

Less than 10% of the study population (8.5%, n=52) died at any point during the study period. Among the 52 infants who died, 41 (78.8%) (6.7% of the 612) died during infancy and an additional 11 (21.1%) (1.8% of the 612) died between the ages of one and four years.

5.6.1.3 Descriptive Results for One-Way Travel Time

Table 5.2 shows descriptive one-way travel time results for birth, post-birth, and all infancy hospitalizations, and for hospitalizations during ages one to four years. Infants had an average one-way travel time of 41.9 minutes and median of 20.3 minutes (standard deviation, SD: 64.9 minutes; and range: 1-571 minutes) to access care at a *birth* hospital. Infants had an average one-way travel time of 50.5 minutes and median of 28.0 minutes (SD: 66.5 minutes; range: 2.4-732 minutes) for all post-birth hospitalizations during infancy .The average one-way travel time for all infancy hospitalizations was 45.1 minutes with a median of 25.9 minutes (SD: 54.6 minutes; range: 2.4-494 minutes).

Table 5.3 shows findings for one-way travel time and distance by child's age. During the first year of life, 56.3% (n=345) of infants had a one-way travel time of 30 minutes or less to access hospital care, while 43.6% (n=267) of infants had a one-way travel time of more than 30 minutes. Approximately 22% (n=136) of infants traveled longer than 60 minutes to access hospital care.

For children with SB ages one to four years (n=251 children), average one-way travel time was 39.8 minutes and the median was 22.7 minutes (SD: 46.0 minutes and range: 2.4-285.6 minutes). Approximately 61% (n=154) of children traveled 30 minutes or less to access hospital care, while 38.6% (n=97) traveled more than 30 minutes to access care. Approximately 21% (n=53) of children ages one to four traveled longer than 60 minutes.

Figures 5.4 and 5.5 are Florida maps indicating average one-way travel patterns for hospitalizations during infancy and ages one to four years, respectively. These two maps suggest that some children traveled significant distances to receive hospital care.

The maps also suggest that some children were not hospitalized at the hospital closest to their maternal residence at birth.

5.6.1.4 Descriptive Results for One-Way Travel Distance

Table 5.2 also shows one-way travel distance for birth, post-birth, and all infancy hospitalizations, and for hospitalizations during ages one to four years. Infants traveled one-way an average distance of 32.2 miles and a median distance of median 13.0 miles (SD: 53.8 miles; range: 0.6-433 miles) to access care at a birth hospital. Infants traveled one-way an average distance of 38.5 miles and a mean distance of 19.7 miles (SD: 54.3 miles; range: 1.3-598 miles) for all post-birth hospitalizations during infancy. The one-way travel for all infancy hospitalizations was an average distance of 34.5 miles and a median distance of 18.1 miles (SD: 45.4 miles; range: 1.2-404 miles). During ages one to four years, average one-way travel distance was 38.2 miles and median one-way travel distance was 19.4 miles (SD: 50.1 miles; range: 1.3-325 miles). Approximately 64% of children ages one to four (n=161) traveled 30 miles or less to access hospital care, while 35.9% traveled more than 30 miles. About 20% (n=51) of children ages one to four traveled more than 60 miles to access hospital care.

5.6.2 Bivariate Results

5.6.2.1 Bivariate Results for Demographic Characteristics

Table 5.4a presents the results of bivariate analysis comparing infants who traveled more than 30 minutes to access hospital care with those who traveled less than or equal to 30 minutes. Among infants who traveled less than or equal to 30 minutes, 41.2% (n=142) were born to mothers who were non-Hispanic White; 30.7% (n=106) were Hispanic; and 26.1% (n=90) were non-Hispanic Black. Infants born to mothers in

racial/ethnic minority groups traveled significantly shorter times than infants born to non-Hispanic White mothers ($p<0.0001$). Compared with infants who traveled 30 minutes or less to access hospital care, infants who traveled more than 30 minutes were more likely to have mothers who were married ($p=0.007$), born in the United States ($p=0.0002$), and were rural residents ($p<0.0001$). Finally, infants who traveled more than 30 minutes were more likely to have hydrocephalus ($p=0.038$) or to have non-isolated SB ($p=0.003$).

Table 5.4b shows the results of bivariate analysis comparing children ages one to four years who traveled 30 minutes or less to access hospital care with those who traveled more than 30 minutes (Table 5.4b). Among children who traveled 30 minutes or less, 40.3% (n=62) were born to mothers who were non-Hispanic White; 29.2% (n=45) were Hispanic; and 26.6% (n=41) were non-Hispanic Black. Mothers in racial/ethnic minority groups traveled significantly shorter times to access care for their children than non-Hispanic White mothers ($p=0.0149$). These was modest evidence that children who traveled more than 30 minutes were more likely to have mothers who lived in rural areas ($p=0.0604$). No other characteristics were statistically significant.

5.6.2.2 Bivariate Results by Hydrocephalus and Isolated or Non-Isolated Spina Bifida

Table 5.2 further shows the one-way travel time and distance results by the presence of comorbidities and by isolated or non-isolated SB for birth, post-birth, and all infancy hospitalizations, as well as hospitalizations during ages one to four years. Infants with hydrocephalus or with non-isolated SB or with both hydrocephalus and non-isolated SB all experienced significantly longer average one-way travel times and distances compared to infants with isolated SB without hydrocephalus. One-way mean and median travel times for infants without hydrocephalus were statistically different compared to

infants with hydrocephalus. Infants without hydrocephalus traveled 37.3 (mean) and 24.1 (median) minutes one-way to access hospital care, while infants with hydrocephalus traveled 51.0 (mean) and 27.4 (median) minutes to access hospital care (p=0.009). Similarly, mean and median travel distances for infants without hydrocephalus were 28.0 and 16.3 miles, respectively, while infants with hydrocephalus had 39.4 (mean) and 20.8 (median) miles (p=0.004).

Observing infancy hospitalizations by birth and post-birth admissions, the presence of hydrocephalus, non-isolated SB, and non-isolated SB with hydrocephalus were all significantly associated with longer one-way travel times and distances to birth hospitalizations (Table 5.2). Among post-birth hospitalizations, non-isolated SB was associated with both longer one-way travel time and distance (time: p=0.0170; distance p=0.0135) and non-isolated SB with hydrocephalus was associated with longer one-way travel distance (p=0.0337). The presence of hydrocephalus was not significantly associated with one-way travel time or distance for post-birth hospitalizations (Table 5.2)

One way mean and median travel times for infants with isolated SB was 42.3 and 24.2 minutes, respectively, while infants with non-isolated SB experienced travel times of 53.2 (mean) and 31.8 (median) minutes (p=0.001). Similarly, one-way mean and median travel distances for infants with isolated SB were 32.1 and 16.6 miles, respectively, while infants with non-isolated SB traveled 41.3 (mean) and 24.6 (median) miles (p=0.001). These findings suggest that infants with more complex presentations of SB that include another major birth defect (non-isolated SB) travel more time and distance to access hospital care.

Notably, infants who had both hydrocephalus and non-isolated SB traveled the longest time and distance to access hospital care: one-way mean of 60.8 minutes and median of 34.2 minutes (SD: 72.4; range: 5-494 minutes) and one-way mean of 48.5 miles and median of 26.9 miles (SD: 61.1; range: 3-404 miles). Both one-way travel time and distance were significantly different from infants who did not have either of these conditions. The average one-way travel time and distance to access hospital care for children ages one to four years did not differ based on the presence of hydrocephalus or isolated or non-isolated SB

5.6.2.3 Stratified by Health Care Payer

Table 5.4c shows results for one-way travel time and distance when stratified by payer for birth hospitalization and by for payer types across all infancy admissions. There were no statistically significant differences in one-way average travel time and distance across payers and payer types.

5.6.4 Multivariable Results

5.6.3.1 Multivariable Results for Primary Exposures of Interest (Presence of Hydrocephalus and Isolated or Non-Isolated SB)

Table 5.5a shows the unadjusted and adjusted OR and 95% CI for the exposures of interest and the average one-way travel time to access birth and post-birth hospitalizations. In the adjusted models, the presence of hydrocephalus was not significantly associated with one-way travel time to either birth or post-birth hospitalizations. The presence of non-isolated SB, however, was associated with longer one-way travel times for both types of hospitalizations (birth: aOR=0.50, 95% CI: 0.32-0.79 and post-birth: aOR=0.61, 95% CI: 0.38-0.96).

Table 5.5b shows the unadjusted and adjusted OR and 95% CI for the exposures of interest and the average one-way travel time to access all infancy hospitalizations. In the adjusted models, the primary exposure of interest, hydrocephalus, was not significantly associated with one-way travel time across all infancy admissions (aOR=0.78, 95% CI: 0.54-1.13). However, non-isolated SB was associated with decreased odds of traveling 30 minutes or less compared to infants with isolated SB (aOR=0.58. 95% CI: 0.38-0.89).

Table 5.5c shows unadjusted and adjusted OR and 95% CI for the exposures of interest and the average one-way travel time to access hospitalizations for children ages one to four years. In the unadjusted and adjusted models, neither hydrocephalus nor non-isolated SB was significantly associated with one-way travel time to access hospital care during ages one to four years (hydrocephalus: aOR=0.77, 95% CI: 0.39-1.51; non-isolated SB: aOR=0.86, 95% CI: 0.46-1.60).

5.6.3.2 Multivariable Results for Predisposing Characteristics

Maternal minority race/ethnicity was associated with about two times shorter one-way travel times for both birth and post-birth hospitalizations during infancy (Hispanic: aOR=2.31, 95% CI: 1.29-4.15 birth and aOR=2.10, 95% CI: 1.13-3.93 post-birth; non-Hispanic Black: aOR=2.33, 95% CI: 1.38-3.93 birth and aOR=1.82, 95% CI: 1.02-3.24 post-birth). For all infancy hospitalizations, infants born to mothers of maternal racial/ethnic minority groups consistently had increased odds of one-way shorter drive times compared with infants born to non-Hispanic White mothers (Hispanic: aOR=2.32, 95% CI: 1.31-4.10; non-Hispanic Black: aOR=2.50, 95% CI: 1.49-4.18). During ages one to four years, children born to mothers of racial/ethnic minority groups had nearly

three times shorter one-way travel times compared with infants born to non-Hispanic White mothers (Hispanic mothers: aOR=2.79, 95% CI: 1.11-6.97; non-Hispanic Black mothers: aOR=2.86, 95% CI: 1.32-6.20).

Lower level of maternal education (< high school education) was associated with decreased odds of traveling less than or equal to 30 minutes to access hospital care for birth hospitalizations (aOR=0.58, 95% CI: 0.35-0.98) than mothers with a high school education. Across all infancy hospitalizations, lower level of maternal education was barely associated with lower odds of traveling 30 minutes or less (aOR=0.61, 95% CI: 0.37-0.99). No other predisposing characteristics were associated with travel time to access hospital care during infancy or ages one to four years.

5.6.3.3 Multivariable Results for Enabling Characteristics

Infants whose mothers were rural residents were consistently more likely to travel longer times to access hospital care for both birth and post-birth hospitalizations than infants whose mothers lived in urban areas (birth: aOR=0.54, 95% CI: 0.21-0.62; post-birth: aOR=0.43, 95% CI: 0.24-0.79). Across all infancy, infants living in rural areas were 63% more likely to travel more than 30 minutes to access hospital care (aOR=0.37, 95% CI: 0.22-0.63) than infants living in urban areas. Surprisingly, rural residence was not statistically associated with one-way travel time for hospitalizations during ages one to four years.

No single payer type had a statistically significant effect on travel time during infancy, however, having multiple health payers across all infancy hospitalizations was marginally associated with lower odds of traveling of less than or equal to 30 minutes to access hospital care (aOR=0.51, 95% CI: 0.27-0.97). No other enabling characteristics

were associated with travel time to access hospital care during infancy or ages one to four.

5.6.3.4 Multivariable Results for Need Characteristics

Parents of infants who were born preterm were 67% more likely to have shorter one-way travel times for birth hospitalization (aOR=1.67, 95% CI: 1.07-2.60) compared to infants who were not born preterm (Table 5.5a). Across all infancy hospitalizations, parents of infants who were born preterm birth were almost two times more likely to travel 30 minutes or less (aOR=1.88, 95% CI: 1.22-1.93), compared to infants who were not born preterm (Table 5.5b). No other need characteristics were associated with one-way travel time to access hospital care during ages one to four (Table 5.5c).

5.6.3.5 Multivariable Results Comparing Separate Models for Predisposing, Enabling, and Need Characteristics

In models that examined travel time by independent categories of predisposing, enabling, and need factors, I found no notable differences when compared to the full model. When modeling only the predisposing characteristics, hydrocephalus was not associated with travel time, however, non-isolated SB was associated with lower odds of traveling 30 minutes or less (aOR=0.51, 95% CI: 0.34-0.75). When modeling only the enabling characteristics, hydrocephalus was not associated with travel time, however, non-isolated SB was again associated with lower odds of traveling 30 minutes or less (aOR=0.58, 95% CI: 0.39-0.86). Finally, when modeling only need characteristics, both hydrocephalus (aOR=0.69; 95% CI: 0.49-0.96) and non-isolated SB (OR=0.60, 95% CI: 0.41-0.87) were associated with lower odds of traveling 30 minutes or less to access hospital care.

5.6.3.6 Multivariable Results Comparing Models with and without Hydrocephalus

Lastly, Table 5.5e shows unadjusted and adjusted OR and 95% CI for all covariables by the presence and absence of hydrocephalus for all infancy hospitalizations. When comparing the adjusted models for: 1) all infants with SB, 2) infants with SB and with hydrocephalus, and 3) infants with SB without hydrocephalus, there were few notable differences across the three models. Infants born to mothers of racial/ethnic minority groups and infants born preterm had consistently lower odds of shorter one-way travel times in each model, while rural residency was consistently associated with longer one-way travel times. Among infants with SB and hydrocephalus, higher maternal parity was associated with increased odds of traveling 30 minutes or less to access hospital care (aOR=1.78; 95% CI: 1.03-3.09), however, no association existed in the model with infants with no hydrocephalus or in the model with all infants. Lower level of maternal education was associated with odds of traveling longer to access hospital care among infants with hydrocephalus (aOR=0.42; 95% CI: 0.22-0.83) and in the model with all infants (OR=0.61; 95% CI: 0.37-0.99), but no association was found in the model with infants without hydrocephalus.

5.7 Discussion

5.7.1 Discussion of Travel Time and Distance by Age Category

This research examined travel time and distance to access hospital care for children born with spina bifida (SB) during infancy (from birth through one year) and ages one to four years. Results showed infants with SB traveled about 7% shorter one-way average times and distances to access care for their birth hospitalization than to access care for all infancy hospitalizations. During ages one to four years, average one-

way travel to access hospital care was slightly longer than travel during infancy. Comparing infancy hospitalizations to hospitalizations during ages one to four years, approximately the same percentage of children traveled over 60 minutes to access hospital care (22.2% of infants; 21.1% of children ages one to four years).

Examining one-way travel time and distance to access hospital care, children with SB, from birth through age four, traveled longer times and greater distances to access hospital care than travel reported to accessing health care in general populations [136, 240]. Findings from this study were consistent with studies that examined travel times and distances in populations of CSHCN [123, 138-140]. Modest differences with previous literature that examined geographic access to care among children with birth defects may be the result of differences in birth defects ascertainment methods, state population density and state-specific differences in the geographic distribution of health care facilities or services. Longer average travel times and distances for children, ages one to four years compared to infancy travel may reflect different patterns of health services needed and used by slightly older children, or may indicate intrastate residential relocations that could not be accounted for in this study. No empirical studies are available for comparison of differences of travel time and distance to access hospital care by child's age, including ages one to four years.

5.7.2 Discussion of Effects of Hydrocephalus and Isolated or Non-Isolated SB on Travel Time and Distance

Further addressing the first research question in terms of differences in travel time and distance for children by the presence of hydrocephalus and by isolated or non-isolated SB, bivariate results showed that infants with hydrocephalus traveled longer one-

way time and distance to access hospital care than infants without hydrocephalus. This result was expected as hydrocephalus is both an indirect indictor of the more severe form of SB known as myelomeningocele and because hydrocephalus itself adds a dimension of complexity to the SB presentation. Similarly, infants with non-isolated SB traveled longer times and distances than infants with isolated SB and was an expected result. Infants with both hydrocephalus and non-isolated SB experienced the greatest travel burden. The influence of hydrocephalus on isolated SB and non-isolated SB was most notable during the birth hospitalization and less so during post-birth hospitalizations.

The American Academy of Pediatrics advises that major congenital anomalies in infants and children be managed by specialists at pediatric referral centers and specifically suggests that infants with myelomeningocele should preferably be cared for by a pediatric neurosurgeon, as part of a medical-surgical team [241]. Among the 108 Florida hospitals used by infants in this study [140], twenty-six had nurseries with a designation of Level III care [242], indicating that the nurseries were able to provide complex neonatal surgery, pediatric neurosurgery, and neonatal cardiovascular surgery services [243]. The findings that children with more complex presentations of SB traveled longer travel times and distances to access hospital care are consistent with the number of Florida hospitals with Level III nursery care. Previous literature that suggests increasingly complex or severe forms of medical conditions may require more specialized and diverse medical care [17, 92, 244] that may not be available in local or community hospital settings.

There were no significant differences for travel time and distance to access hospital care during ages one to four years, based on the presence or absence of

hydrocephalus, by isolated or non-isolated SB, or by the presence of both hydrocephalus and non-isolated SB. These findings are consistent with a previous study that found medical care use for children with SB is highest in the first year of life [12, 14]. Infants with SB typically undergo major surgical repairs during the first few months of life, many of which require specialty care not available in all hospitals [241]. Follow-up care and hospitalizations after infancy may not require the same level of specialty care as during infancy, thus a child ages one to four years may receive adequate care at a community hospital.

I did not examine the reason for admission in this study. For example, treatment of a urinary tract infection, a common reason for hospitalization among individuals with SB [16, 245, 246], could possibly be managed at a community level hospital. Alternately, an admission may have been completely unrelated to the child's SB, for example, a hospitalization for pneumonia or an injury caused by an accident. Local or community hospitals may have the resources necessary to adequately care for routine conditions, thus reducing the travel burden to access care.

5.7.3 Discussion of Effects of Other Covariables on Travel Time and Distance

Addressing the second research question about other factors associated with travel time and distance to access hospital care, two predisposing characteristics were consistently associated with travel time and distance during infancy. Infants born to Hispanic or non-Hispanic Black mothers were less likely to travel more than 30 minutes to access care compared to infants of non-Hispanic White mothers. Maternal race/ethnicity was also the only characteristic that remained associated with greater travel time and distance during ages one to four years. All 108 Florida hospitals used by

children in this study were located in urban or urban cluster areas [140]. Higher percentages of Hispanic and non-Hispanic Black mothers in the study population lived in urban or urban cluster areas, and may provide a possible explanation for the consistently shorter travel distances among infants of mothers of racial/ethnic minority groups.

While shorter drive times may indicate better access to care, shorter travel time may also indicate a more limited understanding of options available for health care or access to fewer resources that facilitate travel and may not lead to better quality of services nor better health outcomes. Previous research has also shown that cultural influences, such as medical mistrust, limited proficiency in the English language, and hesitancy to adopt unfamiliar medical care can influence use of health care services [247-249]. The results of this study suggest that mothers in racial/ethnic minority groups may seek care for their children with SB at the closest and most familiar health care facility. Factors, such as family support, availability of transportation and transportation mode, familiarity with a facility, the inability to take time off from work or no childcare for siblings, may also contribute to shorter travel observed among mothers in racial/ethnic minority groups.

