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The Relationship between Ventriculoperitoneal Shunts and Shunt Revisions
versus Visual Complaints among Patients with Spina Bifida
in the Arkansas Spina Bifida Research Project

Regina McCollum Sullivan

Georgia State University

A Thesis Submitted to the Graduate Faculty
of Georgia State University in Partial Fulfillment
of the Requirements for the Degree

MASTER OF PUBLIC HEALTH

ATLANTA, GEORGIA

2012

The Relationship between Ventriculoperitoneal Shunts and Shunt Revisions
versus Visual Complaints among Patients with Spina Bifida
in the Arkansas Spina Bifida Research Project

REGINA MCCOLLUM SULLIVAN

(Under the direction of Daniel Crimmins, PhD, Clinical Professor)

ABSTRACT

Many patients with Spina Bifida suffer from hydrocephalus as a complication of their developmental disability and surgeons commonly treat this condition with ventriculoperitoneal shunts. Surgeons have speculated for years that these shunts may cause some type of visual disturbance because of their close proximity to the visual pathways in the brain. Little research has been done, however, to support or discourage this commonly held belief. Questions and data from the Arkansas Spina Bifida Research Project were used to examine whether ventriculoperitoneal (VP) shunts *and* VP shunt revisions increase reports of visual complaints for the individuals participating in this research project. This cross sectional design used responses to the vision questions from the 2005 Arkansas Spina Bifida Questionnaire. Results showed a 333% increase in reported vision complaints after receiving a VP shunt, but no significance with the increase in vision complaints for those having three or more VP shunt revisions. Females were 50% to 60% less likely to report vision complaints in both multivariate linear logistic models. While these results indicate the potential relationship between VP shunts and vision concerns, they must be viewed cautiously in light of study limitations due to the small sample size, selection bias, and study design.

INDEX WORDS: Spina Bifida, Myelomeningocele, Hydrocephalus, Vision

APPROVAL PAGE

The Relationship between Ventriculoperitoneal Shunts and Shunt Revisions
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in the Arkansas Spina Bifida Research Project

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ACKNOWLEDGEMENTS

I would like to thank the faculty and staff of Georgia State University's Institute of Public Health and Center for Leadership in Disability. I would also like to thank my Georgia LEND Fellows, staff, and director for an inspirational learning experience. I am grateful for the opportunities and guidance given by Drs. Daniel Crimmins, Daniel Mandel, Mark Swanson, Ike Okosun, and Michael Eriksen. Your encouragements, directions, and passions for public health have proven contagious and motivated me to want to achieve great things.

Thank you to my friends and family for your love, support, and encouragement throughout this process.

Finally, I would like to give special thanks to my husband, Joseph Sullivan, for his support while I pursued my MPH.

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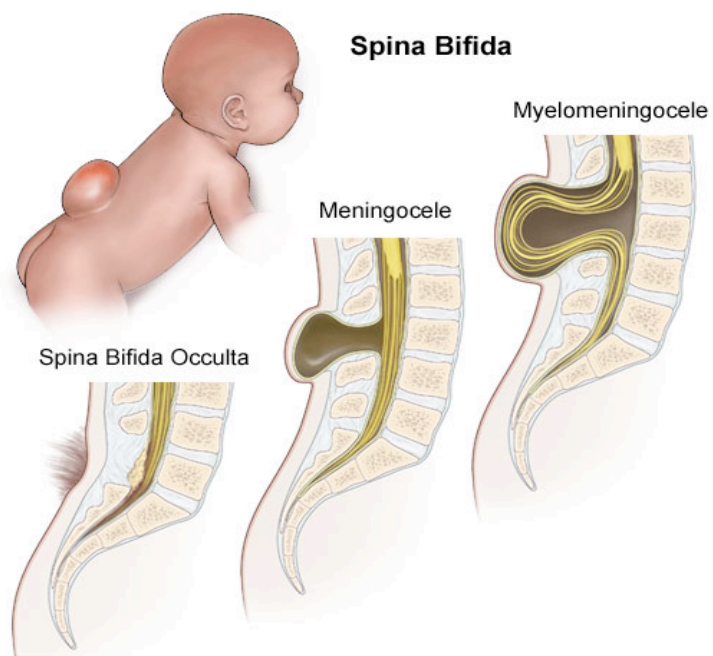
Chapter I

INTRODUCTION

1.1 Background

Spina bifida is a neurodevelopmental disorder with a complex etiology that involves environmental factors intertwined with genetic aspects, and it is considered the most common birth defect affecting the central nerve system. (Frether & Brei, 2010) The term “Spina bifida” encompasses a variety of neural tube malformations that can occur anywhere along the spine. Basically, there are three common forms of spina bifida: myelomeningocele, meningocele, and spina bifida occulta. (Frether & Brei, 2010; Center of Disease Control, 2011) Myelomeningocele (open spina bifida) is the most severe form of the condition and occurs when a sac of spinal cord fluid comes through the opening in the baby’s back causing loss of feeling and the inability to move his or her legs. Meningocele is the second most discussed version of this birth defect but less damaging, because the spinal cord is not in the sac protruding out of the infant’s back. Thus, the structural malformation of the spinal column is less severe and as a result functional limitations of the lower body are less severe. The mildest form of spina bifida is called “spina bifida occulta” and involves only a small opening in the spinal cord, which makes it harder to diagnose. (Figure 1) Many people with spina bifida occulta are not diagnosed until much later in life. (Pico, Wilson, & Haas, 2009; CDC, 2011) The size and location of the openings determine the extent of the physical and mental impact. Most literature refers to the location of these malformations as the “lesion level”.