A second characteristic consistently associated with travel time and distance among infants with SB was a rural maternal residence at birth. As expected, infants of mothers with a rural residential address were less likely to travel less than 30 minutes to access hospital care compared with infants of mothers with an urban maternal residential address. As noted previously, all 108 Florida hospitals used by infants in this study were located in urban or urban cluster areas [140], thus minimizing travel times for people living in urban areas. Surprisingly, however, rural maternal residence was not associated

with longer travel during ages one to four years in either the unadjusted or the adjusted logistic regression models. These findings suggest that rural residents may seek hospital care at nearby community hospitals. It may be that a child's medical condition was stable and thus she or he required less complex care; on the other hand, if the family faced travel barriers, a child could have received less than optimal care.

Other covariables also influenced travel time and distance. Infants of mothers who were married or born in the United States were more likely to travel more than 30 minutes to access care for their infants compared with unmarried or foreign-born mothers. Married and foreign-born mothers in this study were more often of a minority racial/ethnic group. Mothers of a minority racial/ethnic group in this study were more likely to live in urban areas with shorter access to a hospital facility. An infant born preterm was more likely to travel less than 30 minutes to access hospital care. This finding was expected as a premature delivery may be unplanned and result in a *birth* hospitalization at the closest hospital. Infants of mothers with less than a high school education were also less likely to travel less than 30 minutes to access hospital care. I expected that lower maternal education might be associated with rural residence, but no association existed between maternal education and rural residence in this study population (data not shown). However, cell sizes were small (rural, no high school diploma: n=21) and had insufficient power to detect significant associations in this group. Surprisingly, the type of health care payer showed no association with travel time or distance to access hospital care during infancy, although one previous study also reported a similar finding [123]. While health payer has been associated with access to health care, particularly for CSHCN [108, 126, 127, 166, 250, 251], this finding suggests that

payer type may be associated with a different dimension of access to care than the geographic access described by the accessibility component [121].

In models that examined each category of predisposing, enabling, and need factors separately, no notable differences existed when compared to the full model, which included all predisposing, enabling and need factors. These results suggested that all three categories of characteristics described in the Aday and Andersen model [152] contributed to describing travel time and distance.

In summary, infants who have increasingly complex presentations of SB (SB with hydrocephalus or with non-isolated SB or an accompanying preterm birth) may experience greater travel time and distance during infancy than infants with SB but no additional comorbidities; this was particularly true for birth hospitalizations. The increased travel time and distance for children with hydrocephalus or non-isolated SB does not appear to continue after the first year of life, possibly signaling a reduced need for specialty hospital care as the child matures, and medical conditions stabilize. Maternal race/ethnicity and rural residence were the two demographic characteristics most consistently associated with travel time and distance in this study. Surprisingly, health care payer type was not associated with travel time and distance.

The interpretation of travel time and distance presents several challenges. A shorter travel experience may be interpreted positively as a reflection of easier access to hospital care. For CSHCN, however, appropriate care may only be available in a limited number of pediatric specialty centers that may require more travel for families. Some parents of children who travel shorter times and distances may have access to care, but not to appropriate, optimal, or multidisciplinary specialty care. Other children may live

near a hospital with a high level of specialty care and experience a shorter travel time and distance to access care, but face other barriers that may influence hospital use. Conversely, while long distances to access hospital care may be interpreted as a high travel burden, a family with health knowledge and resources, or for other reasons, may intentionally select a distant hospital for their child's care. Families of children with SB may choose hospitals based on a wide variety of factors not directly associated with travel time and distance that we were unable to measure in this study.

5.8 Strengths and Limitations

5.8.1 Innovation and Strengths in the Research Topic

This study examined the differences in geographic access to hospital care in a population-based, statewide study of unduplicated Florida children insured by different health care payer types. The study followed children for the first four years of life, which provided new opportunities to examine changes in access to care and associated predisposing, enabling, and need characteristics over time. The comparison of access to care findings by payer type and in the presence of hydrocephalus was also unique. With the exception of one study, which explored hospital costs for North Carolina children with SB and SB with hydrocephalus born 1995-2002 continuously enrolled in Medicaid [12], no similar work related to comorbidities and SB existed. Additionally, little work has been done to explore travel time and distance to access health care for children with SB [140], so each of these topics represents new or expanded areas of research.

5.8.2 Innovation and Strengths in Methodology

The study population for this research was an important strength in that it represented a robust and diverse group of children. The state of Florida was among the

fastest growing and the fourth most populous state in the 2000 U.S. Census [22, 23]. Florida was fourth in number of annual live births, second in number of live births to non-Hispanic Black women, and third in number of live births to Hispanic women, nationwide [22-24]. Florida also supports a statewide, population-based birth defects registry and a statewide agency for the collection of hospital discharge data that provided information for this project. This statewide, population-based study sample used linked, longitudinal data from the FBDR and the Florida AHCA, which provided a robust source of information for this project.

A second significant strength of the methodology, and perhaps the most significant, was the derivation of the variables for evaluating travel time and distance. The travel time and distance variables were calculated using network methodology, rather than Euclidian "straight line" distances. Topological road networks provide a more accurate measure of travel time and distance because they account for speed limits, one-way restrictions, and reflect connectivity between roads, including highways, overpasses, and access ramps [140, 252, 253]. Straight-line Euclidean distances typically underestimate the actual travel time and distance, and can thus introduce error in analyses [254].

Additionally, this research incorporated several methods that are not frequently used in health services research for birth defects and thus are both strengths and innovations of the proposed project. First, because of the nature of the data, the unit of analysis for this project was the individual child, rather than the more typical observation level of hospital admission or other aggregate level data, such as the hospital visit. Secondly, the dataset provided access to hospital discharge data for children from

multiple payer sources, rather than the more commonly researched single payer source. This allowed for a more complete picture of access to care across multiple payer types. Third, the linked, longitudinal data provided the opportunity to follow each child through early childhood to give unique insights into the differences in access to hospital care over time.

A final strength of this methodology is its ability to be replicated for other birth defects by health services researchers and public health researchers examining different types of birth defects in collaboration with other birth defect registries and state and federal agencies. These methods underscore the value of collaboration between local, state, and federal public health agencies, academic universities, public health and health services researchers and the use of geographic information system (GIS) methods.

The demographic findings are similar to the characteristics of all Florida-born infants during with study period with a few exceptions, which make this study generalizable. In comparison to all infants born in Florida, 1998-2007, the study population included a slightly lower proportion of Hispanic mothers (29% statewide compared to 25% in the study sample), a notably higher proportion of preterm births (27% compared to 11% in Florida 2007, 12.8% nationwide 2006), and a slightly higher proportion of infants with public payer sources (50% compared to 43%) [27, 255]. The higher percentage of preterm births was consistent with previous research that has reported an association between low birth weight and prematurity among infants with birth defects [172, 196]. In comparison specifically to other children with SB, about 26% of this study sample had non-isolated SB. This finding is generally consistent with previous research that found between 15% and 25% of infants with SB have non-isolated

SB [81, 161, 162]. Findings for death (8.5%) were also consistent with previous research on mortality among children with SB [56-59].

5.8.3 Limitations Inherent in the Data Sources

This research faced several limitations based on the data used. Infants identified for this study were based on the passive surveillance methodologies for identifying infants with birth defects using ICD-9-CM codes. Some birth defects surveillance systems actively identify birth defect diagnoses using modifications of the *British Pediatric Association (BPA) Classification of Diseases* [198, 199]. In contrast, passive birth defects surveillance systems, while widely used, do not actively verify the birth defect diagnosis by review of medical records, hospital charts, or nursery logs. Also, limited information on prenatal diagnosis is available in passive surveillance systems [27]. For Florida, there is no access to data on prenatal diagnosis for birth defects through the FBDR.

While passive surveillance techniques may lead to under-reporting or miss reporting of infants with birth defects or a specific defect type [27-29, 200, 201], the FBDR's overall completeness of ascertainment of birth defects has been estimated at 87%, with case ascertainment variation noted by specific defect [28, 29]. Because SB is relatively easy to detect, a passive surveillance system may be less of a limitation than with other birth defects that are more difficult to detect. In the FBDR data, ascertainment of infants with SB without anencephaly was 88.0% [29], a relatively high completeness of ascertainment of SB. In addition, because this analysis used data from the FBDR, it is a state-specific study, which may limit generalizability to other states or regions of the country.

The nature of the study sample also presented a limitation because the sample size was not constant over the ten-year study period. While the Florida AHCA provided ten years of hospital discharge data, the full ten years only applied to the children born in the first year of the study as per ascertainment methods of infants with birth defects used with the FBDR and is common with most birth defect registries in the United States. Each subsequent birth cohort had one less year of data, ending with the birth cohort of 2007, which had only one year of data. This decreasing sample size for each cohort resulted in a decreased statistical power, thus making the outcomes at risk for Type II error, the report of a false negative decision. To reduce this risk, I limited the analyses to the first four years of life.

Additionally, the principal payer source variable that was used in analyses of birth payer and payer types across the four years was an expected principal payer source. It is not known if this was the actual payer source. Furthermore, some infants may have dual payer sources (e.g., private and public payer) for a single hospitalization. Such information is not generally reported with hospital discharge data.

Another limitation was the fact that the data were based on Florida hospital administrative data. Administrative data may be at risk for error or inconsistent coding that could incorrectly code maternal residential addresses and hospital facility codes or introduce error in diagnostic coding. This data did not include information on families that sought care out-of-state for their child. Additionally, while approximately 290 Florida hospitals report data to the Florida AHCA, not all are required to report, including one Shriner's Hospital that provides care at no cost to patients, as well as long and short-term psychiatric hospitals, inpatient residential treatment and rehabilitation facilities, and

military hospitals [202]. The lack of reporting to the Florida AHCA means I was not able to capture access to care data on all the children within the Florida SB population under study. However, because data from 108 different Florida hospitals were represented in the data set [140] and because most of the non-reporting hospitals do not provide newborn care, the amount of data lost was likely limited and thus the findings of this research may be generalizable at least to the state of Florida.

Lastly, the use of administrative data does not capture all aspects of an individual's inclination to use health services resources, and specifically factors related to travel to access care. Characteristics, such as family resources, employment status, access to a personal vehicle, and health beliefs and health literacy, are not available in administrative data, but are all characteristics that may influence the use of hospital services separate from the influence of travel time and distance.

5.8.4 Limitations Inherent in Research Design

This research faced additional limitations resulting from the study design. Because of data limitations, I assumed a single maternal residential address (from infant birth records) for all admissions when calculating average one-way travel time and distance to hospitals of care. Families of children with SB, however, could have made one or more intrastate moves during the study period or children may move to live with someone other than their mothers. The assumption of the maternal address at the infant's delivery for all later hospital admissions likely introduced error in the calculations; however, Florida state law specifically prohibits follow-up contact by the FBDR or FDOH with families of children with birth defects, so addresses associated with subsequent hospital visits could not be confirmed.

No research has examined residential mobility among children with birth defects during infancy or in childhood. However, several studies provide general insight into residential mobility. A 2003 U.S. Census Bureau Current Population Survey found that 14% of all U.S. residents moved between 2002 and 2003 [256]. The same U.S. Census also reported that among individuals who moved, 59% moved within the same county and 19% moved to a different county within the same state [256]. Among all individuals who moved between 2002 and 2003, the U.S. Census reported that 19% moved to a different state [256]. A study of 1984 data from the population-based Maryland Birth Defects Reporting and Information System reported residential mobility among pregnant women of 20% and suggested that residential mobility likely resulted in misclassification of the exposure measured [257]. Similarly, research using geocoded data from the 1993-1997 Birth Defects Risk Factor Surveillance Study found that maternal mobility may introduce non-differential misclassification into analytic findings [258], as did two subsequent studies [259, 260]. Error introduced by residential mobility following an infant's birth is an important limitation that should be acknowledged for this study. In future research, residential addresses could be updated and geocoded at the zip code level for hospitalizations subsequent to the birth hospitalization using the Florida AHCA data. Future research that included collaborative, multistate work could also be useful in addressing the challenges introduced by residential mobility.

The use of network methodology and geocoding techniques involved additional assumptions that may have introduced error into the findings. First, I assumed that all travel was made by personal vehicles. No adjustments in time and travel distances were made for public transportation methods. Second, families were assumed to have driven

the shortest and fastest travel routes to access hospital care. This assumption may not be valid, as parents may have taken more familiar routes, chosen to avoid certain areas or types of roads, or linked trips to the hospital with other family commitments or needs (trip-chaining) [261, 262]. Also, the shortest travel times and distances may change throughout the day based on traffic congestion patterns, thus affecting the actual travel experience. Additionally, about 3% of the infants were geocoded at the ZIP-code level rather than the street address level [140], thus introducing a level of uncertainty about their actual travel time and distance. Any of these assumptions could have resulted in an over or understatement of the actual one-way travel time and distance. Time and travel distance were also reported as one-way travel and did not include the reverse trip. This method of report therefore underestimated the full travel burden to access hospital care.

Finally, while the study population was comprised of children with SB, the reasons for travel to access hospital care was unknown. While hospitalization may have been directly related to a children's SB diagnosis, hospitalizations could also occur because of other illnesses or injuries unrelated to SB. Any interpretations and conclusions must therefore be made with caution.

5.9 Implications for Public Health Practice and Research

This study suggests several points for consideration in the areas of public health and access to health care services. First, this research suggests that the use of birth defects surveillance data combined with geocoding methodology contributes vital information about patterns, predictors, and barriers to accessing hospital care for children with SB. While geocoding methodology has often been used in birth defects research to map geographic distribution of birth defects and to examine exposures to potential

teratogens [145-149, 263], this study demonstrates that the use of GIS methodology in association with birth defect registry data can provide valuable data related to health services use [264]. An understanding of patterns and predictors of the travel burden associated with access to hospital care may be important to the coordination of delivery of services by health care planners or by governmental providers, such a federal state, and local agencies, particularly those serving CSHCN [13, 123, 140]. Further exploration of these data may suggest geographic areas in which shortages of adequate health care resources exist.

Second, results identified several factors associated various travel burdens to access hospital care for children with SB. These findings suggest areas in which to focus heath care resources. Opportunities may exist to better educate parents or caregivers about the best options for health care for their child with SB and how to access those resources, especially if they include greater travel burdens. Information on insurance acceptance, ways to minimize travel costs, and means of obtaining support services such as housing away from home [265] could be helpful, especially if longer travel meant access to more appropriate specialty hospital care for a child with SB. However, the factors most often associated with travel time and distance that are reported in this research (clinical needs including hydrocephalus, non-isolated SB, and preterm birth; maternal race/ethnicity; and rural residence) each have a low degree of mutability. This observation reinforces the importance of primary prevention, with a particular focus on continued support for folic acid fortification of the U.S. food supply and continued education about prenatal intake of folic acid among women of childbearing age [50, 52,

116, 266] and other health practices that have been shown to reduce the risk of birth defects.

This study also suggests opportunities for further public health research or research related to access of health care services. First, future research could further explore factors and patterns associated with travel time and distance to access health care, including not only hospital care, but also outpatient care, and follow-up care at specialty centers. Factors warranting further examination could include the influence of public transportation or ownership of personal vehicles, daily traffic patterns, the effects of direct and indirect travel costs, and travel burdens by type of hospital; for example, comparing travel to children's specialty hospitals versus local community hospitals. Second, a more complete understanding of both individual and area-based socioeconomic measures could provide insights into the influence of socioeconomic status on choice of hospital and associated travel. Third, qualitative research could be important to gaining a deeper understanding of factors that influence parents' decisions related to travel and the selection of a hospital for the care their children with SB. A better understanding of qualitative findings, such as feelings, perceptions, barriers and conflicting pressures, previous hospital experiences, and other reasons for choosing one hospital or doctor over another, could inform our understanding of the role of personal decision-making in the selection of a hospital. Fourth, understanding the reasons for hospitalizations (e.g., the admitting diagnoses) could contribute to a more accurate understanding of the reason for choosing one hospital over an alternate location.

Finally, the findings of this study can help to inform research for other birth defects. Birth defects, such as orofacial clefts, congenital heart disease, and

chromosomal anomalies may also have multiple presentations, associated comorbidities, and require long-term specialty care. A better understanding of factors that influence access to care, including geographic location and travel time and distance, can suggest ways to identify and address barriers to care. Increased access to health care, whether through increased geographic accessibility to hospital care or by facilitating improvements in availability, accommodation, affordability, or acceptability of health care, may improve health outcomes, reduce health costs, and improve long-term quality of life for children with SB or other similar birth defects.

Table 5.1: Selected characteristics of Florida-born children with spina bifida and with a geocoded maternal residential address at birth, 1998-2007

Characteristics	All children with SB (n=612)		*Without* hydrocephalus (n=265, 43.3%)		*With* hydrocephalus (n=347, 56.7%)		p-value
	n	(%)	n	(%)	n	(%)	
Exposures of interest							
Hydrocephalus							
Yes	347	(56.7)					
No	265	(43.3)					
Spina bifida [1]							
Isolated	456	(74.5)	198	(74.7)	258	(74.4)	0.918
Non- isolated	156	(25.5)	67	(25.3)	89	(25.6)	
Hydrocephalus and SB							
No	523	(85.5)					
Yes	89	(14.5)					
Predisposing characteristics							
Maternal age (in years)							
≤ 24	224	(36.6)	78	(29.4)	146	(42.1)	**0.006**
25-29	164	(26.8)	79	(29.8)	85	(24.5)	
≥30	224	(36.6)	108	(40.8)	116	(33.4)	
Maternal race/ethnicity							
Non-Hispanic White	321	(52.4)	136	(51.3)	185	(53.3)	**0.005**
Hispanic	153	(25.0)	73	(27.6)	80	(23.1)	
Non-Hispanic Black	128	(20.9)	54	(20.4)	74	(21.3)	
Other	10	(1.6)	2	(0.7)	8	(2.3)	
Maternal nativity							
Born in U.S.	464	(75.8)	198	(74.7)	266	(76.7)	0.849
Foreign-born	146	(23.9)	66	(24.9)	80	(23.0)	
Missing	2	(0.3)	1	(0.4)	1	(0.3)	
Maternal marital status							
Married	366	(59.8)	162	(61.1)	204	(58.9)	0.558
Not married	246	(40.2)	103	(38.9)	143	(41.2)	
Maternal parity							
First child	237	(38.7)	99	(37.5)	138	(39.8)	0.234
Second subsequent child	374	(61.1)	165	(62.5)	209	(60.2)	
Missing	1	(0.2)					
Maternal education							
High school diploma or more	466	(76.1)	48	(18.3)	90	(26.3)	0.112
No high school diploma	138	(22.6)	214	(81.7)	249	(72.8)	
Missing	8	(1.3)					

Table 5.1 (continued)

	All children		Without hydrocephalus		With hydrocephalus		p-value
Sex of child							
Female	315	(51.5)	142	(53.6)	173	(49.9)	0.360
Male	297	(48.5)	123	(46.4)	174	(50.1)	
Enabling characteristics							
Prenatal care [2]							
Adequate prenatal care	445	(72.7)	194	(77.9)	251	(76.3)	0.647
Inadequate prenatal care	133	(21.7)	55	(22.1)	78	(23.7)	
Missing	34	(5.6)					
Rurality [3]							
Urban/urban cluster	525	(85.8)	226	(85.3)	299	(86.2)	0.756
Rural	87	(14.2)	39	(14.7)	48	(13.8)	
Payer for birth hospitalization [4]							
Public payer	292	(47.7)	128	(48.3)	192	(55.3)	0.113
Private payer	253	(41.3)	123	(46.4)	145	(47.8)	
Self or uninsured	24	(3.9)	14	(5.3)	10	(2.9)	
No birth hospitalization	43	(7.0)					
Payer type during infancy [4]							
Public payer only	305	(49.8)	123	(46.4)	182	(52.4)	0.097
Private payer only	235	(38.4)	114	(43.0)	121	(34.9)	
Self or uninsured	8	(1.3)	5	(1.9)	3	(0.9)	
Multiple payers	64	(10.5)	23	(8.7)	41	(11.8)	
Payer type, ages 1-4 years [4]							
Public payer only	117	(46.6)	29	(47.5)	88	(46.3)	0.157
Private payer only	68	(27.1)	21	(34.4)	47	(24.7)	
Multiple payers	66	(26.3)	11	(18.0)	55	(29.0)	
Need characteristics							
Preterm birth (< 37 weeks gestation)							
Yes	162	(26.5)	205	(78.0)	242	(69.9)	**0.027**
No	447	(73.0)	58	(22.0)	104	(30.1)	
Missing	3	(0.5)					
Low birth weight (< 2500 grams)							
Yes	120	(19.6)	206	(77.7)	285	(82.4)	0.153
No	491	(80.2)	59	(22.3)	61	(17.6)	
Missing	1	(0.2)					
Plurality							
Singleton birth	591	(96.6)	252	(95.1)	339	(97.7)	0.080
Multiple birth	21	(3.4)	13	(4.9)	8	(2.3)	

Table 5.1 (continued)

	All children		Without hydrocephalus		With hydrocephalus		p-value
Level of nursery care in birth hospital [5]							
Level III	437	(71.4)	152	(57.6)	285	(82.1)	**<0.0001**
Level I or II	174	(28.4)	112	(42.4)	62	(17.9)	
Missing	1	(0.2)					
Death [6]							
No death before age 4	560	(91.5)	239	(90.2)	320	(92.2)	0.310
Died during infancy	41	(6.7)	22	(8.3)	19	(5.6)	
Died in ages 1-4	11	(1.8)	4	(1.5)	8	(2.3)	

Note: Columns may not add to 100% because of missing or unknown values

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.