Figure 1. Spina Bifida Types



Bing Pictures

According to the National Spina Bifida Association, more than 70,000 people live with some form of spina bifida in the United States. (Pico et al., 2009) From 1999 to 2004, twenty-one nationwide birth defects surveillance systems revealed that Hispanics had the highest rate of births with neural tube developmental issues (4.7 per 10,000 births), Non-Hispanic Blacks or African Americans had the lowest rates (2.64 per 10,000 births), and Non-Hispanic Whites had an average of 3.22 per 10,000 births. (Boulet et al., 2008; CDC, 2011)

While the etiology of spina bifida is not fully understood, regular consumption of folic acid in early pregnancy is associated with greatly reduced incidence of spina bifida, particularly myelomeningoceles. In fact, folic acid fortification of foods in the mid 1990s

is attributed with a reduction in the U.S. rate neural tubal defects (including spina bifida) from 4000 to 3000 a year. (Pico et al., 2009; Magaron, Poenaru, Bransford, & Albright, 2010; Frether & Brei, 2010; Stevenson et al., 2000) Despite this, the incidence of new cases has remains steady over the past decade because of improvements in the diagnosis of spina bifida via ultrasounds, blood test that measures fetal alpha-fetoprotein levels, and sampling of the amniocentesis. (Alriksson-Schmidt, Swanson, & Thibadeau, 2009; Yen, Khoury, & Erickson et al. 1992; Boulet et al., 2008)

Throughout the lifetime of individuals with spina bifida many complications can occur, especially for those with myelomeningoceles who include 70% of individuals with spina bifida. Of those with myelomeningoceles, 70% to 95% develop hydrocephalus. (Pico, et al., 2005; Matson, Mahone, & Zabel, 2005; Thomas & Barnes, 2010)

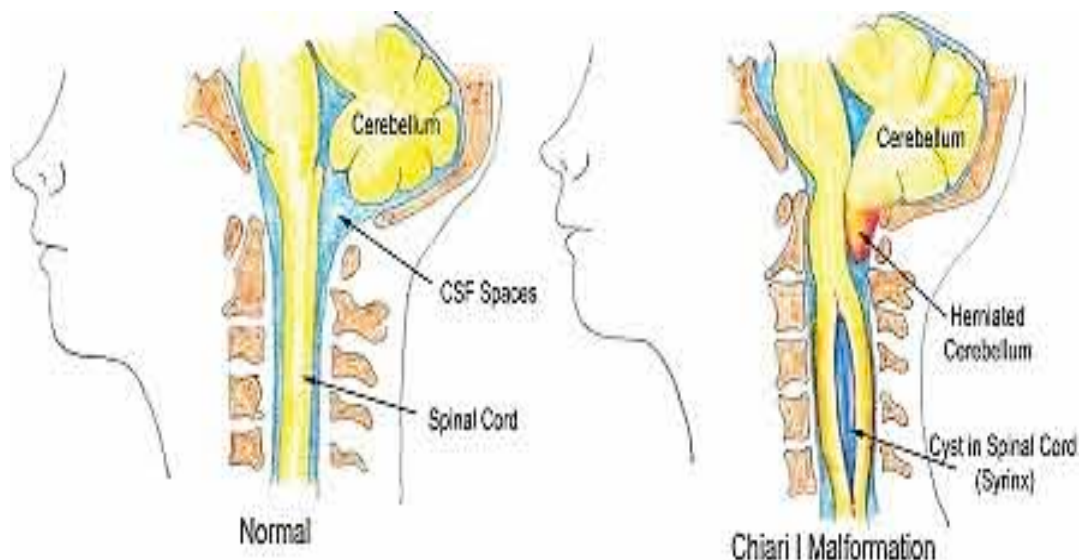
Hydrocephalus occurs when an increase in the head's circumference is produced by a rise in ventricular pressure and cerebrospinal fluid volume in the skull. (Aring et al., 2007)

The most common cause of hydrocephalus in people with myelomeningocele is a common hindbrain abnormality called "Chiari Malformation". (Figure 2) (Bowman & McLone, 2010; Persson, Anderson, Wiklund, & Uvebrant, 2007) The malformation displaces the medulla, lower pons, fourth ventricle, and cerebella vermis into the cervical spinal cord. This displacement interrupts the flow of cerebrospinal fluid thorough the cervical spinal column and causes the fluid to build up in the brain. (Pico et al., 2009)

Since 1970, the most common procedure for treating hydrocephalus is the placement of a ventriculoperitoneal (VP) shunt near vital visual processing pathway in the brain. (Ali, Aman, Khan, Siddique Khanzada, & Ayub, 2009) Many surgeons have speculated that the placement of VP shunts may have an impact on the visual processing and functioning

of patients with hydrocephalus; however, little research has been done to validate this notion. (Alriksson-Schmidt et al., 2009)

Figure 2. Chiari Malformation



Opening Minds

1.2 Purpose of the Study

This study explores the relationship of (VP) shunts and VP shunt revisions with vision complaints among patients with spina bifida surveyed in the Arkansas Spina Bifida Research Project. It is estimated that 95% of patients with myelomeningocele will have hydrocephalus as a complication of their neural tube defect, and surgeons commonly treat 77% of these patient with VP shunts. (Pico, et al., 2005; Matson, Mahone, & Zabel, 2005; Thomas & Barnes, 2010)

Surgeons have speculated for years that VP shunts may cause some type of visual disturbance because of their close proximity to the visual pathways in the brain. However, little research has been done to support this commonly held belief. This study

will add to the limited knowledge on the possible side effects of VP shunts, encourage research into less invasive procedures, and potentially support the inclusion of vision providers in the interdisciplinary treatment of patients with spina bifida.

1.3 Research Question

The objective of this study is to enhance the present body of literature evaluating the relationship of VP shunts and VP shunt revisions with visual complaints via answering the following questions:

1. Is there a relationship between the introduction of VP shunts and reports of visual perception issues, ocular surgery, or visual problems (collectively referred to as “vision complaints”) in the subjects of the Arkansas Spina Bifida Research Project?
2. Is there a relationship between an increased number of VP shunt revisions and vision complaints (as defined above) in the same subjects of the Arkansas Spina Bifida Research Project?

Chapter II REVIEW OF THE LITERATURE

To support the rationale for this study, a review of the literature illustrates the available knowledge on the treatment of hydrocephalus, commonly reported vision issues associated with hydrocephalus, the use of VP shunts and subsequent VP shunt revisions, and an overview of the current protocols for interdisciplinary treatment for spina bifida patients.

2.1 Hydrocephalus

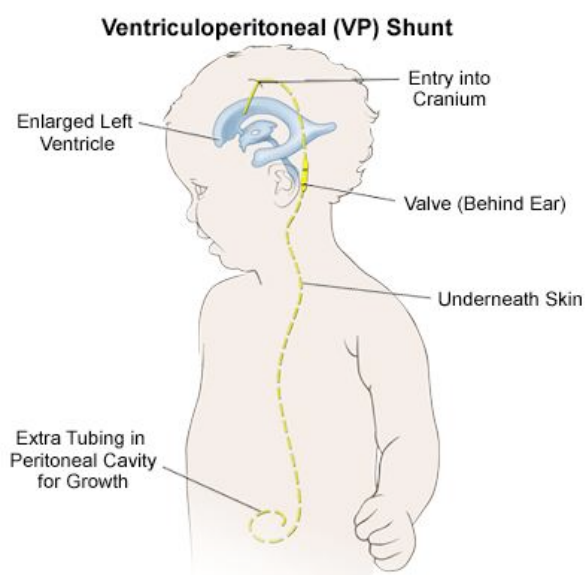
Hydrocephalus is an enlargement of the circumference of the head created by an increase in cerebrospinal fluid in the brain associated with the Chiari malformation. (Aring et al., 2007) The Chiari malformation is located in the hindbrain. (Hindbrain, 2009) In about 80% to 90% of patient with myelomeningocele, the medulla oblongata and pons do not sit on top of the cervical spinal cord; they are blended or displaced into the upper part of the spinal cord and column. This malformation in the brain is called a Chiari II Malformation, and it interferes with the flow of cerebrospinal fluid through the spinal cord. (Figure 2) (Pico et al., 2009; Bowman & McLone, 2010) Research on patients with the co-morbidities of hydrocephalus and spina bifida reveals that 75% to 85% of this population requires surgical management. (Pico et al., 2009; Ali et al, 2009; Persson et al, 2007; Sawin & Bellin, 2010)

2.2 Overall Treatment Strategy for Hydrocephalus

The most common procedure used to treat hydrocephalus is the placement of a VP shunt. (Ali et al., 2011; Bani & Hassler, 2006; Ghritlaharey, Shrivastava, & Srivastav, 2012; Sehati, 2012; Prabhakar, et al., 2005) Shunting is largely a mechanical intervention, which requires a lifetime commitment from the surgeon to the patient. (Ali,

et al., 2009) The technique was introduced in the 1950's and offered the possibility of long-term survival for a selected group of infants. In the typical VP shunt placement procedure, the surgeon first makes a small incision toward the back of the skull (above and behind the ear). Then he or she places a long and thin plastic tube (catheter) inside the area where the cerebrospinal fluid has accumulated (the lateral ventricles in the brain). This catheter is then attached to a valve (which controls the flow of the CSF) and the opposite end of the catheter (the distal end) is tunneled under the skin of the scalp, downward toward the lower torso. Finally, a small incision is made in the lower part of the abdomen to help the surgeon place the distal end of the catheter inside the peritoneal cavity, where the excess fluid is absorbed. (Figure 3) (Sehati, 2012; Prabhakar et al., 2005; Margaron et al., 2010; Mitchell, Adzick, Melchionne, Pasquariello, & Sutton, 2004)