[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.

[2] Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care".

[3] Rurality calculated using the 2000 U.S. Census block group data corresponding to the maternal residential address at birth.

[4] Payers are expected health care payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no insurance or no third party coverage or less than 30%. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.

[5] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.

[6] Death were those that occurred during the study period, prior to December 31, 2008.

Table 5.2: One-way time and distance traveled to access hospital care by age and presence of comorbidities for Florida-born children with spina bifida, 1998-2007

Age of child (years)	n	Travel Time (minutes)				Travel Distance (miles)			
		Mean (SD)	Median (IQR)	Range	p-value	Mean (SD)	Median (IQR)	Range	p-value
Age 0 (Birth hospitalizations)	569	41.9 (64.9)	20.3 (31.9)	1.0-571		32.2 (53.8)	13.0 (25.7)	0.6-433	
Hydrocephalus									
No	249	31.0 (41.2)	18.5 (25.3)	1.0-346	**0.0004**	22.9 (35.0)	11.5 (19.2)	0.6-334	**<0.0001**
Yes	320	50.4 (77.6)	22.2 (40.4)	1.7-571		39.5 (63.9)	15.2 (33.5)	0.9-433	
Spina bifida [1]									
Isolated	426	39.3 (62.3)	19.3 (27.5)	1.7-571	**0.021**	30.0 (51.3)	12.3 (22.8)	0.9-433	**0.034**
Non-isolated	143	49.7 (71.8)	24.3 (39.1)	1.0-526		39.0 (60.4)	16.4 (32.8)	0.6-420	
Hydrocephalus and non-isolated SB									
No	491	38.8 (59.9)	19.4 (28.6)	1.0-571	**0.002**	29.4 (49.2)	12.3 (23.0)	0.6-433	**0.001**
Yes	78	61.8 (88.2)	27.3 (48.8)	4.1-526		50.1 (74.8)	21.9 (46.6)	2.6-420	
Age 0 (Post-birth hospitalizations)	461	50.5 (66.5)	28.0 (46.6)	2.4-732		38.5 (54.3)	19.7 (35.5)	1.3-598	
Hydrocephalus									
No	178	44.0 (41.2)	28.6 (46.7)	2.4-205	0.592	33.2 (34.4)	19.2 (36.2)	1.3-176	0.432
Yes	283	54.7 (78.1)	27.9 (48.7)	2.7-732		41.9 (63.5)	20.6 (37.4)	1.3-598	
Spina bifida									
Isolated	335	47.9 (59.6)	26.4 (48.1)	2.6-438	**0.017**	36.5 (48.5)	18.5 (34.8)	1.3-338	**0.014**
Non-isolated	126	57.6 (81.8)	33.5 (43.7)	2.4-732		44.1 (67.1)	25.0 (33.9)	1.3-598	
Hydrocephalus and non-isolated SB									
No	384	48.2 (27.8)	27.4 (49.7)	2.4-438	0.073	36.6 (47.0)	19.1 (36.2)	1.3-338	**0.034**
Yes	77	62.1 (98.6)	30.7 (36.4)	4.9-732		48.5 (81.0)	25.0 (31.7)	2.7-598	

Table 5.2 (continued)

	n	Travel time (minutes)				Travel distance (miles)			
		Mean (SD)	Median (IQR)	Range	p-value	Mean (SD)	Median (IQR)	Range	p-value
Age 0 (All infants)	612	45.1 (54.6)	25.9 (38.8)	2.4-494		34.5 (45.4)	18.1 (32.5)	1.2-404	
Hydrocephalus									
No	265	37.3 (40.1)	24.1 (33.5)	2.4-346	**0.009**	28.0 (34.5)	16.3 (27.6)	1.2-334	**0.004**
Yes	347	51.0 (62.9)	27.4 (47.3)	2.7- 494		39.4 (51.7)	20.8 (38.2)	1.3-404	
Spina bifida									
Isolated	456	42.3 (52.0)	24.2 (37.7)	2.7-338	**0.001**	32.1 (43.0)	16.6 (30.6)	1.3-338	**0.001**
Non-isolated	156	53.2 (61.1)	31.8 (43.7)	2.4-494		41.3 (51.2)	24.6 (36.9)	1.2-404	
Hydrocephalus and non-isolated SB									
No	523	42.4 (50.7)	24.8 (38.2)	2.4-437	**0.001**	32.1 (41.7)	16.9 (30.7)	1.2-388	**0.0002**
Yes	89	60.8 (72.4)	34.2 (49.3)	5.0-494		48.5 (61.1)	26.9 (42.2)	2.7-404	
Age 1-4 years	251	39.8 (46.0)	22.7 (38.2)	2.4-285.6		38.2 (50.1)	19.4 (40.4)	1.3-325.1	
Hydrocephalus									
No	61	32.5 (32.6)	17.2 (38.0)	2.4-116.6	0.088	30.9 (34.8)	14.6 (40.1)	1.3-124.0	0.141
Yes	190	42.2 (49.4)	24.3 (39.3)	2.7-285.6		40.5 (54.0)	21.1 (40.5)	1.3-325.1	
Spina bifida									
Isolated	180	39.4 (46.0)	22.4 (37.1)	2.6-262.4	0.362	37.9 (50.0)	19.1 (39.7)	1.3-301	0.343
Non -isolated	71	41.0 (46.4)	24.2 (43.2)	2.2-285.6		39.0 (50.7)	19.6 (42.8)	1.3-325.1	
Hydrocephalus and non-isolated SB									
No	204	38.6 (44.1)	22.4 (37.0)	2.4-262.4	0.217	37.1 (48.0)	18.4 (39.2)	1.3-301.4	0.261
Yes	47	45.1 (53.6)	24.3 (44.3)	4.7-285.6		43.0 (58.8)	21.8 (41.9)	2.4-325.1	

Note: Not all children had a hospitalization in each age category. SD, standard deviation. IQR, interquartile range. Statistical significance noted in bold at p-value of < 0.05 (based on Wilcoxon Rank Sum test)

Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.

[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.

Table 5.3: One-way time and distance traveled to access hospital care for Florida-born children with spina bifida by age category and presence of hydrocephalus, 1998-2007

Age of child (years)	n	Travel Time (minutes)						Travel Distance (miles)					
		≤ 30 minutes	>30 minutes	p-value	≤ 60 minutes	> 60 minutes	p-value	≤ 30 miles	>30 miles	p-value	≤ 60 miles	> 60 miles	p-value
		n (%)	n (%)		n (%)	n (%)		n (%)	n (%)		n (%)	n (%)	
All infants, age 0	612	345	267		476	136		403	209		515	97	
Hydrocephalus													
No	265	162 (47.0)	103 (38.6)	**0.038**	218 (45.8)	47 (34.6)	**0.020**	189 (46.9)	76 (36.4)	**0.013**	234 (45.4)	31 (32.0)	**0.014**
Yes	347	183 (53.0)	164 (61.4)		258 (54.2)	89 (65.4)		214 (53.1)	133 (63.6)		281 (54.6)	66 (68.0)	
All children, age 1-4	251	154	97		198	53		161	90		200	51	
Hydrocephalus													
No	61	41 (26.6)	20 (20.6)	0.280	50 (25.3)	11 (20.7)	0.498	42 (26.1)	19 (21.1)	0.378	50 (25.0)	11 (21.6)	0.610
Yes	190	113 (73.4)	77 (79.4)		148 (74.8)	42 (79.3)		119 (73.9)	71 (78.9)		150 (75.0)	40 (78.4)	

Note: Not all children had a hospitalization in each age category.
Note: Presence of hydrocephalus was based on coded data and not clinically verified.
Statistical significance noted in bold at p-value of < 0.05.

Table 5.4a: Selected characteristics for Florida-born infants with spina bifida by travel time and distance to access hospital care during infancy, 1998-2007 (n=612)

Characteristics	≤ 30 Minutes (n=345)		> 30 Minutes (n=267)		p-value	≤ 30 Miles (n=403)		> 30 Miles (n=209)		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Exposures of interest										
Hydrocephalus										
Yes	183	(53.0)	164	(61.4)	**0.038**	214	(53.1)	133	(63.6)	**0.013**
No	162	(47.0)	103	(38.6)		189	(46.9)	76	(36.4)	
Spina bifida [1]										
Isolated spina bifida	273	(79.1)	183	(68.5)	**0.003**	313	(77.7)	143	(68.4)	**0.013**
Non-isolated spina bifida	72	(20.9)	84	(31.5)		90	(22.3)	66	(31.6)	
Predisposing characteristics										
Maternal age (in years)										
< 25	129	(37.4)	95	(35.6)	0.897	150	(37.2)	74	(35.4)	0.883
25-29	91	(26.4)	73	(27.3)		108	(26.8)	56	(26.8)	
≥ 30	125	(36.2)	99	(37.1)		145	(36.0)	79	(37.8)	
Maternal race/ethnicity										
Non-Hispanic White	142	(41.2)	179	(67.0)	**<0.0001**	177	(43.9)	144	(68.9)	**<0.0001**
Hispanic	106	(30.7)	47	(17.6)		118	(29.3)	35	(16.7)	
Non-Hispanic Black	90	(26.1)	38	(14.2)		100	(24.8)	28	(13.4)	
Other	7	(2.0)	3	(1.1)		8	(2.0)	2	(1.0)	
Maternal nativity										
Born in U.S.	240	(70.0)	224	(83.9)	**0.0002**	287	(71.6)	177	(84.7)	**0.002**
Foreign-born	103	(30.0)	43	(16.1)		114	(28.4)	32	(15.3)	
Maternal marital status										
Married	190	(55.1)	176	(65.9)	**0.007**	226	(56.1)	140	(67.0)	**0.009**
Not married	155	(44.9)	91	(34.1)		177	(43.9)	69	(33.0)	

Table 5.4a (continued)

	≤ 30 Minutes		> 30 Minutes		p-value	≤ 30 Miles		> 30 Miles		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Maternal parity										
First child	127	(36.9)	110	(41.2)	0.282	147	(36.6)	90	(43.1)	0.118
Second or subsequent child	217	(63.1)	157	(58.8)		255	(63.4)	119	(56.9)	
Maternal education										
High school diploma or more	264	(77.6)	202	(76.5)	0.742	311	(78.1)	155	(75.2)	0.421
No high school diploma	76	(22.4)	62	(23.5)		87	(21.9)	51	(24.8)	
Sex of child										
Female	181	(52.5)	134	(50.2)	0.576	211	(52.4)	104	(49.8)	0.542
Male	164	(47.5)	133	(49.8)		192	(47.6)	105	(50.2)	
Enabling characteristics										
Prenatal care [2]										
Adequate prenatal care	248	(76.5)	197	(77.6)	0.773	292	(77.0)	153	(76.9)	0.965
Inadequate prenatal care	76	(23.5)	57	(22.4)		87	(23.0)	46	(23.1)	
Residential rurality [3]										
Urban /urban cluster	317	(91.9)	208	(77.9)	**<0.0001**	366	(90.8)	159	(76.1)	**<0.0001**
Rural	28	(8.1)	59	(22.1)		37	(9.2)	50	(23.9)	
Payer for birth hospitalization [4]										
Public payer	167	(48.4)	125	(46.8)	0.742	198	(49.1)	94	(45.0)	0.684
Private payer	137	(39.1)	116	(43.5)		161	(40.0)	92	(44.0)	
Self or uninsured	15	(4.4)	9	(3.4)		17	(4.2)	7	(3.4)	
No birth hospitalization	26	(7.5)	17	(6.4)		27	(6.7)	16	(7.7)	

Table 5.4a (continued)

	≤ 30 Minutes		> 30 Minutes		p-value	≤ 30 Miles		> 30 Miles		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Payer type infancy [4]										
Public payer only	183	(53.0)	122	(45.7)	0.202	210	(52.1)	95	(45.5)	0.381
Private payer only	127	(36.8)	108	(40.5)		148	(36.7)	87	(41.6)	
Self or uninsured	5	(1.5)	3	(1.1)		6	(1.5)	2	(1.0)	
Multiple payers	30	(8.7)	34	(12.7)		39	(9.7)	25	(12.0)	
Need characteristics										
Preterm birth (< 37 weeks gestation)										
Yes	105	(30.7)	57	(21.4)	**0.010**	117	(29.3)	45	(21.5)	**0.041**
No	237	(69.3)	210	(78.6)		283	(70.7)	164	(78.5)	
Low birth weight (< 2500 grams)										
Yes	75	(21.8)	45	(16.9)	0.127	84	(20.9)	36	(17.2)	0.279
No	269	(78.2)	222	(83.1)		318	(79.1)	173	(82.8)	
Plurality										
Singleton birth	332	(96.2)	259	(97.0)	0.603	387	(96.0)	204	(97.6)	0.309
Multiple birth	13	(3.8)	8	(3.0)		16	(4.0)	5	(2.4)	
Level of nursery care in birth hospital [5]										
Level III	247	(71.8)	190	(71.2)	0.844	285	(70.9)	152	(72.7)	0.647
Level I or II	97	(28.2)	77	(28.8)		117	(29.1)	57	(27.3)	
Death [6]										
No death before age 4 years	316	(91.6)	244	(91.4)	0.992	369	(91.6)	191	(91.4)	0.988
Died in infancy	23	(6.7)	18	(6.7)		27	(6.7)	14	(6.7)	
Died in ages 1-4	6	(1.7)	5	(1.9)		7	(1.7)	4	(1.9)	

Note: Columns may not add to 100% because of missing or unknown values. Numbers in bold are statistically significant at a p-value of <0.05
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.
[2] Adequacy of prenatal care was determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered “adequate prenatal care”.

[3] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[4] Payers are expected health care payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no insurance or no third party coverage or less than 30%. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[5] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.
[6] Deaths were those that occurred during the study period, prior to December 31, 2008

Table 5.4b: Selected characteristics for Florida-born children with spina bifida by travel time and distance to access hospital care during ages 1-4 years, 1998-2007 (n=251)

Characteristics	≤ 30 Minutes (n=154)		> 30 Minutes (n=97)		p-value	≤ 30 Miles (n=161		> 30 Miles (n=90)		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Exposures of interest										
Hydrocephalus										
Yes	113	(73.4)	77	(79.4)	0.280	119	(73.9)	71	(78.9)	0.378
No	41	(26.6)	20	(20.6)		42	(26.1)	19	(21.1)	
Spina bifida [1]										
Isolated	114	(74.0)	66	(68.0)	0.305	118	(73.3)	62	(68.9)	0.458
Non-isolated	40	(26.0)	31	(32.0)		43	(26.7)	28	(31.1)	
Predisposing characteristics										
Maternal age (in years)										
< 25	61	(39.6)	42	(43.3)	0.729	66	(41.0)	37	(41.1)	0.898
25-29	33	(21.4)	22	(22.7)		34	(21.1)	21	(23.3)	
≥ 30	60	(39.0)	33	(34.0)		61	(37.9)	32	(35.6)	
Maternal race/ethnicity										
Non-Hispanic White	62	(40.3)	61	(62.9)	0.015	66	(41.0)	57	(63.3)	0.022
Hispanic	45	(29.2)	21	(21.7)		47	(29.2)	19	(21.1)	
Non-Hispanic Black	41	(26.6)	14	(14.4)		42	(26.1)	13	(14.4)	
Other	6	(3.9)	1	(1.0)		6	(3.7)	1	(1.1)	
Maternal nativity										
Born in U.S.	114	(74.5)	77	(79.4)	0.493	119	(74.4)	72	(80.0)	0.455
Foreign-born	39	(25.5)	20	(20.6)		41	(25.6)	18	(20.0)	
Maternal marital status										
Married	84	(54.6)	56	(57.7)	0.621	88	(56.7)	52	(57.8)	0.633
Not married	70	(45.4)	41	(42.3)		73	(45.3)	38	(42.2)	

Table 5.4b (continued)

	≤ 30 Minutes		> 30 Minutes		p-value	≤ 30 Miles		> 30 Miles		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Maternal parity										
First child	57	(37.0)	39	(40.2)	0.612	62	(38.5)	34	(37.8)	0.909
Second or subsequent child	97	(63.0)	58	(59.8)		99	(61.5)	56	(62.2)	
Maternal education										
High school diploma or more	112	(73.7)	68	(72.3)	0.817	117	(73.6)	63	(72.4)	0.843
No high school diploma	40	(26.3)	26	(27.7)		42	(26.4)	24	(28.6)	
Sex of child										
Female	83	(53.9)	50	(51.6)	0.717	87	(54.0)	46	(51.1)	0.656
Male	71	(46.1)	47	(48.5)		74	(46.0)	44	(48.9)	
Enabling characteristics										
Rurality [2]										
Urban/urban cluster	143	(92.9)	83	(85.6)	0.060	148	(91.9)	78	(86.7)	0.182
Rural	11	(7.1)	14	(14.4)		13	(8.1)	12	(13.3)	
Payer type ages 1-4 years [3]										
Public payer only	74	(48.0)	43	(44.3)	0.375	79	(49.1)	38	(42.2)	0.373
Private payer only	37	(24.0)	32	(32.0)		39	(24.2)	29	(32.2)	
Multiple payers	43	(28.0)	23	(23.7)		43	(26.7)	23	(25.6)	
Need characteristics										
Preterm birth (< 37 weeks gestation)										
Yes	46	(30.1)	25	(25.8)	0.463	46	(28.8)	25	(72.2)	0.870
No	107	(69.9)	72	(74.2)		114	(71.2)	65	(27.8)	

Table 5.4b (continued)

	≤ 30 Minutes		> 30 Minutes		p-value	≤ 30 Miles		> 30 Miles		p-value
	n	(%)	n	(%)		n	(%)	n	(%)	
Low birth weight (< 2500 grams)										
Yes	25	(16.3)	16	(16.5)	0.974	25	(15.6)	16	(17.8)	0.659
No	128	(86.7)	81	(83.5)		135	(84.4)	74	(82.2)	
Plurality										
Singleton birth	151	(98.1)	95	(97.9)	0.950	158	(98.1)	88	(97.8)	0.845
Multiple birth	3	(1.9)	2	(2.1)		3	(1.9)	2	(2.2)	
Death [4]										
No death before age 4 years	149	(96.8)	93	(95.9)	0.716	155	(96.3)	87	(96.7)	0.872
Died in ages 1-4	5	(3.2)	4	(4.1)		6	(3.7)	3	(3.3)	

Note: Columns may not add to 100% because of missing or unknown values. Numbers in bold are statistically significant at a p-value of <0.05.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Isolated spina bifida is defined as SB with no additional major defects, other than the sequence of defects related to SB.
[2] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[3] Payers are expected health care payers. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[4] Deaths were those that occurred during the study period, prior to December 31, 2008.