Figure 3. Ventriculoperitoneal Shunt



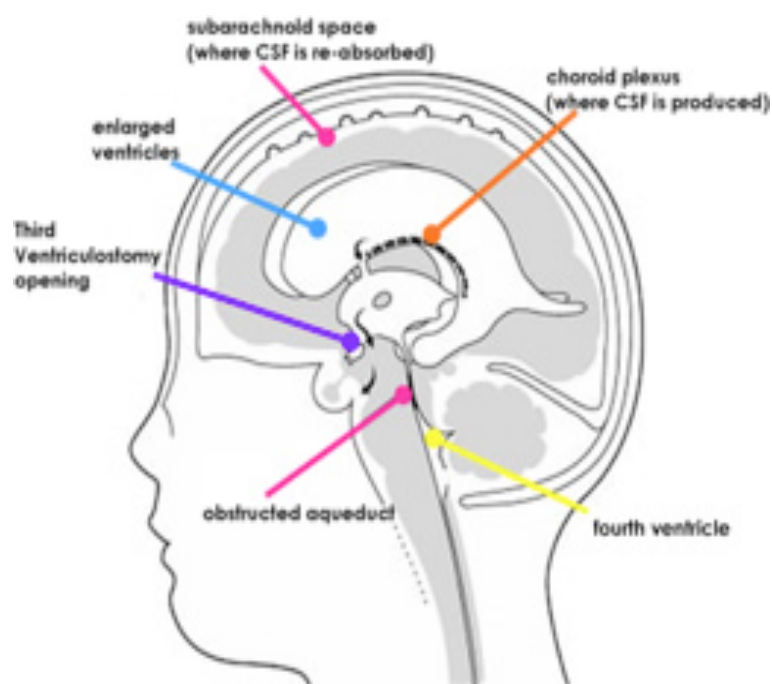
Bing Pictures

During the late 50's and 60's, providers advocated for selective criteria of infants treated with this procedure. They reasoned that allowing the most severe cases to survive would cause a burden to their parents and society. This reasoning led to restricted access to the VP shunting procedure, with only 30 percent of infants born with spina bifida and hydrocephalus receiving treatment. Untreated patients were given only supportive nursing care and survival for these infants was considered an unacceptable outcome. (Bowman & McLone, 2010; Lorber & Salfield, 1981) Current practice extends the use of the procedure to all infants, which along with prenatal diagnosis and fetal surgery, has lead to a decreased in mortality rate from 50% in the 1950's to 10% today. (Laurence, 1974; Bowan & McLone, 2010; Persson et al., 2007)

More recently, the procedure called “endoscopic third ventriculostomy” has come to be considered to be a promising alternative for internal cerebrospinal fluid diversion. (El-Ghandour, 2010; Jallo, Kothbauer, & Abbott, 2005) Surprisingly, ventriculostomies were performed before VP shunting, but the procedure lost popularity because VP shunting was faster and less complicated. The return of ventriculostomies as a viable treated for hydrocephalus has to do with the invention of sophisticated endoscopic instruments and detailed images provided by the MRI. (Jallo et al., 2005) The combination of these technologies allows the surgeon to divert cerebrospinal fluid to a different area of the brain with minimal damage to other structures. Specifically, endoscopic guided third ventriculolostomy allows for the formation of a stoma (an opening that connects a part of the body cavity to the outside environment) on the floor of the third ventricle, which directs CSF into the subarachnoid spaces of the interpeduncular cistern, thus bypassing the obstruction. (Figure 4) This procedure removes the patient's

dependency on shunts and it can be used after a patient has had numerous shunt revisions. (De Ribaupierre, Rillet, Vernet, Regli, & Villemure, 2007; Garton, Kestle, Cochrane, & Steinbok, 2002; Javadpour, Mallucci, Brodbelt, Golash, & May, 2001) The disadvantage of this procedure is that it can only be applied to hydrocephalus cases caused by aqueductal stenosis and a MRI is required to confirm diagnoses. (Jallo et al., 2005) Neurosurgeons are currently looking for ways to apply this less invasive procedure to more individuals with hydrocephalus.

Figure 4. Endoscopic third ventriculostomy



Bing Pictures

2.3 Revision Rates

VP shunts have been the most commonly used method for treating hydrocephalus for the past 5 decades, but the consequences of shunting have remained a major problem. This procedure mandates regular monitoring of the known long-term risks, including shunt infections, distal blockage, and mechanical failure of the shunt. The most common reason for revisions was blockage of the shunt, usually in the upper end of the catheter, and the second most common reason was infections, especially in children. (Ali et al., 2011; Kaplan, Yakar, Orhan, & Erol, 2007) A long-term study reviewed children born with myelomeningocele and hydrocephalus found an average rate of two shunt revisions over a 25-year period. (Bowman & McLone, 2010; Bowman et al., 2001) Monitoring and management of VP shunts clearly requires a lifelong commitment by the patient and surgeon.