Table 5.4c: One-way time and distance traveled to access hospital care by health care payer type for Florida-born infants with spina bifida, 1998-2007

Health care payers[1]	n	Travel Time (minutes)				Travel Distance (miles)			
		Mean (SD)	Median (IQR)	Range	p-value	Mean (SD)	Median (IQR)	Range	p-value
Birth hospitalizations [2]									
Private	253	39.8 (61.3)	20.9 (25.4)	2.4-571		30.2 (49.0)	14.2 (21.8)	1.2-433	
Public	292	44.5 (69.3)	19.8 (35.5)	1.1-526	0.5943	34.6 (58.8)	12.2 (28.8)	0.6-421	0.4389
Self-payer or under-insured	24	33.7 (42.0)	16.8 (32.2)	4.9-180		24.9 (35.1)	11.0 (24.4)	2.5-156	
Infancy hospitalizations									
Private	235	53.9 (77.8)	27.2 (49.4)	3.1-732		41.8 (64.6)	19.1 (36.2)	1.2-598	
Public	305	46.7 (64.5)	22.6 (39.5)	2.9-433	0.1180	35.9 (54.3)	14.9 (31.6)	1.3-334	0.1092
Self-payer or under-insured	8	50.5 (56.9)	25.9 (38.9)	8.1-180		38.7 (49.9)	20.1 (29.7)	3.9-155	
Multiple	64	49.4 (58.6)	30.5 (47.0)	4.9-311		38.2 (48.6)	24.4 (36.5)	2.5-243	

Note: Not all children had a hospitalization in each age category. SD, standard deviation. IQR, interquartile range.
Statistical significance noted in bold at p-value of < 0.05 (based on Wilcoxon Rank Sum test).
[1] Health care payer is expected payer. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different health care payer sources over multiple hospitalizations
[2] Birth hospitalizations defined as a first hospitalization with age at admission of 0 days or a first hospitalization with an age at admission of 1 day with an accompanying indication of hospital transfer

Table 5.5a Unadjusted and adjusted odds ratio estimates and 95% confidence intervals for the association between selected characteristics and average one-way travel time to access birth and post-birth hospitalizations during infancy for Florida-born children with spina bifida, 1998-2007 [>30 minutes vs. ≤ 30 minutes (reference)]

Characteristics	Birth hospitalizations[1] (n=569)				Post-Birth Hospitalizations (n=461)			
	Unadjusted model		Adjusted model		Unadjusted models		Adjusted model	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Exposures of interest								
Hydrocephalus								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	0.66	**(0.47-0.92)**	0.70	(0.47-1.04)	0.97	(0.67-1.42)	1.11	(0.73-1.70)
Spina Bifida								
Isolated [2]	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	**0.52**	**(0.36-0.77)**	**0.50**	**(0.32-0.79)**	**0.60**	**(0.40-0.90)**	**0.61**	**(0.38-0.96)**
Predisposing characteristics								
Maternal age (in years)								
<25	1.02	(0.72-1.43)	1.10	(0.65-1.75)	1.07	(0.73-1.56)	1.20	(0.68-2.13)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	1.04	(0.74-1.47)	0.88	(0.55-1.42)	0.95	(0.65-1.39)	0.82	(0.49-1.37)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	**2.88**	**(1.87-4.44)**	**2.31**	**(1.29-4.15)**	**2.58**	**(1.64-4.08)**	**2.10**	**(1.13-3.92)**
Non-Hispanic Black	**2.95**	**(1.89-4.62)**	**2.33**	**(1.38-3.93)**	**2.02**	**(1.23-3.31)**	**1.82**	**(1.02-3.24)**
Other	2.96	(0.75-11.7)	1.67	(0.36-7.44)	2.61	(0.47-14.6)	1.56	(0.24-10.1)
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	**2.36**	**(1.53-3.62)**	1.75	(0.97-3.13)	**2.06**	**(1.31-3.26)**	1.52	(0.83-2.83)

Table 5.5a (continued)

Characteristics	Birth hospitalizations[1]				Post-Birth Hospitalizations			
	Unadjusted model		Adjusted model		Unadjusted models		Adjusted model	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	**1.54**	**(1.09-2.17)**	1.32	(0.82-2.15)	1.16	(0.80-1.69)	0.93	(0.56-1.54)
Maternal parity								
First child	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Second or subsequent child	1.14	(0.81-1.61)	1.17	(0.78-1.75)	1.32	(0.91-1.92)	1.29	(0.84-1.99)
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	0.85	(0.57-1.27)	**0.58**	**(0.35-0.98)**	1.07	(0.70-1.65)	0.75	(0.43-1.30)
Sex of child								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.86	(0.62-1.20)	0.87	(0.60-1.27)	0.89	(0.61-1.28)	0.83	(0.55-1.24)
Enabling characteristics								
Adequate of prenatal care[3]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	1.05	(0.70-1.57)	0.98	(0.61-1.57)	1.03	(0.66-1.58)	0.95	(0.58-1.56)
Residential rurality[4]								
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	**0.30**	**(0.18-0.49)**	**0.37**	**(0.21-0.62)**	**0.34**	**(0.19-0.60)**	**0.43**	**(0.24-0.79)**
Payer for birth hospitalization[5]								
Public payer	1.13	(0.81-1.59)	0.92	(0.59-1.43)				
Private payer	1.00	(1.00)	1.00	(1.00)				
Self or uninsured	1.41	(0.60-3.34)	0.89	(0.34-2.32)				

Table 5.5a (continued)

Characteristics	Birth hospitalizations[1]				Post-Birth Hospitalizations			
	Unadjusted model		Adjusted model		Unadjusted models		Adjusted model	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Payer type for infancy [5]								
Public payer only					1.21	(0.81-1.80)	0.89	(0.53-1.51)
Private payer only					1.00	(1.00)	1.00	(1.00)
Multiple payers					0.78	(0.44-1.39)	0.55	(0.28-1.07)
Need characteristics								
Preterm birth (< 37 weeks)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.58**	**(1.08-2.33)**	**1.67**	**(1.07-2.60)**	**1.57**	**(1.02-2.40)**	1.61	(0.99-2.62)
Level of nursery care [6]								
Level III	1.00	(1.00)	1.00	(1.00)				
Level I or II	0.97	(0.68-1.39)	0.93	(0.60-1.42)				
Death [7]								
No death	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Death during infancy	0.96	(0.50-1.82)	0.93	(0.43-2.02)	0.89	(0.35-2.27)	0.88	(0.30-2.58)
Death in ages 1-4 years	0.94	(0.28-3.11)	1.53	(0.25-9.26)	1.07	(0.32-3.55)	1.75	(0.34-8.98)

uOR=odds ratio, aOR=adjusted odds ratio. 95% CI= 95% confidence interval. Values in bold are statistically significant. Models are adjusted for all covariables.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Birth hospitalizations defined as a first hospitalization with age at admission of 0 days or a first hospitalization with an age at admission of 1 day with an accompanying indication of hospital transfer.
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Payer is expected payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations
[6] Level 3 is highest level of hospital nursery care. Level 1 is the lowest level of hospital nursery care.
[7] Deaths were those that occurred during the study period, prior to December 31, 2008

Table 5.5b Unadjusted and adjusted odds ratio estimates and 95% confidence intervals for the association between selected characteristics and average one-way travel time to access hospitalizations during all infancy for Florida-born children with spina bifida, 1998-2007 [>30 minutes vs. ≤ 30 minutes (reference)]

Characteristics	Hospitalizations, *Infancy*							
	Unadjusted models				Adjusted models [1]			
	all infants (n=612)		excludes deaths (n=571)		all infants (n=612)		excludes deaths (n=571)	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Exposures of interest								
Hydrocephalus								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**0.71**	**(0.51-0.98)**	0.75	(0.53-1.05)	0.78	(0.54-1.13)	0.80	(0.55-1.17)
Spina Bifida								
Isolated	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	**0.58**	**(0.40-0.83)**	**0.58**	**(0.39-0.85)**	**0.58**	**(0.38-0.89)**	**0.58**	**(0.37-0.91)**
Predisposing characteristics								
Maternal age (in years)								
<25	1.08	(0.78-1.51)	1.12	(0.79-1.57)	1.12	(0.67-1.86)	1.04	(0.61-1.76)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	0.96	(0.69-1.34)	0.88	(0.63-1.25)	0.80	(0.51-1.27)	0.78	(0.49-1.25)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	**2.84**	**(1.89-4.28)**	**2.68**	**(1.77-4.06)**	**2.32**	**(1.31-4.10)**	**2.21**	**(1.24-3.95)**
Non-Hispanic Black	**2.99**	**(1.93-4.63)**	**3.15**	**(1.97-5.03)**	**2.50**	**(1.49-4.18)**	**2.72**	**(1.57-4.69)**
Other	2.94	(0.75-11.6)	2.90	(0.77-11.4)	1.67	(0.38-7.44)	1.67	(0.37-7.45)
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	**2.24**	**(1.50-3.33)**	**2.17**	**(1.43-3.27)**	1.74	(0.99-3.05)	1.72	(0.97-3.06)

Table 5.5b (continued)

Characteristics	Hospitalizations, *Infancy*							
	Unadjusted models				Adjusted models [1]			
	all infants		excludes deaths		all infants		excludes deaths	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	**1.58**	**(1.13-2.20)**	**1.68**	**(1.19-2.37)**	1.28	(0.81-2.03)	1.40	(0.87-2.25)
Maternal parity								
First child	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Second or subsequent child	1.20	(0.86-1.66)	1.14	(0.81-1.60)	1.22	(0.82-1.80)	1.17	(0.78-1.76)
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	0.94	(0.64-1.37)	0.94	(0.63-1.40)	**0.61**	**(0.37-0.99)**	**0.58**	**(0.34-0.97)**
Sex of child								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.91	(0.66-1.26)	0.94	(0.67-1.30)	0.94	(0.65-1.35)	0.94	(0.65-1.37)
Enabling characteristics								
Adequate prenatal care [3]								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	0.94	(0.64-1.40)	0.96	(0.64-1.44)	0.87	(0.55-1.37)	0.87	(0.54-1.40)
Residential rurality [4]								
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	**0.31**	**(0.19-0.51)**	**0.31**	**(0.19-0.51)**	**0.37**	**(0.22-0.63)**	**0.37**	**(0.21-0.64)**
Payer type for infancy [5]								
Public payer only	1.28	(0.90-1.80)	1.38	(0.97-1.97)	0.92	(0.59-1.45)	0.95	(0.59-1.51)
Private payer only	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Self or uninsured	1.42	(0.33-6.07)	1.35	(0.22-8.21)	0.90	(0.18-4.48)	1.18	(0.16-8.67)
Multiple payers	0.75	(0.43-1.31)	0.87	(0.49-1.54)	0.51	(0.27-0.97)	0.54	(0.28-1.04)

Table 5.5b (continued)

Characteristics	Hospitalizations, *Infancy*							
	Unadjusted models				Adjusted models [1]			
	all infants		excludes deaths		all infants		excludes deaths	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Need characteristics								
Preterm birth (< 37 weeks)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.63**	**(1.13-2.37)**	**1.70**	**(1.15-2.51)**	**1.88**	**(1.22-2.89)**	**1.93**	**(1.23-3.03)**
Death [6]								
No death	1.00	(1.00)			1.00	(1.00)		
Death during infancy	0.99	(0.52-1.87)		excluded	0.92	(0.45-1.99)		excluded
Death in ages 1-4 years	0.93	(0.28-3.07)			1.53	(0.27-8.70)		

uOR=odds ratio, aOR=adjusted odds ratio. 95% CI= 95% confidence interval. Values in bold are statistically significant.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Adjusted for all covariates in the model.
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Payer is expected payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations
[6] Deaths were those that occurred during the study period, prior to December 31, 2008

Table 5.5c Unadjusted and adjusted odds ratio estimates and 95% confidence intervals for the association between selected characteristics and average one-way travel time to access hospitalizations during ages 1-4 years for Florida-born children with spina bifida, 1998-2007 [>30 minutes vs. ≤ 30 minutes (reference)]

Characteristics	Hospitalizations, *Ages 1-4 years*							
	Unadjusted models				Adjusted models [1]			
	all children (n=251)		excludes deaths (n=242)		all children (n=251)		excludes deaths (n=242)	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Exposures of interest								
Hydrocephalus								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	0.72	(0.39-1.32)	0.77	(0.42-1.43)	0.77	(0.39-1.51)	0.81	(0.41-1.58)
Spina Bifida [2]								
Isolated	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	0.75	(0.43-1.31)	0.78	(0.44-1.39)	0.86	(0.46-1.60)	0.86	(0.46-1.60)
Predisposing characteristics								
Maternal age (in years)								
<25	0.86	(0.51-1.44)	0.88	(0.52-1.48)	0.77	(0.33-1.79)	0.78	(0.34-1.81)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≥30	1.24	(0.73-2.10)	1.18	(0.69-2.03)	1.29	(0.60-2.74)	1.29	(0.59-2.82)
Maternal race/ethnicity								
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	**2.11**	**(1.13-3.95)**	**2.29**	**(1.20-4.37)**	**2.79**	**(1.11-6.97)**	**3.00**	**(1.18-7.67)**
Non-Hispanic Black	**2.88**	**(1.43-5.82)**	**2.56**	**(1.26-5.21)**	**2.86**	**(1.32-6.20)**	**2.51**	**(1.15-5.47)**
Other	5.90	(0.69-50.5)	5.80	(0.68-49.6)	7.72	(0.79-75.2)	7.64	(0.78-74.6)
Maternal nativity								
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	1.32	(0.71-2.43)	1.34	(0.71-2.53)	0.80	(0.33-1.97)	0.75	(0.30-1.89)

Table 5.5c (continued)

Characteristics	Hospitalizations, *Ages 1-4 years*							
	Unadjusted models				Adjusted models [1]			
	All children		excludes deaths		all children		excludes deaths	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Maternal marital status								
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	1.14	(0.68-1.90)	1.16	(0.69-1.96)	0.84	(0.42-1.64)	0.87	(0.44-1.72)
Maternal parity								
First child	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Second or subsequent child	1.14	(0.68-1.93)	1.08	(0.63-1.84)	1.04	(0.58-1.87)	1.01	(0.55-1.84)
Maternal education								
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
No high school diploma	0.93	(0.53-1.67)	1.01	(0.56-1.82)	0.73	(0.36-1.51)	0.74	(0.35-1.54)
Sex of child								
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.91	(0.55-1.51)	0.87	(0.52-1.46)	0.89	(0.51-1.56)	0.87	(0.50-1.53)
Enabling characteristics								
Rurality [3]								
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	0.46	(0.20-1.05)	0.49	(0.21-1.15)	0.51	(0.21-1.26)	0.53	(0.21-1.33)
Payer type [4]								
Public payer only	1.44	(0.79-2.65)	1.56	(0.84-2.91)	1.73	(0.79-3.82)	1.73	(0.77-3.90)
Private payer only	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Multiple payers	1.57	(0.78-3.14)	1.53	(0.75-3.10)	1.99	(0.87-4.53)	1.88	(0.81-4.37)

Table 5.5c (continued)

Characteristics	Hospitalizations, *Ages 1-4 years*							
	Unadjusted models				Adjusted models [1]			
	all children		excludes deaths		all children		excludes deaths	
	uOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Need characteristics								
Preterm birth (< 37 weeks gestation)								
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	1.24	(0.70-2.19)	1.29	(0.72-2.32)	1.15	(0.61-2.16)	2.24	(0.65-2.37)
Death [5]								
No death	1.00	(1.00)		Deaths excluded	1.00	(1.00)		Deaths excluded
Death ages 1-4 years	0.78	(0.20-2.98)			0.93	(0.18-4.86)		

uOR=odds ratio, aOR=adjusted odds ratio. 95% CI= 95% confidence interval. Values in bold are statistically significant.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Adjusted for all covariates in the model.
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[4] Payer is expected payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[5] Deaths were those that occurred during the study period, prior to December 31, 2008

Table 5.5d Predisposing, enabling, and need models for unadjusted and adjusted odds ratio estimates and 95% confidence intervals for the association between selected characteristics and average one-way travel time to access all hospitalizations during infancy for Florida-born children with spina bifida, 1998-2007 [>30 minutes vs. ≤30 minutes (reference)]

Characteristics	Full model				Model 1		Model 2		Model 3	
	Unadjusted model		Adjusted model [1]		Adjusted for *Predisposing* factors only		Adjusted for *Enabling* factors only		Adjusted for *Need* factors only	
	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Exposures of interest										
Hydrocephalus										
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	0.71	(0.51-0.98)	0.78	(0.54-1.13)	0.75	(0.53-1.06)	0.77	(0.54-1.08)	**0.69**	**(0.49-0.96)**
Spina Bifida										
Isolated [2]	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	**0.58**	**(0.40-0.83)**	**0.58**	**(0.38-0.89)**	**0.51**	**(0.34-0.75)**	**0.58**	**(0.39-0.86)**	**0.60**	**(0.41-0.87)**
Predisposing characteristics										
Maternal age (in years)										
<25	1.08	(0.78-1.51)	1.12	(0.67-1.86)	1.03	(0.64-1.66)				
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
≥30	0.96	(0.69-1.34)	0.80	(0.51-1.27)	0.94	(0.61-1.44)				
Maternal race/ethnicity										
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
Hispanic	**2.84**	**(1.89-4.28)**	**2.32**	**(1.31-4.10)**	**2.32**	**(1.36-3.96)**				
Non-Hispanic Black	**2.99**	**(1.93-4.63)**	**2.50**	**(1.49-4.18)**	**2.41**	**(1.49-3.89)**				
Other	2.94	(0.75-11.6)	1.67	(0.38-7.44)	1.77	(0.40-7.88)				
Maternal nativity										
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
Foreign-born	2.23	(1.50-3.33)	1.74	(0.99-3.05)	1.56	(0.93-2.65)				

Table 5.5d (continued)

Characteristics	Full model				Model 1		Model 2		Model 3	
	Unadjusted model		Adjusted model [1]		Adjusted for *Predisposing* factors only		Adjusted for *Enabling* factors only		Adjusted for *Need* factors only	
	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Maternal marital status										
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
Not married	**1.58**	**(1.13-2.20)**	1.28	(0.81-2.03)	**1.62**	**(1.07-2.45)**				
Maternal parity										
First child	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
Second or subsequent child	1.20	(0.86-1.66)	1.22	(0.82-1.80)	1.22	(0.85-1.75)				
Maternal education										
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
< High school diploma	0.94	(0.64-1.37)	**0.61**	**(0.37-0.99)**	**0.59**	**(0.38-0.94)**				
Child sex										
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)				
Male	0.91	(0.66-1.26)	0.94	(0.65-1.35)	1.02	(0.72-1.43)				
Enabling characteristics										
Adequate prenatal care [3]										
No	1.00	(1.00)	1.00	(1.00)			1.00	(1.00)		
Yes	0.94	(0.64-1.40)	0.87	(0.55-1.37)			0.89	(0.59-1.36)		
Residential rurality [4]										
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)			1.00	(1.00)		
Rural	**0.31**	**(0.19-0.51)**	**0.37**	**(0.22-0.63)**			**0.31**	**(0.19-0.51)**		

Table 5.5d (continued)

Characteristics	Full model				Model 1		Model 2		Model 3	
	Unadjusted model		Adjusted model [1]		Adjusted for *Predisposing* factors only		Adjusted for *Enabling* factors only		Adjusted for *Need* factors only	
	uOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)	aOR	(95% CI)
Payer type for infancy [5]										
Public payer only	1.28	(0.90-1.80)	0.92	(0.59-1.45)			1.34	(0.92-1.96)		
Private payer only	1.00	(1.00)	1.00	(1.00)			1.00	(1.00)		
Self or uninsured	1.42	(0.33-6.07)	0.90	(0.18-4.48)			1.72	(0.38-7.68)		
Multiple payers	0.75	(0.43-1.31)	**0.51**	**(0.27-0.97)**			0.728	(0.41-1.31)		
Need characteristics										
Preterm birth (< 37 weeks)										
No	1.00	(1.00)	1.00	(1.00)					1.00	(1.00)
Yes	**1.63**	**(1.13-2.37)**	**1.88**	**(1.22-2.89)**					**1.64**	**(1.12-2.41)**
Death[6]										
No death	1.00	(1.00)	1.00	(1.00)					1.00	(1.00)
Death during infancy	0.99	(0.52-1.87)	0.92	(0.43-1.99)					0.97	(0.49-1.91)
Death in ages 1-4 years	0.93	(0.28-3.07)	1.53	(0.27-8.70)					0.96	(0.28-3.32)

uOR=odds ratio, aOR=adjusted odds ratio. 95% CI= 95% confidence interval. Values in bold are statistically significant.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Adjusted for all covariates in the model.
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Payer is expected payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[6] All deaths occurred during the study period, prior to December 31, 2008.