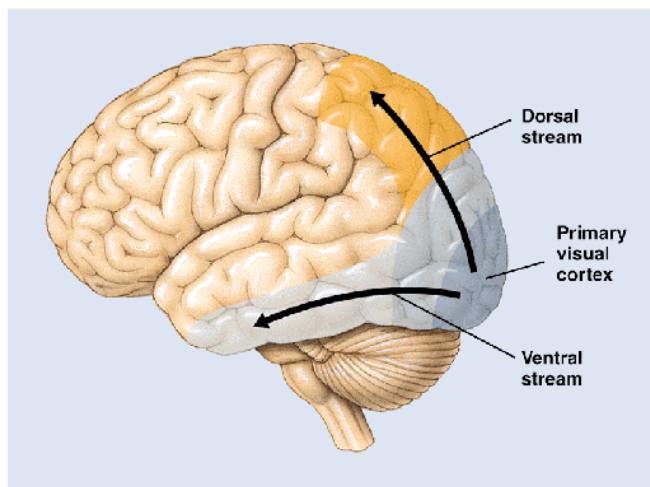
2.4 Associated visual complaints

A large part of the human brain is devoted to vision and visual information processing. First of all, vision is more than just collecting images; it entails visual search, visual attention, and visual guidance. The details captured in visual processing control the body's movement in response to vision information. Understanding all of the components of the visual process is important to evaluating the effects of VP shunts and hydrocephalus on vision. The areas of the brain responsible for sorting and interpreting of visual information is called the ventral and dorsal stream (figure 5). The ventral stream, called the "what pathway", projects from the occipital lobe to the inferotemporal cortex and the dorsal stream, called the "where pathway", runs from the occipital lobes to the posterior parietal cortex. (Macintyre-Beon et al., 2010)

A multitude of neuro-ophthalmic problems are associated with hydrocephalus: ocular motility problems, visual field defects, optic atrophy, and amblyopia. In fact, ocular signs of a shunt obstruction or uncontrolled hydrocephalus involve the presentation of a lateral rectus palsy, divergence palsies, and many other ocular motility disorders. (Caines, Dahl, & Holmstrom, 2007; Arling, et al., 2007; Holgrove, Leach, Herwadkar, & Gnanalingham, 2009; Rudolph, Sterker, Till, Grafe, & Geyer, 2009) Thus, understanding if VP shunts are causing visual problems is difficult because hydrocephalus may cause some of the same complaints, which has lead to some researchers comparing prior vision assessment records to assessments during hydrocephalus and after shunting.

Figure 5. Ventral and Dorsal Streams

► Visual Information Pathways



Bing Pictures

The verdict on VP shunting causing or relieving vision complaints is mixed. Some researchers have found that revisions of shunts have been shown to be a risk factor

for strabismus and amblyopia. (Caines et al., 2007) Others noted that medical stability following shunt revisions revealed that spatial visual memory and object visual memory did not improve after revisions and continued to decline. (Matson, Mahone, & Zabel, 2005) Altintas and associates (Altintas et al., 2005) found that shunt treatment improved some ocular issues but others persisted. They also found that VP shunt revisions were related to increased reports of strabismus and reduced best-corrected visual acuity. Another group of investigators found no difference in the strabismus, abnormal head posture, nystagmus, or ocular motilities among children with hydrocephalus that had or did not have shunt revisions. (Arling et al., 2007)

Chapter III

METHODS AND PROCEDURES

3.1 Data Source

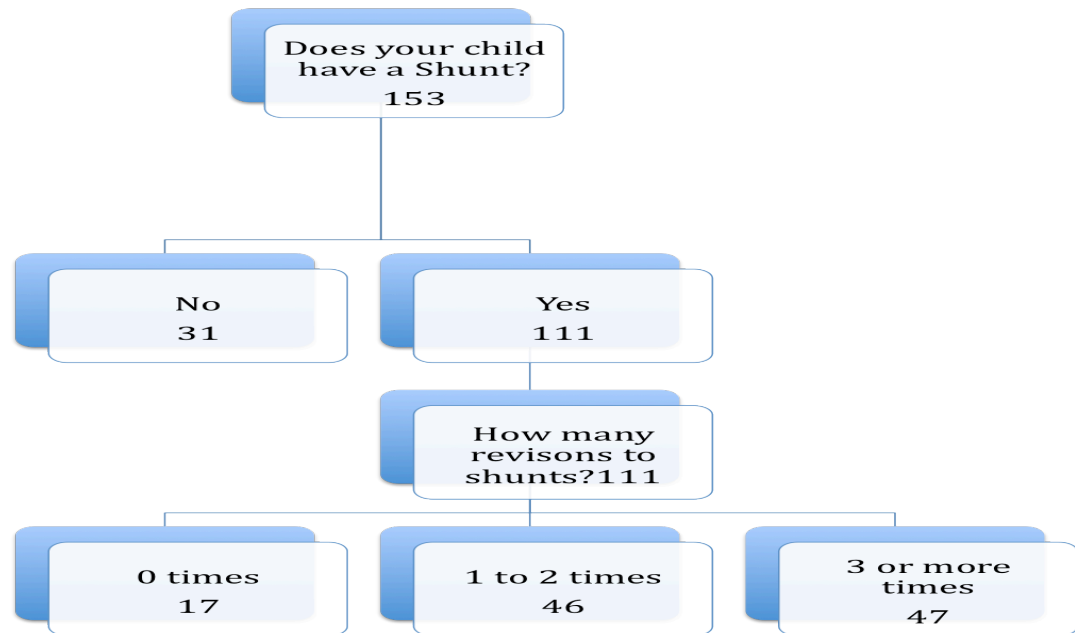
The data used in this study was obtained from a project sponsored by the Arkansas Spinal Cord Commission (ASCC), which maintains a statewide registry of spinal cord disabilities. The mission of the ASCC is to identify and meet the unique and lifelong needs of people with spinal cord disabilities caused by trauma, infection, tumors, or birth defects. The project included a longitudinal database that contained responses from a survey of persons with spina bifida in the state of Arkansas. The questionnaire was first administered in 1993 to individuals from 12 through 31 years of age with spina bifida living in Arkansas, including their parents. The questions covered several details (such as shunt complications, school experiences, illicit drug use, vision problems, and secondary health conditions). A similar follow-up questionnaire was administered to the same subjects in 2005. Both surveys contained 132 questions, which were divided into seven sections: interview identification, biological mother information, biological father information, secondary conditions, medical care providers, client employment, and client alcohol and drug use. The 1993 and 2005 surveys were linked to each child-parent responses, which created the ability to conduct a detailed comparison of changes over time. A total of 165 children-parent teams participated in both the 1993 and 2005 Arkansas Spina Bifida Questionnaires (ASBQs). This thesis only used information from the 2005 parental questionnaire because the responses were more consistent and easier to incorporate in a cross sectional design.

3.2 Variables

We identified several questions from the secondary conditions section of the ASBQ that focused on vision. The specific items used for this study included the questions: 1) Has your child ever been diagnosed with having a visual perception problem or a visual motor problem? 2) Has your child ever had eye surgery to repair or improve his/her vision? and 3) Has your child had vision problems within the last year? All of these questions were answered in a dichotomous modality with simple “yes” (scored as 1) and “no” (scored as 0) responses. The dependent variables (based on these questions) were abbreviated for charting purposes and were labeled as *Visual Perception*, *Ocular Surgery*, and *Vision Problems*, respectively. Due to issues with low cell counts in the statistical calculations, the three dependent variables were combined into one variable called *Vision Complaints*.

The next two questions were selected because this investigation was interested in the relationship of vision complaints with simply having a VP shunt versus having multiple VP shunt revisions. The inquiries selected from the survey were “Does your child have a shunt? ”, which was coded as “yes” (1) or “no” (0), and labeled as independent variable *Have a Shunt* for data presentation. The second item chosen from the questionnaire was “How many times was your child’s shunt revised? ”, which was abbreviated as independent variable *Shunt Revisions* in the data set and recoded into three categories: “yes to shunt, but no revisions” (0), “yes, with 1 to 2 revisions” (1), and “yes, with 3 or more revisions” (2). (See Figure 6.) The rationale for the distributions of shunts was based on research from Bowman and McLone (2010) citing the average number of shunt revisions as approximately two during a 25-year period.

Figure 6: Diagram of Shunt Distribution



3.3 Co variables

The co variables, also referred to as confounders, were selected based on variables known to have an effect on outcomes: *Age, Sex, Racial/Ethnic Background, Lesion Level, and Social Economic Status (SES)*.

SES was operationally defined as household income, which was dichotomized into “family incomes between 0 and \$30,000” (0) and “family incomes above \$30,000” (1); this was based on the structure of the survey questions and U.S. Census data reporting the mean average income in Arkansas of \$32,000 in 2000. *Racial/ethnic background* was initially divided into three categories: white, black, and other; only two categories were maintained because over 80% of the respondents answered white, with 18% black and less than 1% other; several racial categories that were included in the “other” category had zero responses, which produced false significance in the distribution

of demographics. *Lesion Level* was chosen as a confounder because historically the location of the lesion determines the impact of spina bifida; this response was divided into upper and lower lesion locations. *Age* at the time of the interview was recoded into four categories: 0= “12 to 16 years of age”, 1=”17 to 20 years of age”, 2=” 21 to 25 years of age”, and 3= “26 to 31 years of age”.

3.4 Statistical Analyses

Statistical analyses were performed using SPSS 20.0 statistical analysis software. All the frequencies, chi squares, univariate, and multivariate analyses were done unweighted due to the small size of the sample. This study design was cross sectional and the design is low in power or statistical strength. Statistical significance, in this investigation, was determined by $p < .05$ and 95% confidence intervals.

Descriptive analyses involved performing frequency procedures on the total study population and on all five variables with respect to age, sex, race, household income, VP shunt distribution, VP shunt revisions, vision complaints, vision problems, and ocular surgeries. The frequency evaluations allowed for the identification of potential problems for the univariate analyses such as low cell count. Pearson’s chi-squared p-values were calculated to determine differences between groups. However, when the cell counts were low Fisher’s exact values were used to determine the p-values. Univariate analyses were used to obtain 95% CIs, odds ratios, and p values for each dependent variable (visual perception, ocular surgery, and vision problems) separately versus each independent variable (have a shunt and shunt revisions) separately. Multivariate analyses were used to determine the impact of confounders on the dependent variables (visual perception, ocular surgery, and vision problems) separately versus independent variable interactions.

Chapter IV

RESULTS

4.1 Background Descriptors of the Sample

Basic demographic descriptors of the children and young adults (n=153) with spina bifida in this study are in Table 4.1. Individuals were distributed into four age groupings, with 21 to 25 year-olds the largest age group representing 29% of the sample, and 12 to 16 year-olds the smallest at 19%. Overall, sex (male and female) and annual household income were evenly distributed. The racial/ethnic diversity of the entire sample revealed that 80% were Non-Hispanic white and 20% other. The majority of the people in this sample (64%) reported having lower level lesions.