Table 5.5e Unadjusted and adjusted odds ratio estimates and 95% confidence intervals for the association between selected characteristics and average one-way travel time to access hospitalizations during infancy for Florida-born children with spina bifida, 1998-2007 with and without hydrocephalus [>30 minutes vs. ≤ 30 minutes (reference)]

Characteristics	All infants (n=612)				Infants with SB *without* HC (n=265)				Infants with SB *with* HC (n=347)			
	Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Exposures of interest												
Hydrocephalus												
No	1.00	(1.00)	1.00	(1.00)								
Yes	**0.71**	**(0.51-0.98)**	0.78	(0.54-1.13)								
Spina Bifida [2]												
Isolated	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Non-isolated	**0.58**	**(0.40-0.83)**	**0.58**	**(0.38-0.89)**	0.61	(0.35-1.07)	0.53	(0.27-1.04)	**0.55**	**(0.34-0.89)**	0.64	(0.36-1.15)
Predisposing characteristics												
Maternal age (in years)												
<25	1.08	(0.78-1.51)	1.12	(0.67-1.86)	0.89	(0.51-1.51)	0.73	(0.32-1.65)	1.33	(0.87-2.04)	1.73	(0.84-3.53)
25-29	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
≤30	0.96	(0.69-1.34)	0.80	(0.51-1.27)	1.14	(0.69-1.89)	0.76	(0.38-1.54)	0.81	(0.52-1.26)	0.93	(0.49-1.77)
Maternal race/ethnicity												
Non-Hispanic White	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Hispanic	**2.84**	**(1.89-4.28)**	**2.32**	**(1.31-4.10)**	**2.48**	**(1.35-4.55)**	**2.32**	**(1.01-5.34)**	**3.12**	**(1.79-5.41)**	**2.57**	**(1.13-5.86)**
Non-Hispanic Black	**2.99**	**(1.93-4.63)**	**2.50**	**(1.49-4.18)**	**2.86**	**(1.42-5.73)**	**3.13**	**(1.36-7.22)**	**3.13**	**(1.77-5.52)**	**2.14**	**(1.06-4.30)**
Other	2.94	(0.75-11.6)	1.67	(0.38-7.44)	0	(0)	0	(0)	2.50	(0.58-10.8)	1.22	(0.24-6.14)
Maternal nativity												
Born in U.S.	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Foreign-born	**2.23**	**(1.50-3.33)**	1.74	(0.99-3.05)	**2.01**	**(1.09-3.69)**	1.21	(0.60-3.33)	**2.41**	**(1.42-4.09)**	2.08	(0.95-4.52)

Table 5.5e (continued)

Characteristics	All infants				Infants with SB *without* HC				Infants with SB *with* HC			
	Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Maternal marital status												
Married	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Not married	**1.58**	**(1.13-2.20)**	1.28	(0.81-2.03)	**1.41**	**(0.84-2.35)**	0.95	(0.46-1.97)	**1.75**	**(1.13-2.70)**	1.67	(0.88-3.15)
Maternal parity												
First child	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Second or subsequent child	1.20	(0.86-1.66)	1.22	(0.82-1.80)	0.90	(0.54-1.50)	0.77	(0.42-1.43)	1.46	(0.95-2.24)	1.78	(1.03-3.09)
Maternal education												
High school diploma or more	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
< High school diploma	0.94	(0.64-1.37)	**0.61**	**(0.37-0.99)**	1.23	(0.64-2.35)	0.97	(0.42-2.21)	0.85	(0.53-1.38)	**0.42**	**(0.22-0.83)**
Sex of child												
Female	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Male	0.91	(0.66-1.26)	0.94	(0.65-1.35)	0.82	(0.50-1.34)	0.77	(0.44-1.35)	1.01	(0.66-1.54)	1.04	(0.63-1.72)
Enabling characteristics												
Adequate prenatal care [3]												
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	0.94	(0.64-1.40)	0.87	(0.55-1.37)	0.97	(0.53-1.79)	0.92	(0.43-1.95)	0.92	(0.55-1.53)	0.81	(0.44-1.48)
Residential rurality [4]												
Urban /urban cluster	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Rural	**0.31**	**(0.19-0.51)**	**0.37**	**(0.22-0.63)**	**0.43**	**(0.22-0.86)**	**0.51**	**(0.24-1.11)**	**0.22**	**(0.11-0.45)**	**0.28**	**(0.13-0.62)**
Payer type for infancy [5]												
Public payer only	1.28	(0.90-1.80)	0.92	(0.59-1.45)	1.57	(0.92-2.67)	1.29	(0.64-2.60)	1.17	(0.74-1.86)	0.72	(0.38-1.34)
Private payer only	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Self or uninsured	1.42	(0.33-6.07)	0.90	(0.18-4.48)	1.09	(0.18-6.78)	0.71	(0.09-5.42)	1.97	(0.17-22.3)	1.88	(0.11-33.0)
Multiple payers	0.75	(0.43-1.31)	0.51	(0.27-0.97)	0.47	(0.19-1.17)	0.31	(0.10-0.92)	1.03	(0.51-2.10)	0.68	(0.29-1.60)

Table 5.5e (continued)

Characteristics	All infants				Infants with SB *without* HC				Infants with SB *with* HC			
	Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]		Unadjusted model		Adjusted model [1]	
	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)	uOR	(95% CI)	aOR	(95% CI)
Need characteristics												
Preterm birth (< 37 weeks)												
No	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Yes	**1.63**	**(1.13-2.37)**	**1.88**	**(1.22-2.89)**	1.74	(0.93-3.27)	2.07	(0.97-4.40)	1.68	**(1.05-2.69)**	**2.00**	**(1.14-3.51)**
Death [6]												
No death	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)	1.00	(1.00)
Death during infancy	0.99	(0.52-1.87)	0.92	(0.43-1.99)	1.40	(0.55-3.56)	1.14	(0.34-3.83)	0.64	(0.25-1.62)	0.73	(0.25-2.10)
Death in ages 1-4 years	0.93	(0.28-3.07)	1.53	(0.27-8.70)	1.28	(0.11-14.2)	0.74	(0.05-10.7)	0.89	(0.22-3.63)	3.29	(0.30-36.0)

uOR=odds ratio, aOR=adjusted odds ratio. 95% CI= 95% confidence interval. Values in bold are statistically significant.
Note: Presence of hydrocephalus and major birth defects used to identify non-isolated SB were based on coded data and not clinically verified.
[1] Adjusted for all covariates in the model.
[2] Isolated spina bifida is SB with no additional major defects, other than the sequence of defects related to SB.
[3] Adequacy of prenatal care is determined using the Kotelchuck Index. Based on Kotelchuck scoring, adequate and adequate plus are considered "adequate prenatal care"; inadequate and intermediate care were considered inadequate.
[4] Rural residence is identified using geocoded maternal residence and 2000 U.S. Census data.
[5] Payer is expected payer. Public insurance included Medicare, Medicaid, KidCare, and Veterans Administration insurance. Private included employer-based insurance, including military coverage (CHAMPUS/TriCare). Self or under-insured defined as no third party coverage or less than 30% estimated insurance coverage. Multiple payer type means child had different types of health care payer sources over multiple hospitalizations.
[6] Deaths were those that occurred during the study period, prior to December 31, 2008.

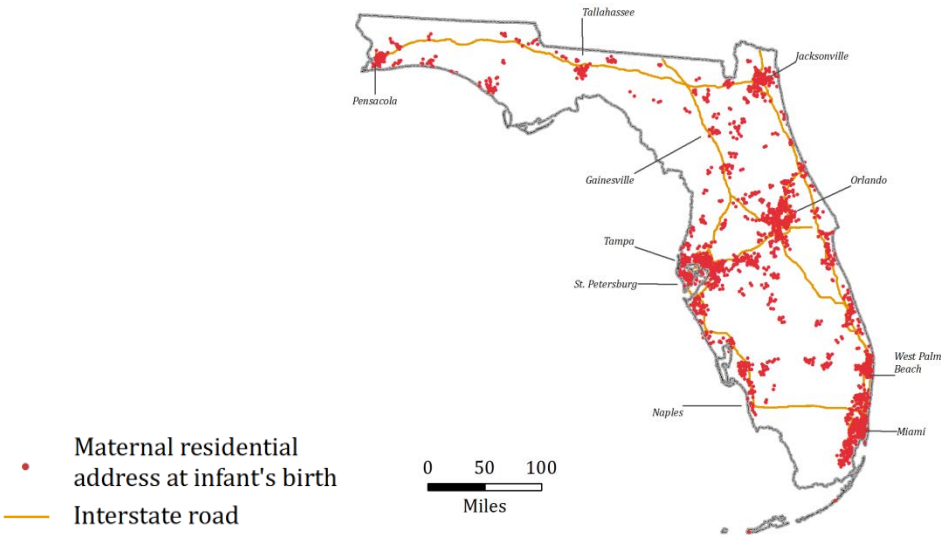

Figure 5.1 Map of geomasked maternal residential addresses at delivery for Florida-resident infants with spina bifida, 1998-2007 (n=612)

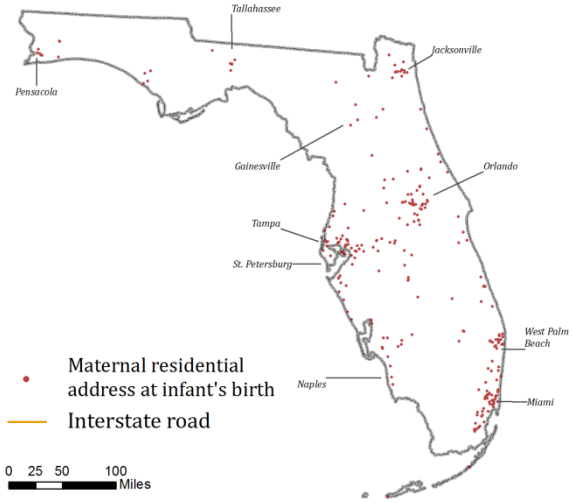

Figure 5.2 Map of geomasked maternal residential addresses at delivery for Florida-resident children with spina bifida, ages one to four years, 1998-2007 (n=251)

Data sources: Agency for Health Care Administration, 1998-2008; Florida Birth Defects Registry, 1998-2007; U.S. Census Bureau, 2000 (www.census.gov); Florida Department of Transportation, 2007

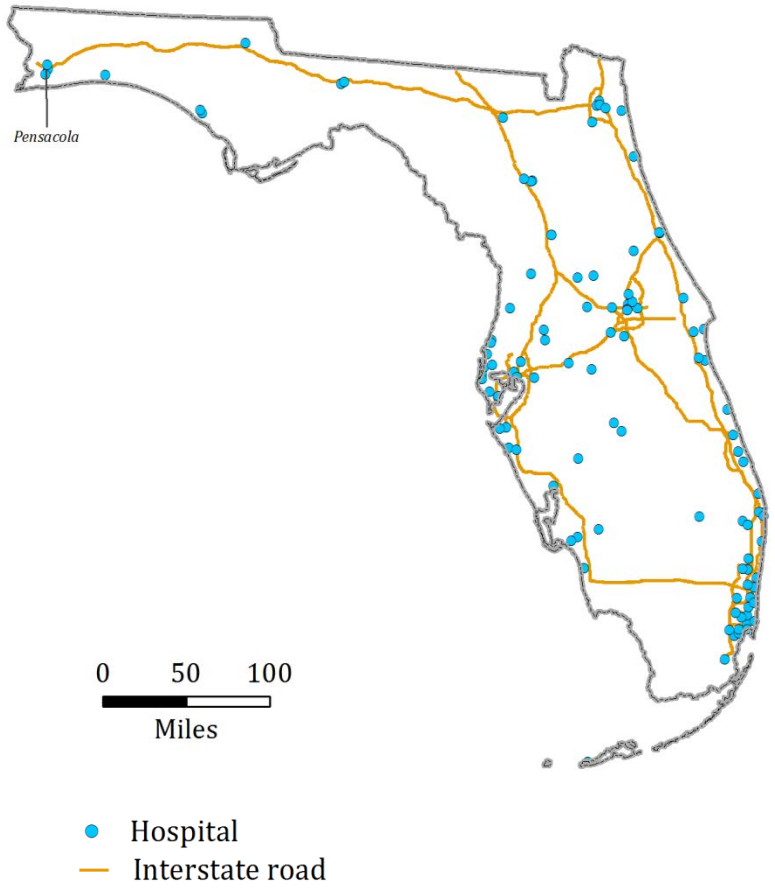

Figure 5.3 Map of Florida hospitals where Florida-resident children with spina bifida were hospitalized, 1998-2008 (n=108 hospitals)

Data sources: Agency for Health Care Administration, 1998-2008; Florida Birth Defects Registry, 1998-2007; U.S. Census Bureau, 2000 (www.census.gov); Florida Department of Transportation, 2007

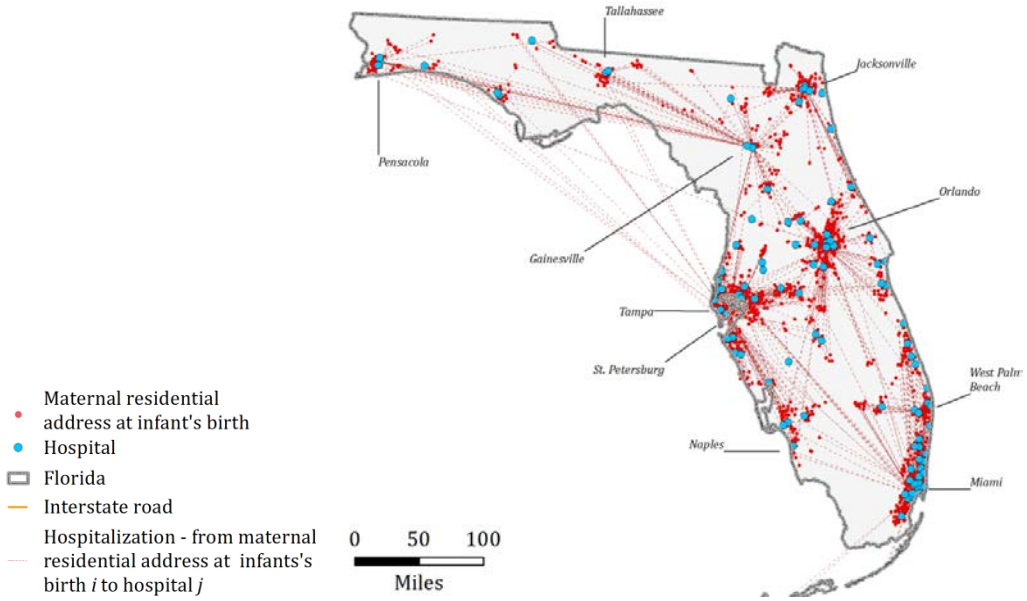

Figure 5.4 Map of travel patterns to access hospital care for Florida-resident infants with spina bifida, 1998-2007 (n=612)

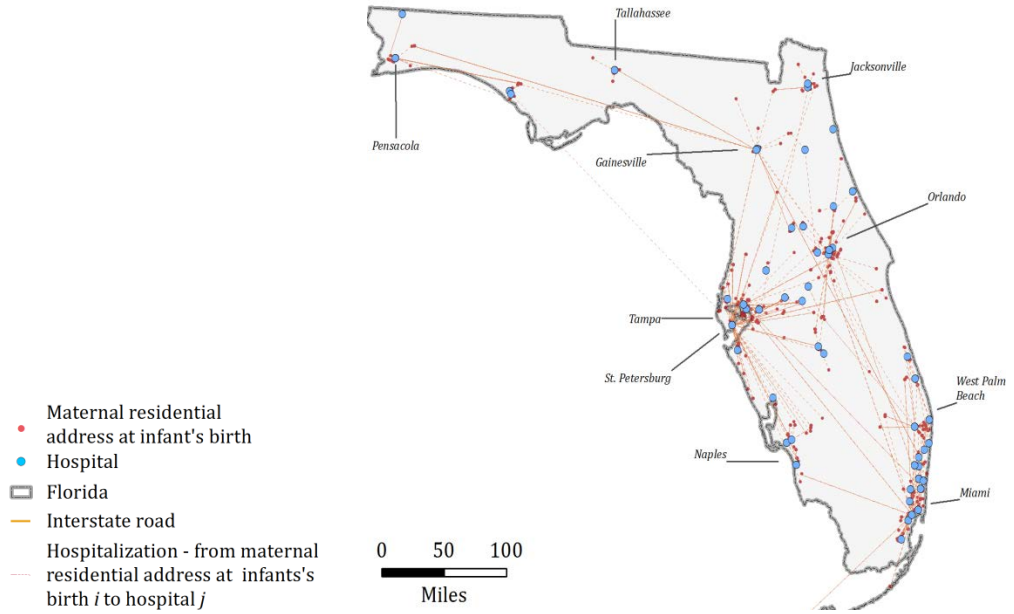

Figure 5.5 Map of travel patterns to access hospital care for Florida-resident children with spina bifida, ages one to four, 1998-2007 (n=251)