Table 4.1 Demographic Profile of Study Sample

Variable	N = 153	Percentage
Age		
12 to 16 years old	29	19%
17 to 20 years old	40	26%
21 to 25 years old	44	29%
26 to 31 years old	40	26%
Sex		
Male	72	47%
Female	81	53%
Race		
White	123	80%
*Other	30	20%
Lesion Level		
Upper	52	34%
Lower	98	64%
Missing	3	2%
Household Income		
\$30,000 and below	63	41%
\$30,001 and above	67	44%
Missing	23	15%

The next five frequency tables evaluate the distribution of the selected variables, which helps predict possible statistical issues affecting more complex evaluations.

Table 4.2 Demographics by Dependent Variable 1 – Visual Perception Problems

<i>Demographics</i>	<i>Visual Perception Problems</i>		<i>Significance</i>
	<i>No</i>	<i>Yes</i>	
Age			
12 to 16 years old	18(62.1%)	11(37.9%)	X ² = 2.097 p= .552 df(3)
17 to 20 years old	22(56.4%)	17(43.6%)	
21 to 25 years old	27(67.5%)	13(32.5%)	
26 to 31 years old	16(51.6%)	15(48.4%)	
N = 139			
Sex			
Male	39(53.4%)	34(46.6%)	X ² = 2.527 p=.123 df(1)
Female	44(66.7%)	22(33.3%)	
N = 139			
Race			
White	66(58.9%)	46(41.1%)	X ² = .147 p= .701 df(1)
Other	17(63.0%)	10(37.0%)	
N = 139			
Lesion Level			
Upper	27(56.2%)	21(43.8%)	X ² = .507 p= .583 df(1)
Lower	55(62.5%)	33(37.5%)	
N = 136			
Household Income			
\$30,000 and below	35(57.4%)	26(42.6%)	X ² = .089 p= .857 df(1)
\$30,000 and above	39(60%)	26(40%)	
N = 126			

Table 4.2 demonstrates that none of the co-variables of interest should have a significant impact on the variable *Perception Problems* and the majority of the respondents answered “no” to visual perception problems.

Table 4.3 Demographics by Dependent Variable 2 - Ocular Surgery

<i>Demographics</i>	<i>Ocular surgery</i>		<i>Significance</i>
	<i>No</i>	<i>Yes</i>	
Age			
12 to 16 years old	28 (96.6%)	1(3.4%)	p=.53* df(1)
17 to 20 years old	32 (84.2%)	6(15.8%)	
21 to 25 years old	35(83.3%)	7(16.7%)	
26 to 31 years old	29(90.6%)	3(9.4%)	
N = 141			
Sex			
Female	65(86.7%)	10(13.3%)	p=.80* df(1)
Male	59(89.4%)	7(10.6%)	
N = 141			
Race			
White/ Non-Hispanic	98(86.7%)	15(13.3%)	p=.53* df(1)
Black/ Non-Hispanic	26(92.9%)	2(7.1%)	
N = 141			
Lesion Level			
Upper	42(87.5%)	6(12.5%)	X ² = .002 p= .96 df(1)
Lower	79(87.8%)	11(12.2%)	
N = 138			
Household Income			
\$30,000 and below	57(90.5%)	6(9.5%)	X ² = .94 p= .33 df(1)
\$30,000 and above	56(84.8%)	10(15.2%)	
N = 129			

* Fishers exact test was used for p-value when cells were low.

Table 4.3 reveals the majority of the people in this sample did not have ocular surgery to correct vision problems. The cell counts were low in the age, race, and sex categories, which means Fisher's exact test was used for p-values. The category *Lesion Level* reveals a significant difference between upper and lower lesion levels with the majority of the sample having lower lesions.

Table 4.4 Demographics by Dependent Variable Visual Problems in Past Year

<i>Demographics</i>	<i>Visual Problems</i>		<i>Significance</i>
	<i>No</i>	<i>Yes</i>	
Age			
12 to 16 years old	27 (93.1%)	2 (6.9 %)	X ² = 2.25 p=.52 df(3)
17 to 20 years old	34 (87.2%)	5 (12.8%)	
21 to 25 years old	34 (81.0%)	8 (19.0%)	
26 to 31 years old	28 (87.5%)	4 (12.5%)	
N = 142			
Sex			
Male	60 (84.0%)	12(16%)	p= .46* df(1)
Female	63 (89.6%)	7(10.4%)	
N = 142			
Race			
White/ Non-Hispanic	100 (87.7%)	14(12.3%)	p= .53* df(1)
Other	25(84.0%)	6(16.0%)	
N = 142			
Lesion Level			
Upper	40 (81.6%)	9 (18.4%)	p= .30* df(1)
Lower	80 (88.9%)	10 (11.1%)	
N = 139			
Household Income			
\$30,000 and below	52 (83.9%)	10 (16.1%)	p= .23* df(1)
\$30,000 and above	61 (91.0%)	6 (9.0%)	
N = 129			

* Fisher's exact test was used for p-value when cell counts are low.

Table 4.4 shows that the majority of the sample answered “no” to visual problems within the last year. Again, Fisher’s exact test p values were used to establish significance because of low cell counts with sex, race, lesion level, and income; none of the confounders have a p-value that suggest anything outside of chance.