Data sources: Agency for Health Care Administration, 1998-2008; Florida Birth Defects Registry, 1998-2007; U.S. Census Bureau, 2000 (www.census.gov); Florida Department of Transportation, 2007

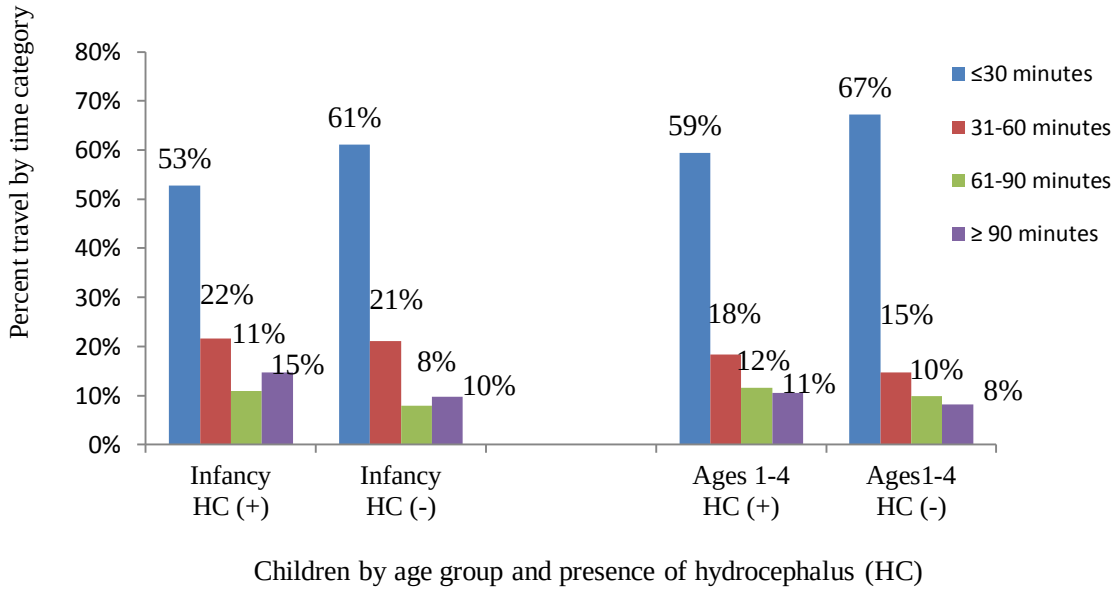

Figure 5.6: Average one-way travel time to access hospital care for Florida-born children with spina bifida, birth to 4 years, 1998-2007

REFERENCES

1. Parker, S.E., et al., *Updated national birth prevalence estimates for selected birth defects in the United States, 2004-2006.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2010. 88(12): p. 1008-1016.

2. Centers for Disease Control and Prevention, *Hospital stays, hospital charges, and in-hospital deaths among infants with selected birth defects-United States, 2003.* Morbidity and Mortality Weekly Report, 2007. 56(02): p. 25-29.

3. Centers for Disease Control and Prevention, *Update on overall prevalence of major birth defects,1978-2005.* Morbidity and Mortality Weekly Report 2008. 57(01**)**: p. 1-5.

4. WHO. *Provisional agenda item 11.7: Birth Defects*. 2010 [cited 2013 January 13]; Available from: http://apps.who.int/gb/ebwha/pdf_files/WHA63/A63_10-en.pdf.

5. Centers for Disease Control and Prevention. *Facts on birth defects*. 2011 [cited 2013 January 13]; Available from: http://www.cdc.gov/ncbddd/birthdefects/facts.html.

6. Yoon, P.W., et al., *Contribution of birth defects and genetic diseases to pediatric hospitalizations. A population-based study.* Archives of Pediatrics & Adolescent Medicine, 1997. 151(11): p. 1096-103.

7. Centers for Disease Control and Prevention, *Economic costs of birth defects and cerebral palsy-United States, 1992.* Morbidity and Mortality Weekly Report, 1995. 44(37): p. 694-699.

8. Russo, C.A. and A. Elixhauser, *Hospitalizations for birth defects, 2004*, in *Healthcare Cost and Utilization Project (HCUP) Statistical Brief #24.* 2007, U.S. Agency for Healthcare Research and Quality: Rockville, MD.

9. Martin, J.A., et al., *Annual summary of vital statistics: 2006.* Pediatrics, 2008. 121(4): p. 788-801.

10. Greene, N.D. and A.J. Copp, *The Embryonic Basis of Neural Tube Defect*, in *Neural Tube Defects: From Origin to Treatment*, D. Wyszynski, Editor 2006, Oxford University Press: New York. p. 15-28.

11. Moore, C., *Classification of Neural Tube Defects*, in *Neural Tube Defects: from Origin to Treatment*, D. Wyszynski, Editor 2006, Oxford University Pres: New York. p. 66-75.

12. Cassell, C.H., et al., *Health care expenditures among children with and those without spina bifida enrolled in Medicaid in North Carolina.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2011. 91(12): p. 1019-27.

13. Newacheck, P.W. and S.E. Kim, *A national profile of health care utilization and expenditures for children with special health care needs.* Archives of Pediatrics and Adolescent Medicine, 2005. 159(1): p. 10-7.

14. Ouyang, L.J., et al., *Health care expenditures of children and adults with spina bifida in a privately insured U.S. population.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2007. 79(7): p. 552-8.

15. Waitzman, N., R.M. Scheffler, and P.S. Romano, *The cost of birth defects: estimates of the value of prevention* 1996, Lanham, Md.: University Press of America. xiv, 262 p.

16. Tarcan, T., et al., *The timing of primary neurosurgical repair significantly affects neurogenic bladder prognosis in children with myelomeningocele.* Journal of Urology, 2006. 176(3): p. 1161-1165.

17. Valderas, J.M., et al., *Defining comorbidity: implications for understanding health and health services.* Annals of Family Medicine, 2009. **7**(4): p. 357-63.

18. Gamache, F.W., Jr., *Treatment of hydrocephalus in patients with meningomyelocele or encephalocele: a recent series.* Child's Nervous System, 1995. 11(8): p. 487-488.

19. Pinto, F.C., et al., *Surgical treatment of myelomeningocele carried out at 'time zero' immediately after birth.* Pediatric Neurosurgery, 2009. 45(2): p. 114-118.

20. Steinbok, P., et al., *Long-term outcome and complications of children born with meningomyelocele.* Child's Nervous System, 1992. **8**(2): p. 92-96.

21. Worley, G., J.M. Schuster, and W.J. Oakes, *Survival at 5 years of a cohort of newborn infants with myelomeningocele.* Developmental Medicine & Child Neurology, 1996. 38(9): p. 816-22.

22. United States Census Bureau. *United States Census 2000: Resident Population by State*. 2000 [cited 2012 November 26]; Available from: http://www.census.gov/population/www/cen2000/maps/respop.html.

23. Perry, M.J. and P.J. Makun. *Census 2000 Brief: Population change and distribution 1990-2000*. 2001 [cited 2012 November 26]; Available from: http://www.census.gov/prod/2001pubs/c2kbr01-2.pdf.

24. Martin, J.A., et al. *Births: Final Data for 2000, Volume 50(5)*. 2002 [cited 2012 November 26]; Available from: http://www.cdc.gov/nchs/data/nvsr/nvsr50/nvsr50_05.pdf.

25. Florida Agency for Healthcare Administration. *Florida Agency for Healthcare Administration*. 2010 [cited 2010 November 22]; Available from: http://ahca.myflorida.com/.

26. Salemi, J.L., et al., *Developing a database management system to support birth defects surveillance in Florida.* Journal of Registry Management, 2010. 37(1): p. 10-15; quiz 38-39.

27. Block, S., S.M. Watkins, and J.A. Correia. *Report on Birth Defects in Florida, 1998-2007*. Florida Department of Health and Florida Birth Defects Registry Report 2011 [cited 2014 January 20]; 1-39]. Available from: http://www.floridahealth.gov/alternatesites/fbdr/pdf/FBDR_report_May2011.pdf.

28. Salemi, J.L., et al., *The relative contribution of data sources to a birth defects registry utilizing passive multisource ascertainment methods: does a smaller birth defects case ascertainment net lead to overall or disproportionate loss?* Journal of Registry Management, 2011. 38(1): p. 30-8.

29. Salemi, J.L., et al., *A comparison of two surveillance strategies for selected birth defects in Florida.* Public Health Reports, 2012. 127(4): p. 391-400.

30. Salemi, J.L., et al., *Creation and Evaluation of a Multi-layered Maternal and Child Health Database for Comparative Effectiveness Research.* Journal of Registry Management, 2013. 40(1): p. 14-28.

31. U.S. Department of Health and Human Services. *Healthy People 2020 Topics and Objectives: Maternal, Infant, and Child Health.* 2012 September 7, 2012]; Available from: http://www.healthypeople.gov/2020/topicsobjectives2020/objectiveslist.aspx?topicid=26.

32. Spina Bifida Association (SBA). *Advocacy Priorities: 2012 Policy Agenda, 112th Congress, 2nd Session*. 2012 [cited 2012 October 1]; Available from: http://www.spinabifidaassociation.org/atf/cf/%7B43f44563-b00d-445d-ab6e-c3a49979b5ad%7D/SBA-2012-PACKET-HEALTH-POLICY-AGENDA.PDF.

33. Oster, M.E., et al., *Public health science agenda for congenital heart defects: report from a centers for disease control and prevention experts meeting.* Journal of the American Heart Association, 2013. 2(5): p. e000256.

34. Rasmussen, S.A., et al., *Setting a public health research agenda for Down syndrome: summary of a meeting sponsored by the Centers for Disease Control*

and Prevention and the National Down Syndrome Society. American Journal of Medical Genetics Part A, 2008. 146A(23): p. 2998-3010.

35. Rasmussen, S.A., et al., *Priorities for public health research on craniosynostosis: summary and recommendations from a Centers for Disease Control and Prevention-sponsored meeting.* American Journal of Medical Genetics Part A, 2008. 146A(2): p. 149-158.

36. Yazdy, M.M., et al., *Priorities for future public health research in orofacial clefts.* Cleft Palate-Craniofacial Journal, 2007. 44(4): p. 351-7.

37. Williams, M.A., et al., *Priorities for hydrocephalus research: report from a National Institutes of Health-sponsored workshop.* Journal of Neurosurgery, 2007. **107**(5 Suppl): p. 345-57.

38. Centers for Disease Control and Prevention. *Birth Defects: Facts about Down Syndrome*. 2011 [cited 2013 January 13]; Available from: http://www.cdc.gov/ncbddd/birthdefects/DownSyndrome.html.

39. Centers for Disease Control and Prevention. *Guidance for Preventing Birth Defects*. 2011 [cited 2013 January 13]; Available from: http://www.cdc.gov/ncbddd/birthdefects/prevention.html.

40. Kirby, R.S., *Methodological Issues in the Study of Neural Tube Defects*, in *Neural Tube Defects: From Origin to Treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 103-108.

41. Hunter, A.G.W., *Brain and Spinal Cord*, in *Human malformations and related anomalies*, R.E. Stevenson, J.G. Hall, and R.M. Goodman, Editors. 1993, Oxford University Press: New York. p. 109-137.

42. Agopian, A.J., et al., *Spina bifida subtypes and sub-phenotypes by maternal race/ethnicity in the National Birth Defects Prevention Study.* American Journal of Medical Genetics Part A, 2012. 158A(1): p. 109-15.

43. March of Dimes. *Birth Defects: Spina Bifida.* 2009 [cited 2012 October 1]; Available from: http://www.marchofdimes.com/baby/birthdefects_spinabifida.html.

44. Joseph, D.B., *Urological Care and Management of Patients with Neural Tube Defects*, in *Neural Tube Defects: From Origin to Treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 261-270.

45. Shin, M., L.M. Besser, and A. Correa, *Prevalence of Spina Bifida among Children and Adolescents in Metropolitan Atlanta.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2008. ***82***(11): p. 748-754.

46. Liptak, G.S. and A. El Samra, *Optimizing health care for children with spina bifida.* Developmental Disabilities Research Reviews, 2010. 16(1): p. 66-75.

47. Liptak, G.S., *Cognition, Education, and Learning Disorders,* in *Neural Tube Defects: From Origin to Treatment,* D.F. Wyszynsk, Editor 2006, Oxford University Press: New York. p. 271-282.

48. Williams, L.J., et al., *Decline in the prevalence of spina bifida and anencephaly by race/ethnicity: 1995-2002.* Pediatrics, 2005. 116(3): p. 580-586.

49. Sutton, L.N., *Fetal surgery for neural tube defects.* Best Practice & Research in Clinical Obstetrics & Gynaecology, 2008. 22(1): p. 175-188.

50. Boulet, S.L., et al., *Trends in the postfortification prevalence of spina bifida and anencephaly in the United States.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2008. 82(7): p. 527-532.

51. Canfield, M.A., et al., *Changes in the birth prevalence of selected birth defects after grain fortification with folic acid in the United States: findings from a multi-state population-based study.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2005. 73(10): p. 679-89.

52. Honein, M.A., et al., *Impact of folic acid fortification of the US food supply on the occurrence of neural tube defects.* Journal of the American Medical Association, 2001. 285(23): p. 2981-2986.

53. Williams, L.J., et al., *Prevalence of spina bifida and anencephaly during the transition to mandatory folic acid fortification in the United States.* Teratology, 2002. 66(1): p. 33-9.

54. Boulet, S.L., et al., *Racial/ethnic differences in the birth prevalence of spina bifida - United States, 1995-2005.* CDC Morbidity and Mortality Weekly Report, 2009. 57(53): p. 1409-1413.

55. Shin, M., et al., *Prevalence of spina bifida among children and adolescents in 10 regions in the United States.* Pediatrics, 2010. 126(2): p. 274-279.

56. Bennett, K.A., et al., *Fetal Surgery for Myelomeningoele,* in *Neural Tube Defects: From Origin to Treatment,* D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 217-230.

57. Hunt, G.M., *The median survival time in open spina bifida.* Developmental Medicine & Child Neurology, 1997. 39(8): p. 568.

58. Wong, L.Y. and L.J. Paulozzi, *Survival of infants with spina bifida: a population study, 1979-94.* Paediatric and Perinatal Epidemiology, 2001. 15(4): p. 374-8.

59. Shin, M., et al., *Improved survival among children with spina bifida in the United States.* Journal of Pediatrics, 2012. 161(6): p. 1132-1137 e3.

60. McClugage, S.G., et al., *The history of the surgical repair of spina bifida.* Child's Nervous System, 2012. 28(10): p. 1693-1700.

61. Bowman, R.M. and D.G. McLone, *Neurological Management of Spina Bifida: Research Issues.* Developmental Disabilities Research Reviews, 2010. 16(1): p. 82-87.

62. Singh, D.K., *Families of children with spina bifida: A review.* Journal of Developmental and Physical Disabilities, 2003. 15(1): p. 37-55.

63. Rob de Jong, T.H., *Deliberate termination of life of newborns with spina bifida, a critical reappraisal.* Child's Nervous System, 2008. 24(1): p. 13-28.

64. Schoenmakers, M., et al., *Determinants of functional independence and quality of life in children with spina bifida.* Clinical Rehabilitation, 2005. 19(6): p. 677-685.

65. Merkens, M.J. and Spina Bifida Association, *Guidelines for Spina Bifida Health Care Services Throughout the Lifespan (Third Edition).* Spina Bifida Association, 2006.

66. Miller, J.P. and A.R. Cohen, *Postnatal Surgery for Myelomeningocele,* in *Neural Tube Defects: From Origin to Treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 231-240.

67. National Institute of Neurological Disorders and Stroke and National Institutes of Health. *Spina Bifida.* 2013 [cited 2013 October 15]; Available from: http://www.ninds.nih.gov/disorders/spina_bifida/detail_spina_bifida.htm#241483258.

68. Adzick, N.S., et al., *A randomized trial of prenatal versus postnatal repair of myelomeningocele.* New England Journal of Medicine, 2011. 364(11): p. 993-1004.

69. Aaronson, O.S., et al., *Robot-assisted endoscopic intrauterine myelomeningocele repair: a feasibility study.* Pediatric Neurosurgery, 2002. 36(2): p. 85-9.

70. Adzick, N.S., *Fetal myelomeningocele: natural history, pathophysiology, and in-utero intervention.* Seminars in Fetal & Neonatal Medicine, 2010. 15(1): p. 9-14.

71. Bruner, J.P., et al., *Fetal surgery for myelomeningocele and the incidence of shunt-dependent hydrocephalus.* Journal of the American Medical Association, 1999. 282(19): p. 1819-25.

72. Tulipan, N. and J.P. Bruner, *Myelomeningocele repair in utero: a report of three cases.* Pediatric Neurosurgery, 1998. 28(4): p. 177-80.

73. Tulipan, N., et al., *Intrauterine myelomeningocele repair reverses preexisting hindbrain herniation.* Pediatric Neurosurgery, 1999. 31(3): p. 137-42.

74. Management of Myelomeningocele Study. *Management of Myelomeningocele Study (MOMS)*. 2010 [cited 2010 November 21]; Available from: http://www.spinabifidamoms.com/english/overview.html.

75. Blount, J., et al. *A Randomized Trial of Pre-natal versus Post-natal Repair of Myelomeningocele: Summary and Commentary*. 2011 [cited 2013 November 1]; Available from: http://www.spinabifidaassociation.org/atf/cf/%7Beed435c8-f1a0-4a16-b4d8-a713bbcd9ce4%7D/FINAL%20NEJM_MOMS_REVIEW.PDF.

76. Williams, H., *Questioning the rationale and conduct of the management of myelomeningocele study.* Medical Hypotheses, 2011. 77(1): p. 101-5.

77. van Lith, J.M., M.P. Johnson, and R.D. Wilson, *Current controversies in prenatal diagnosis 3: fetal surgery after MOMS: is fetal therapy better than neonatal?* Prenatal Diagnosis, 2013. 33(1): p. 13-6.

78. Correa, A., et al., *Diabetes mellitus and birth defects.* American Journal of Obstetrics and Gynecology, 2008. 199(3): p. 237 e1-9.

79. Lisi, A., et al., *Surveillance of adverse fetal effects of medications (SAFE-Med): findings from the International Clearinghouse of Birth Defects Surveillance and Research.* Reproductive Toxicology, 2010. 29(4): p. 433-42.

80. Rasmussen, S.A., et al., *Guidelines for case classification for the National Birth Defects Prevention Study.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2003. 67(3): p. 193-201.

81. Seaver, L.H. and R.E. Stevenson, *Syndromes with Neural Tube Defects*, in *Neural Tube Defects: From Origin to Treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 76-83.

82. Stevenson, R.E., et al., *Neural tube defects and associated anomalies in South Carolina.* Birth Defects Research Part A: Clinical and Molecular Teratolology, 2004. 70(9): p. 554-8.

83. National Birth Defects Prevention Network Inc. (NBDPN). *Guidelines for Conducting Birth Defects Surveillance*. 2004 [cited 2013 December 20]; Available from: http://www.nbdpn.org/current/resources/bdsurveillance.html.

84. Jeng, S., et al., *Prevalence of congenital hydrocephalus in California, 1991-2000.* Pediatric Neurology, 2011. 45(2): p. 67-71.

85. Cate, I.M., C. Kennedy, and J. Stevenson, *Disability and quality of life in spina bifida and hydrocephalus.* Developmental Medicine & Child Neurology, 2002. 44(5): p. 317-22.

86. Hampton, L.E., et al., *Hydrocephalus status in spina bifida: an evaluation of variations in neuropsychological outcomes.* Journal of Neurosurgery: Pediatrics, 2011. **8**(3): p. 289-98.

87. Hetherington, R., et al., *Functional outcome in young adults with spina bifida and hydrocephalus.* Child's Nervous System, 2006. 22(2): p. 117-24.

88. Vinck, A., et al., *Arnold-Chiari-II malformation and cognitive functioning in spina bifida.* Journal of Neurology, Neurosurgery, and Psychiatry, 2006. 77(9): p. 1083-6.

89. Yeates, K.O., et al., *Do children with myelomeningocele and hydrocephalus display nonverbal learning disabilities? An empirical approach to classification.* Journal of the International Neuropsychological Society, 2003. 9(4): p. 653-62.

90. Burmeister, R., et al., *Attention problems and executive functions in children with spina bifida and hydrocephalus.* Child Neuropsychology, 2005. 11(3): p. 265-83.

91. *Evidence-based Practice in Spina Bifida: Developing a Research Agenda*, G.S. Liptak, Editor 2003, Convened meeting May 9-10, 2003: Agency for Healthcare Research and Quality, Centers for Disease Control and Prevention, National Institutes of Health, Spina Bifida Association of America: Washington DC.

92. Simon, T.D., et al., *Hospital care for children with hydrocephalus in the United States: utilization, charges, comorbidities, and deaths.* Journal of Neurosurgery: Pediatrics, 2008. 1(2): p. 131-7.

93. Frimberger, D., E. Cheng, and B.P. Kropp, *The current management of the neurogenic bladder in children with spina bifida.* Pediatric Clinics of North America, 2012. 59(4): p. 757-767.

94. Simeonsson, R.J., J.S. McMillen, and G.S. Huntington, *Secondary conditions in children with disabilities: spina bifida as a case example.* Mental Retardation and Developmental Disabilities Research Reviews, 2002. 8(3): p. 198-205.

95. Burke, R. and G.S. Liptak, *Providing a primary care medical home for children and youth with spina bifida.* Pediatrics, 2011. 128(6): p. e1645-57.

96. Bier, J.A.B., et al., *Medical, functional, and social determinants of health-related quality of life in individuals with myelomeningocele.* Developmental Medicine and Child Neurology, 2005. 47(9): p. 609-612.

97. Stevenson, J. and I.M.P. Cate, *Assessment of Health-related Quality of Life in Individuals with Neural Tube Defects*, in *Neural Tube Defects: From Origin to Treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York. p. 361-370.

98. Soe, M.M., et al., *Health risk behaviors among young adults with spina bifida.* Developmental Medicine & Child Neurology, 2012. 54(11): p. 1057-64.

99. Newacheck, P.W., *Characteristics of children with high and low usage of physician services.* Medical Care, 1992. 30(1): p. 30-42.

100. Aday, L.A., et al., *Health insurance and utilization of medical care for children with special health care needs.* Medical Care, 1993. 31(11): p. 1013-26.

101. Shannon, C.N., et al., *The economic impact of ventriculoperitoneal shunt failure.* Journal of Neurosurgery: Pediatrics, 2011. 8(6): p. 593-9.

102. Institute of Medicine (U.S.). Committee on Quality of Health Care in America., *Crossing the quality chasm: a new health system for the 21st century* 2001, Washington, D.C.: National Academy Press. 337 pp.

103. Cassell, C.H., J. Daniels, and R.E. Meyer, *Timeliness of primary cleft lip/palate surgery.* Cleft Palate-Craniofacial Journal, 2009. 46(6): p. 588-97.

104. Dawson, A.L., et al., *Factors associated with late detection of critical congenital heart disease in newborns.* Pediatrics, 2013. 132(3): p. e604-11.

105. Peterson, C., et al., *Hospitalizations, costs, and mortality among infants with critical congenital heart disease: How important is timely detection?* Birth Defects Research Part A: Clinical and Molecular Teratology, 2013.

106. Cassell, C. and R. Meyer, *Timeliness of services during the first two years of life among Medicaid-eligible children with orofacial clefts in North Carolina, 1995-2002*, 2008, State Center for Health Statistics: Raleigh.

107. McLone, D.G., *Results of treatment of children born with a myelomeningocele.* Clinical Neurosurgery, 1983. 30: p. 407-12.

108. Newacheck, P.W., et al., *Health insurance and access to primary care for children.* New England Journal of Medicine, 1998. 338(8): p. 513-9.

109. Newacheck, P.W., M. Inkelas, and S.E. Kim, *Health services use and health care expenditures for children with disabilities.* Pediatrics, 2004. 114(1): p. 79-85.

110. Radcliff, E., et al., *Hospital use, associated costs, and payer status for infants born with spina bifida.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2012. 94(12): p. 1044-53.

111. Houtrow, A.J., J.H. Maselli, and M.J. Okumura, *Inpatient care for children, ages 1-20 years, with spina bifida in the United States.* Journal of Pediatric Rehabilitation Medicine, 2013. 6(2): p. 95-101.

112. Folland, S., A.C. Goodman, and M. Stano, *The economics of health and health care*. 6th ed 2010, Boston: Prentice Hall. xxii, 601 p.

113. Agency for Healthcare Research and Quality (AHRQ). *MEPS Topics: Health Care Costs and Expenditures*. 2009 [cited 2012 February 20]; Available from: http://meps.ahrq.gov/mepsweb/data_stats/MEPS_topics.jsp?topicid55Z-1.

114. NIH. *Health economics information resources. U.S. National Library of Medicine: National Institutes of Health.* 2010 [cited 2012 February 20]; Available from: http://www.nlm.nih.gov/nichsr/edu/healthecon/glossary.html.

115. Wyszynski, D.F., P.S. Romano, and S.D. Grosse, *The Half-Life of Cost-of-Illness Estimates: The Case of Spina Bifida*, in *Neural tube defects: from origin to treatment*, D.F. Wyszynski, Editor 2006, Oxford University Press: New York, NY. p. xvii, 399 p.

116. Grosse, S.D., et al., *Reevaluating the benefits of folic acid fortification in the United States: economic analysis, regulation, and public health.* American Journal of Public Health, 2005. 95(11): p. 1917-22.

117. Grosse, S.D., *Lifetime direct costs for a child with spina bifida (unpublished data)*, 2011, Centers for Disease Control and Prevention, National Center on Birth Defects and Developmental Disabilities: Atlanta.

118. American College of Physicians-American Society of Internal Medicine ACP-ASIM. *White Paper: No Health Insurance? It's Enough to Make You Sick: Scientific Research Linking the Lack of Health Coverage to Poor Health*. 2000 October 20, 2012]; Available from: http://www.acponline.org/pressroom/uninsured_factsheet.htm.

119. McLaughlin, C.G. and L. Wyszewianski, *Access to care: remembering old lessons.* Health Services Research, 2002. 37(6): p. 1441-3.

120. Penchansky, R., *The concept of access: a definition.* Vol. HRP-0900113. 1977, Hyattsville, MD: National Health Planning Information Center. 69 p.

121. Penchansky, R. and J.W. Thomas, *The concept of access: definition and relationship to consumer satisfaction.* Medical Care, 1981. 19(2): p. 127-40.

122. Betz, C.L., et al., *Secondary analysis of primary and preventive services accessed and perceived service barriers by children with developmental disabilities and their families.* Issues in Comprehensive Pediatric Nursing, 2004. 27(2): p. 83-106.

123. Cassell, C.H., et al., *Factors associated with distance and time traveled to cleft and craniofacial care.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2013.

124. Cassell, C.H., D.D. Mendez, and R.P. Strauss, *Maternal perspectives: qualitative responses about perceived barriers to care among children with orofacial clefts in North Carolina.* Cleft Palate-Craniofacial Journal, 2012. 49(3): p. 262-9.

125. Newacheck, P.W., D.C. Hughes, and J.J. Stoddard, *Children's access to primary care: differences by race, income, and insurance status.* Pediatrics, 1996. 97(1): p. 26-32.

126. Newacheck, P.W., Y.Y. Hung, and K.K. Wright, *Racial and ethnic disparities in access to care for children with special health care needs.* Ambulatory Pediatrics, 2002. 2(4): p. 247-54.

127. Newacheck, P.W., et al., *Access to health care for children with special health care needs.* Pediatrics, 2000. 105(4 Pt 1): p. 760-6.

128. Skinner, A.C. and R.T. Slifkin, *Rural/urban differences in barriers to and burden of care for children with special health care needs.* Journal of Rural Health, 2007. 23(2): p. 150-7.

129. Gulliford, M., et al., *What does 'access to health care' mean?* Journal of Health Services Research and Policy, 2002. 7(3): p. 186-8.

130. Silver, E.J. and R.E. Stein, *Access to care, unmet health needs, and poverty status among children with and without chronic conditions.* Ambulatory Pediatrics, 2001. 1(6): p. 314-20.

131. St Clair, P.A., et al., *Situational and financial barriers to prenatal care in a sample of low-income, inner-city women.* Public Health Reports, 1990. 105(3): p. 264-7.

132. Mikhail, B.I., *Perceived impediments to prenatal care among low-income women.* Western Journal of Nursing Research, 1999. 21(3): p. 335-50; discussion 351-5.

133. Flores, G. and S.C. Tomany-Korman, *Racial and ethnic disparities in medical and dental health, access to care, and use of services in US children.* Pediatrics, 2008. 121(2): p. e286-98.

134. Ngui, E.M. and G. Flores, *Unmet needs for specialty, dental, mental, and allied health care among children with special health care needs: are there racial/ethnic disparities?* Journal of Health Care for the Poor and Underserved, 2007. 18(4): p. 931-49.

135. Granados, G., et al., *Health care for Latino children: impact of child and parental birthplace on insurance status and access to health services.* American Journal of Public Health, 2001. 91(11): p. 1806-7.

136. Probst, J.C., et al., *Effects of residence and race on burden of travel for care: cross sectional analysis of the 2001 U.S. National Household Travel Survey.* BMC Health Services Research, 2007. **7**: p. 40.

137. Singh, G.K., et al., *Geographic disparities in access to the medical home among US CSHCN.* Pediatrics, 2009. 124 Suppl 4: p. S352-60.

138. Case, A.P., et al., *Proximity of pediatric genetic services to children with birth defects in Texas.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2008. 82(11): p. 795-8.

139. Fixler, D.E., et al., *Effect of acculturation and distance from cardiac center on congenital heart disease mortality.* Pediatrics, 2012. **129**(6): p. 1118-24.

140. Delmelle, E.M., et al., *Modeling travel impedance to medical care for children with birth defects using Geographic Information Systems.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2013. 97(10), 673-684.

141. Krieger, N., *Place, space, and health: GIS and epidemiology.* Epidemiology, 2003. 14(4): p. 384-5.

142. Aschengrau, A. and G.R. Seage, *Essentials in Epidemiology in Public Health.* 2003 Sudbury, MA: Jones and Bartlett Publishers. 468 pp.

143. Siffel, C., et al., *Role of geographic information systems in birth defects surveillance and research.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2006. 76(11): p. 825-833.

144. Kirby, R., *Incorporating Geographical Analysis into the Study of Mental Retardation and Developmental Disabilities*, in *International Review of Research in Mental Retardation*, R. Urbano and R. Hodapp, Editors. 2006, Academic Press/Elsevier Inc. p. 79-91.

145. Root, E.D., R.E. Meyer, and M.E. Emch, *Evidence of localized clustering of gastroschisis births in North Carolina, 1999-2004.* Social Science & Medicine, 2009. 68(8): p. 1361-7.

146. Wang, Y., et al., *Development of web-based geocoding applications for the population-based Birth Defects Surveillance System in New York state.* Journal of Registry Management, 2010. 37(1): p. 16-21.

147. Vinceti, M., et al., *Risk of congenital anomalies around a municipal solid waste incinerator: a GIS-based case-control study.* International Journal of Health Geographics, 2009. 8: p. 8.

148. Kuehn, C.M., et al., *Risk of malformations associated with residential proximity to hazardous waste sites in Washington State.* Environmental Research, 2007. 103(3): p. 405-12.

149. Rushton, G., et al., *The spatial relationship between infant mortality and birth defect rates in a U.S. city.* Statistics in Medicine, 1996. 15(17-18): p. 1907-19.

150. Nuckols, J.R., et al., *Linking Water Utility Data and Residences in the National Birth Defects Prevention Study* 2004. Accessed March 20, 2014 from http://www.waterrf.org/PublicReportLibrary/2832.pdf.

151. Andersen, R., T.H. Rice, and G.F. Kominski, *Changing the U.S. health care system: key issues in health services policy and management.* 3rd ed 2007, San Francisco: Jossey-Bass. xlv, 693 p.

152. Aday, L.A. and R. Andersen, *A framework for the study of access to medical care.* Health Services Research, 1974. 9(3): p. 208-20.

153. Okumura, M.J., et al., *Understanding factors associated with work loss for families caring for CSHCN.* Pediatrics, 2009. 124 Suppl 4: p. S392-8.

154. Yu, S.M. and G.K. Singh, *Household language use and health care access, unmet need, and family impact among CSHCN.* Pediatrics, 2009. 124 Suppl 4: p. S414-9.

155. Andersen, R.M., *National health surveys and the behavioral model of health services use.* Medical Care, 2008. 46(7): p. 647-53.

156. Andersen, R. and J.F. Newman, *Societal and individual determinants of medical care utilization in the United States.* Milbank Memorial Fund Quarterly: Health and Society, 1973. 51(1): p. 95-124.

157. Strickland, B.B., et al., *Access to the medical home: new findings from the 2005-2006 National Survey of Children with Special Health Care Needs.* Pediatrics, 2009. 123(6): p. e996-1004.

158. van den Akker, M., et al., *Multimorbidity in General Practice: Prevalence, Incidence, and Determinants of Co-Occurring Chronic and Recurrent Diseases.* Journal of Clinical Epidemiology, 1998. 51(5): p. 367-375.

159. Bowman, R.M., et al., *Spina bifida outcome: a 25-year prospective.* Pediatric Neurosurgery, 2001. 34(3): p. 114-20.

160. Hall, J.G., et al., *Clinical, genetic, and epidemiological factors in neural tube defects.* American Journal of Human Genetics, 1988. 43(6): p. 827-37.

161. Kallen, B., E. Robert, and J. Harris, *Associated malformations in infants and fetuses with upper and lower neural tube defects.* Teratology, 1998. 57: p. 56-63.

162. Seaver, L.H., R.E. Stevenson, and J. Dean, *Neural tube defects with associated anomalies: recognized patterns of malformation,* in *Second Biennial Neural Tube Defects Conference* 2002: Seabrook Island, SC.

163. Patwardhan, R.V. and A. Nanda, *Implanted ventricular shunts in the United States: the billion-dollar-a-year cost of hydrocephalus treatment.* Neurosurgery, 2005. 56(1): p. 139-44; discussion 144-5.

164. Caldarelli, M., C. Di Rocco, and F. La Marca, *Shunt complications in the first postoperative year in children with meningomyelocele.* Child,s Nervous System, 1996. 12(12): p. 748-54.

165. Cassedy, A., G. Fairbrother, and P.W. Newacheck, *The impact of insurance instability on children's access, utilization, and satisfaction with health care.* Ambulatory Pediatrics, 2008. 8(5): p. 321-8.

166. Connor, J.A., K. Gauvreau, and K.J. Jenkins, *Factors associated with increased resource utilization for congenital heart disease.* Pediatrics, 2005. 116(3): p. 689-95.

167. Rosenbach, M.L., C. Irvin, and R.F. Coulam, *Access for low-income children: is health insurance enough?* Pediatrics, 1999. 103(6 Pt 1): p. 1167-74.

168. Nelson, A., *Unequal treatment: confronting racial and ethnic disparities in health care.* Journal of the National Medical Association, 2002. 94(8): p. 666-8.

169. Smedley, B.D., A.Y. Stith, and A.R. Nelson, *Unequal treatment: confronting racial and ethnic disparities in health care* Institute of Medicine (U.S.). Committee on Understanding and Eliminating Racial and Ethnic Disparities in Health Care 2003, Washington, D.C.: National Academy Press. xvi, 764 p.

170. Flores, G. and S.C. Tomany-Korman, *The language spoken at home and disparities in medical and dental health, access to care, and use of services in US children.* Pediatrics, 2008. 121(6): p. e1703-14.

171. Beal, A.C., *High-quality health care: the essential route to eliminating disparities and achieving health equity.* Health Affairs (Millwood), 2011. 30(10): p. 1868-71.

172. Honein, M.A., et al., *The association between major birth defects and preterm birth.* Maternal and Child Health Journal, 2009. 13(2): p. 164-75.

173. Lorch, S.A., et al., *The differential impact of delivery hospital on the outcomes of premature infants.* Pediatrics, 2012. 130(2): p. 270-8.

174. Samuelson, J.L., et al., *Maternal characteristics associated with place of delivery and neonatal mortality rates among very-low-birthweight infants, Georgia.* Paediatric and Perinatal Epidemiology, 2002. 16(4): p. 305-13.

175. Aday, L.A. and R.M. Andersen, *Equity of access to medical care: a conceptual and empirical overview.* Medical Care, 1981. 19(12 Suppl): p. 4-27.

176. Andersen, R.M., *Revisiting the behavioral model and access to medical care: does it matter?* Journal of Health and Social Behavior, 1995. 36(1): p. 1-10.

177. Colvin, L. and C. Bower, *A retrospective population-based study of childhood hospital admissions with record linkage to a birth defects registry.* BMC Pediatrics, 2009. 9: p. 32.

178. Agency for Healthcare Research and Quality (AHRQ). *Health Care Utilization Project. Cost-to-charge ratio files.* 2012 [cited 2012 May 22]; Available from: http://www.hcup-us.ahrq.gov/db/state/costtocharge.jsp.

179. U.S. Bureau of Labor Statistics. *Producer price index industry data: hospitals. Series PCU622.* [cited 2012 May 22]; Available from: http://www.bls.gov/cpi.

180. U.S. Bureau of Labor Statistics. *United States Department of Labor: Division of Consumer Prices and Price Indexes.* Division of Consumer Prices and Price Indexes 2011 [cited 2012 November 16]; Available from: http://www.bls.gov/cpi/.

181. *The Kotelchuck Index*. 2012 [cited 2012 December 20]; Available from: http://health.utah.gov/opha/IBIShelp/kotelchuck.html.

182. Kotelchuck, M., *The Adequacy of Prenatal Care Utilization Index: its US distribution and association with low birthweight.* American Journal of Public Health, 1994. 84(9): p. 1486-9.

183. United States Census Bureau. *Geography: Census 2000 Urban and Rural Classification*. 2000 [cited 2013 September 17]; Available from: http://www.census.gov/geo/reference/ua/urban-rural-2000.html.

184. AHCA. *AHCA Florida Hospital Inpatient Discharge Data Dictionary*. 2011 [cited 2013 September 17]; Available upon request from Florida Agency for Health Care Administration:[Available from: http://www.healthdatastore.com/ahca-florida-hospital-discharge-data.aspx.

185. American Academy of Pediatrics Committee on Fetus and Newborn, *Levels of neonatal care.* Pediatrics, 2004. 114(5): p. 1341-7.

186. American Academy of Pediatrics Committee on Fetus And Newborn, *Levels of neonatal care.* Pediatrics, 2012. 130(3): p. 587-97.

187. Barfield, W.D. and The Committee on Fetus and Newborn, *Standard Terminology for Fetal, Infant, and Perinatal Deaths.* Pediatrics, 2011. 128(1): p. 177-181.

188. Manning, W.G. and J. Mullahy, *Estimating log models: to transform or not to transform?* Journal of Health Economics, 2001. 20(4): p. 461-94.

189. Allison, P., *Logistic Regression Using the SAS System: Theory and Application* 1999, Cary, NC: SAS Institute Inc.

190. *Introduction to SAS*. UCLA: Statistical Consulting Group 2013 [cited 2013 September 4]; Available from: http://www.ats.ucla.edu/stat/sas/notes2/.

191. MacDorman, M.F., T.J. Mathews, and E. Declercq, *Home births in the United States, 1990-2009*, 2012, NCHS data brief, #84, National Center for Health Statistics: Hyattsville, MD.

192. Fortin, M., et al., *Multimorbidity's many challenges.* BMJ, 2007. 334(7602): p. 1016-1017.

193. Starfield, B., *Threads and yarns: weaving the tapestry of comorbidity.* Annals of Family Medicine, 2006. 4(2): p. 101-3.

194. Farrell, K., C. Hess, and D. Justice, *The Affordable Care Act and Children with Special Health Care Needs: An Analysis and Steps for State Policymakers,* 2011, National Academy for State Health Policy for The Catalyst Center: Boston, MA.

195. Ray, K.N. and S.A. Lorch, *Hospitalization of rural and urban infants during the first year of life.* Pediatrics, 2012. 130(6): p. 1084-93.

196. Petrini, J., et al., *Contribution of birth defects to infant mortality in the United States.* Teratology, 2002. 66 Supplement 1: p. S3-6.

197. Purisch, S.E., et al., *Preterm birth in pregnancies complicated by major congenital malformations: a population-based study.* American Journal of Obstetrics and Gynecology, 2008. 199(3): p. 287 e1-8.

198. National Center on Birth Defects and Developmental Disabilities (NCBDDD). Centers for Disease Control and Prevention. *Birth defects and genetic diseases: branch 6-digit code for reportable congenital anomalies.* 2007 [cited 2014 January 27]; Available from: http://www.cdc.gov/ncbddd/birthdefects/documents/macdpcode0807.pdf.

199. National Center on Birth Defects and Developmental Disabilities (NCBDDD). Centers for Disease Control and Prevention. *Metropolitan Atlanta Congenital Defects Program (MACDP).* 2013 [cited 2014 January 27]; Available from: http://www.cdc.gov/ncbddd/birthdefects/macdp.html.

200. Holmes, L.B. and M.N. Westgate, *Using ICD-9 codes to establish prevalence of malformations in newborn infants.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2012. 94(4): p. 208-14.

201. Lary, J.M. and L.D. Edmonds, *Prevalence of spina bifida at birth--United States, 1983-1990: a comparison of two surveillance systems.* CDC MMWR Surveillance Summaries, 1996. 45(2): p. 15-26.

202. Florida Agency for Health Care Administration. *Florida Agency for Health Care Administration, Disclaimer* 2013 [cited 2013 June 3]; Available from: http://www.floridahealthfinder.gov/CompareCare/Disclaimer.aspx

203. Allison, P.D., *Missing Data,* in *Sage University Paper Series on Quantitative Application in the Social Sciences* 2001, Sage: Thousand Oaks, CA. p. 07-136.

204. Centers for Disease Control and Prevention. *Spina Bifida: About the National Spina Bifida Patient Registry* 2013 [cited 2013 August 24]; Available from: http://www.cdc.gov/ncbddd/spinabifida/nsbprregistry.html.

205. Tinker, S.C., et al., *Estimate of the potential impact of folic acid fortification of corn masa flour on the prevention of neural tube defects.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2013. 97(10): p. 649-57.

206. Strauss, R.P. and C.H. Cassell, *Critical issues in craniofacial care: quality of life, costs of care, and implications of prenatal diagnosis.* American Academy of Pediatrics, 2009. 9(6): p. 427-32.

207. Kuehl, K.S., C.A. Loffredo, and C. Ferencz, *Failure to diagnose congenital heart disease in infancy.* Pediatrics, 1999. 103(4 Pt 1): p. 743-7.

208. Centers for Disease Control and Prevention, *Economic Costs of Birth Defects and Cerebral Palsy -- United States, 1992*, in *Morbidity and Mortality Weekly Report September 22,* 1995, Centers for Disease Control and Prevention: Atlanta, Georgia. p. 694-9.

209. National Center on Birth Defects and Developmental Disabilities. Centers for Disease Control and Prevention. *Spina Bifida: Facts*. 2013 [cited 2013 January 10]; Available from: http://www.cdc.gov/ncbddd/spinabifida/facts.html.

210. Patet, J.D., C. Lapras, and J.N. Guilburd, *[Spina bifida aperta-- myelomeningocele. Hydrocephalus]*. Neurochirurgie, 1988. 34 Suppl 1: p. 47-52.

211. Stein, S.C. and L. Schut, *Hydrocephalus in myelomeningocele.* Childs Brain, 1979. **5**(4): p. 413-9.

212. Langlois, P.H., M.A. Canfield, and M.D. Swartz, *Poisson versus logistic regression in a descriptive epidemiologic analysis of data from a Birth Defects Registry.* Birth Defects Research Part A: Clinical and Molecular Teratology, 2013.

213. Lee, J., C.S. Tan, and K.S. Chia, *A practical guide for multivariate analysis of dichotomous outcomes.* Annals of the Academy of Medicine, Singapore, 2009. 38(8): p. 714-9.

214. Charney, E.B., et al., *Management of the newborn with myelomeningocele: time for a decision-making process.* Pediatrics, 1985. 75(1): p. 58-64.

215. Centers for Disease Control and Prevention. *Classification of Diseases, Functioning, and Disability: International Classification of Diseases, Tenth Revision, Clinical Modification (ICD-10-CM)*. 2013 [cited 2013 October 7]; Available from: http://www.cdc.gov/nchs/icd/icd10cm.htm.

216. Centers for Disease Control and Prevention. *Birth Defects: Diagnosis During Pregnancy* 2011 [cited 2013 October 27]; Available from: http://www.cdc.gov/ncbddd/birthdefects/diagnosis.html.

217. Roberts, C.D., L.D. Stough, and L.H. Parrish, *The role of genetic counseling in the elective termination of pregnancies involving fetuses with disabilities.* Journal of Special Education, 2002. 36(1): p. 48-55.

218. Saxton, M., *Prenatal screening and discriminatory attitudes about disability.* Women Health, 1987. 13(1-2): p. 217-24.

219. Spina Bifida Association of Central Florida. *Redefining Spina Bifida*. 2013 [cited 2013 August 24]; Available from: http://sbacentralflorida.org/about-us/redefining-spina-bifida/.

220. *Truven Health Analytics: Better Understand Health Economics and Treatment Outcomes*. 2013 [cited 2013 September 6]; Available from: http://www.truvenhealth.com/your_healthcare_focus/pharmaceutical_and_medical_device/data_databases_and_online_tools.aspx.

221. Agency for Healthcare Research and Quality (AHRQ). *The Kids' Inpatient Database*. 2013 [cited 2014 January 23]; Available from: http://www.hcup-us.ahrq.gov/kidoverview.jsp.

222. Krieger, N., et al., *Painting a truer picture of US socioeconomic and racial/ethnic health inequalities: the Public Health Disparities Geocoding Project.* American Journal of Public Health, 2005. 95(2): p. 312-23.

223. McPherson, M., et al., *Implementing community-based systems of services for children and youths with special health care needs: how well are we doing?* Pediatrics, 2004. 113(5 Suppl): p. 1538-44.

224. van Dyck, P.C., et al., *Prevalence and characteristics of children with special health care needs.* Archives of Pediatrics and Adolescent Medicine, 2004. 158(9): p. 884-90.

225. Strickland, B., et al., *Access to the medical home: results of the National Survey of Children with Special Health Care Needs.* Pediatrics, 2004. 113(5 Suppl): p. 1485-92.

226. Chiri, G. and M.E. Warfield, *Unmet need and problems accessing core health care services for children with autism spectrum disorder.* Maternal and Child Health Journal, 2012. 16(5): p. 1081-91.

227. Romaire, M.A., J.F. Bell, and D.C. Grossman, *Medical home access and health care use and expenditures among children with special health care needs.* Archives of Pediatrics and Adolescent Medicine, 2012. 166(4): p. 323-30.

228. Tu, W., S. Tedders, and J. Tian, *An exploratory spatial data analysis of low birth weight prevalence in Georgia.* Applied Geography, 2012. 32(2): p. 195-207.

229. Tian, J., et al., *A spatial-temporal analysis of low birth weight prevalence in Georgia, USA.* GeoJournal, 2013. 78(5): p. 885-895.

230. Rushton, G. and P. Lolonis, *Exploratory spatial analysis of birth defect rates in an urban population.* Statistics in Medicine, 1996. 15(7-9): p. 717-26.

231. Bowman, R.M. and D.G. McLone, *Neurosurgical management of spina bifida: research issues.* Developmental Disabilities Research Reviews, 2010. 16(1): p. 82-87.

232. Dijkstra, E.W., *A Note on Two Problems in Connexion with Graphs.* Numerische Mathematik, 1959. 1: p. 269 - 27 I.

233. Zhan, F.B. and C.E. Noon, *Shortest Path Algorithms: An Evaluation using Real Road Networks.* Transportation Science, 1998. 32(1): p. 65-73.

234. Black, P.E. *Dijkstra's algorithm*. Dictionary of Algorithms and Data Structures 2006 [cited 2013 January 14]; Available from: http://xlinux.nist.gov/dads/HTML/dijkstraalgo.html.

235. Henry, K.A., et al., *Breast cancer stage at diagnosis: is travel time important?* Journal of Community Health, 2011. 36(6): p. 933-42.

236. Huang, B., et al., *Does distance matter? Distance to mammography facilities and stage at diagnosis of breast cancer in Kentucky.* Journal of Rural Health, 2009. 25(4): p. 366-71.

237. Pinto, N.M., et al., *Regionalization in neonatal congenital heart surgery: the impact of distance on outcome after discharge.* Pediatric Cardiology, 2012. 33(2): p. 229-38.

238. Allshouse, W.B., et al., *Geomasking sensitive health data and privacy protection: an evaluation using an E911 database.* Geocarto International, 2010. 25(6): p. 443-452.

239. Hampton, K.H., et al., *Mapping health data: improved privacy protection with donut method geomasking.* American Journal of Epidemiology, 2010. 172(9): p. 1062-9.

240. Mobley, L.R. and H.E. Frech, 3rd, *Managed care, distance traveled, and hospital market definition.* Inquiry, 2000. 37(1): p. 91-107.

241. Surgical Advisory Panel: American Academy of Pediatrics, *Guidelines for referral to pediatric surgical specialists.* Pediatrics, 2002. 110(1 Pt 1): p. 187-91.

242. Florida Agency for Health Care Administration. *Hospital Beds and Services List*. 2010 [cited 2014 January 20]; Available from: http://ahca.myflorida.com/MCHQ/CON_FA/Publications/archives/docs/HospBedSrvList/Jan2010_HospitalBedsandServicesList.pdf.

243. Florida Agency for Health Care Administration. *Neonatal Intensive Care Service*. Florida Administrative Weekly and Florida Administrative Code 2012 [cited 2012 April 27]; Available from: https://www.flrules.org/gateway/ruleno.asp?id=59C-1.042.

244. Verhoef, M., et al., *Secondary impairments in young adults with spina bifida.* Developmental Medicine & Child Neurology, 2004. 46(6): p. 420-427.

245. Armour, B.S., et al., *Hospitalization for urinary tract infections and the quality of preventive health care received by people with spina bifida.* Disability and Health Journal, 2009. 2(3): p. 145-52.

246. de Jong, T.P., et al., *Treatment of the neurogenic bladder in spina bifida.* Pediatric Nephrology, 2008. 23(6): p. 889-96.

247. Richardson, L.D. and M. Norris, *Access to health and health care: how race and ethnicity matter.* Mount Sinai Journal of Medicine, 2010. **77**(2): p. 166-77.

248. Gresenz, C.R., J. Rogowski, and J.J. Escarce, *Community demographics and access to health care among U.S. Hispanics.* Health Services Research, 2009. 44(5 Pt 1): p. 1542-62.

249. Groeneveld, P.W., et al., *Racial differences in attitudes toward innovative medical technology.* Journal of General Internal Medicine, 2006. 21(6): p. 559-63.

250. Bisgaier, J. and K.V. Rhodes, *Auditing access to specialty care for children with public insurance.* New England Journal of Medicine, 2011. 364(24): p. 2324-33.

251. Kogan, M.D., et al., *Association between underinsurance and access to care among children with special health care needs in the United States.* Pediatrics, 2005. 116(5): p. 1162-9.

252. Miller, H.J. and S.L. Shaw, *Geographic information systems for transportation: principles and applications*. Spatial Information Systems. 2001, New York: Oxford University Press. xi, 458 p.

253. Cromley, E.K. and S. McLafferty, *GIS and public health*. 2nd ed 2012, New York: The Guilford Press. xxiv, 503 p.

254. Jones, S.G., et al., *Spatial implications associated with using Euclidean distance measurements and geographic centroid imputation in health care research.* Health Services Research, 2010. 45(1): p. 316-27.

255. Martin, J.A. and M.J.K. Osterman, *Preterm Birth - United States, 2006 and 2010.* CDC Morbidity and Mortality Weekly Report, 2013. 62(03): p. 136-138.

256. U.S. Census Bureau. *Geographical Mobility: 2002 to 2003.* 2004 [cited 2014 January 20]; Available from: http://www.census.gov/prod/2004pubs/p20-549.pdf.

257. Khoury, M.J., et al., *Residential mobility during pregnancy: implications for environmental teratogenesis.* Journal of Clinical Epidemiology, 1988. 41(1): p. 15-20.

258. Miller, A., C. Siffel, and A. Correa, *Residential mobility during pregnancy: patterns and correlates.* Maternal and Child Health Journal, 2010. 14(4): p. 625-34.

259. Canfield, M.A., et al., *Residential mobility patterns and exposure misclassification in epidemiologic studies of birth defects.* Journal of Exposure Science and Environmental Epidemiology, 2006. 16(6): p. 538-43.

260. Fell, D.B., L. Dodds, and W.D. King, *Residential mobility during pregnancy.* Paediatric and Perinatal Epidemiology, 2004. 18(6): p. 408-14.

261. Primerano, F., et al., *Defining and understanding trip chaining behavior.* Transportation 2008. 35: p. 55-72.

262. Strathman, J., K.J. Dueker, and J.S. Davis, *Effects of household structure and selected travel characteristics on trip chaining.* Transportation 1994. 21: p. 23-45.

263. Nuckols, J.R., M.H. Ward, and L. Jarup, *Using geographic information systems for exposure assessment in environmental epidemiology studies.* Environmental Health Perspectives, 2004. 112(9): p. 1007-15.

264. Wang, Y., et al., *Geocoding capacity of birth defects surveillance programs: results from the National Birth Defects Prevention Network Geocoding Survey.* Journal of Registry Management, 2010. 37(1): p. 22-6.

265. *Ronald McDonald House Charities*. 2013 [cited 2013 October 4]; Available from: www.rmhc.org/home.

266. Oakley, G.P. and J.S. Mandel, *Folic acid fortification remains an urgent health priority.* BMJ, 2004. 329(7479): p. 1376.

APPENDIX A: NBDPN MAJOR BIRTH DEFECTS

Birth Defects Included in the Case Definition
of the National Birth Defects Prevention Network (NBDPN)

Birth Defects	ICD-9-CM Codes	CDC/BPA Codes
Central Nervous System		
Anencephalus	740.0 - 740.1	740.00 - 740.10
Spina bifida without anencephalus	741.0, 741.9 w/o 740.0 -740.10	741.00 - 741.99 w/o 740.0 - 740.10
Hydrocephalus without spina bifida	742.3 w/o 741.0, 741.9	742.30 - 742.39 w/o 741.00 - 741.99
Encephalocele	742.0	742.00 - 742.09
Microcephalus	742.1	742.10
Eye		
Anophthalmia/microphthalmia	743.0, 743.1	743.00 - 743.10
Congenital cataract	743.30 - 743.34	743.32 - 743.326
Aniridia	743.45	743.42
Ear		
Anotia/microtia	744.01, 744.23	744.01, 744.21
Cardiovascular		
Common truncus	745.0	745.00 - 745.01
Transposition of great arteries	745.10, .11, .12, .19	745.10 - 745.19
Tetralogy of Fallot	745.2	745.20 - 745.21, 746.84
Ventricular septal defect	745.4	745.40 - 745.490 (exclude 745.498)
Atrial septal defect	745.5	745.50 - 745.59 (exclude 745.50)
Endocardial cushion defect	745.60, .61, .69	745.60 - 745.69
Pulmonary valve atresia and stenosis	746.01, 746.02	746.00 - 746.01
Tricuspid valve atresia and stenosis	746.1	746.10 (exclude 746.105)
Ebstein's anomaly	746.2	746.20
Aortic valve stenosis	746.3	746.30
Hypoplastic left heart syndrome	746.7	746.70
Patent ductus arteriosus (Include only if weight=>2500 grams or note if unable to exclude <2500 grams infants.)	747.0	747.00
Coarctation of aorta	747.10	747.10 - 747.19
Orofacial		
Cleft palate without cleft lip	749.0	749.00 - 749.09
Cleft lip with and without cleft palate	749.1, 749.2	749.10 - 749.29
Choanal atresia	748.0	748.00

Birth Defects	ICD-9-CM Codes	CDC/BPA Codes
Gastrointestinal		
Esophageal atresia/tracheoesophageal fistula	750.3	750.30 - 750.35
Rectal and large intestinal atresia/stenosis	751.2	751.20 - 751.24
Pyloric stenosis	750.5	750.51
Hirshsprung's disease (congenital megacolon)	751.3	751.30 - 751.34
Biliary atresia	751.61	751.65
Genitourinary		
Renal agenesis/hypoplasia	753.0	753.00 - 753.01
Bladder exstrophy	753.5	753.50
Obstructive genitourinary defect	753.2, 753.6	753.20-29 - 753.60- 69
Hypospadias and Epispadias	752.61, 752.62	752.600 - 752.627 (excluding 752.621)
Musculoskeletal		
Reduction deformity, upper limbs	755.20 - 755.29	755.20 - 755.29
Reduction deformity, lower limbs	755.30 - 755.39	755.30 - 755.39
Gastroschisis	756.79	756.71
Omphalocele	756.79	756.70
Congenital hip dislocation	754.30, .31, .35	754.30
Diaphragmatic hernia	756.6	756.610 - 756.617
Chromosomal		
Trisomy 13	758.1	758.10 - 758.19
Down syndrome (Trisomy 21)	758.0	758.00 - 758.09
Trisomy 18	758.2	758.20 - 758.290
Other		
Fetal alcohol syndrome	760.71	760.71
Amniotic bands	No code	658.80

Source: National Birth Defects Prevention Network Inc. (NBDPN), *Guidelines for Conducting Birth Defects Surveillance*, L. Sever, Editor 2004: Atlanta, GA