Table 4.5 Demographics by Independent Variable 1 - Have a Shunt

<i>Demographics</i>	<i>Have a Shunt</i>		<i>Significance</i>
	<i>No</i>	<i>Yes</i>	
Age			
12 to 16 years old	9 (31%)	20 (69%)	X ² = 5.06 p= .17 df(3)
17 to 20 years old	4 (10%)	36 (90%)	
21 to 25 years old	10 (24%)	31(76%)	
26 to 31 years old	8 (25%)	24(75%)	
N = 142			
Sex			
Male	14(21%)	54(80%)	p=.84* df(1)
Female	17(23%)	57(77%)	
N = 142			
Race			
White	25(21.9%)	89(78.1%)	p=1.00* df(1)
Other	6(21.4%)	22(78.6%)	
N = 142			
Lesion Level			
Upper	7(14.3%)	42(85.7%)	p= .14* df(1)
Lower	24(26.7%)	66(73.3%)	
N = 139			
Household Income			
\$30,000 and below	12(19.4%)	50(80.6%)	p= .83* df(1)
\$30,000 and above	15(22.4%)	52(77.6%)	
N = 129			

* Fisher's exact test was used for p-value when cell counts are low.

Table 4.5 reveals that a large number of the individuals (in this population) have a shunt, but the p-values suggests that the distribution is not beyond chance.

Table 4.6 Demographics by Independent Variable 2 - How many revisions?

<i>Distribution</i>	<i>How Many Revisions</i>			<i>Significance</i>
	<i>0</i>	<i>1 or 2</i>	<i>3 or more</i>	
Age				
12 to 16 years old	2(10.5%)	11(57.9%)	6(31.6%)	X ² = 10.57 p = .10 df(6)
17 to 20 years old	4(10.8%)	16(43.2%)	17(45.4%)	
21 to 25 years old	3(9.7%)	12(38.7%)	16(51.6%)	
26 to 31 years old	8(34.8%)	7(30.4%)	8(34.8%)	
N = 110				
Sex				
Male	8(14.0%)	23(40.4%)	26(45.6%)	X ² = .45 p = .80 df(2)
Female	9(17.0%)	23(40.4%)	21(39.6%)	
N = 110				
Race				
White	10(11.2%)	37(41.6%)	42(47.2%)	X ² = 7.55 p = .02 df(2)
Other	7(33.3%)	9(42.9%)	5(23.8%)	
N = 110				
Lesion Level				
Upper	5(11.9%)	15(35.7%)	22(52.4%)	X ² = 2.05 p = .36 df(2)
Lower	11(16.9%)	29(44.6%)	25(38.5%)	
N = 110				
Household Income				
\$30,000 and below	10(20.4%)	18(36.7%)	21(42.9%)	X ² = 2.74 p = .25 df(2)
\$30,000 and above	5(9.6%)	25(48.1%)	22(42.3%)	
N = 101				

Table 4.6 illustrates that most of the respondents have had at least one shunt revision and a majority of them have had 1 or more shunt revisions. The distribution of “race/ethnicity” appears to be the only significant finding, with Non-Hispanic whites having a larger percentage of 3 or more shunt revisions.

4.2 Univariate Analyses of Dependent Variables Separately

The next step of this evaluation looks at the univariate relationships between each dependent variable (Visual Perception, Ocular Surgery, and Vision Problems) and each independent variable (Have a Shunt and Shunt Revisions). The results for the dependent variable *Ocular Surgery* were not included, because the extremely low cell count resulted in an uninterpretable odds ratio.

Table 4.7 Univariate Analyses of Dependent Variables (Visual Perception, Ocular Surgery, and Vision Problems) versus Independent Variable 1 - Have a Shunt

<i>Dependent Variables</i>		<i>Have a Shunt</i>		<i>Significance</i>
		<i>No</i>	<i>Yes</i>	
Visual Perception	No	24	59	
Visual Motor Problems		28.9%	71.1%	p=.02*
N = 139		77.4%	54.6%	OR=2.85
df(1)	Yes	7	49	95%CI
		12.5%	87.5%	(1.13,7.17)
		22.6%	45.4%	
Vision Problems	No	26	97	
within last year		21.1%	78.9%	p= 1.00*
N = 141		86.7%	87.4%	OR=.94
df(1)	Yes	4	14	95%CI
		22.2%	77.8%	(.28,3.09)
		13.3%	12.6%	

* Fisher's exact test was used for p-value when cell counts are low.

Table 4.7 shows that having a shunt increases the likely of a client reporting a visual perception problem by 2.85 times or 185% with $p < .05$ (significant). The univariate analysis on visual problems within the past year revealed no significant relationship with having a shunt.

Table 4.8 Univariate Analyses of Dependent Variables (Visual Perception, Ocular Surgery, and Vision Problems) versus Independent Variable (Shunt Revisions)

<i>Dependent Variables</i>		<i>Shunt Revisions</i>			<i>Chi Square</i>
		<i>0</i>	<i>1 or 2</i>	<i>3 or more</i>	
Visual Perception N=107 df(2)	No	10 17.2%	25 43.1%	23 39.7%	$X^2 = .804$
	Yes	6 12.2%	20 40.8%	23 46.9%	
		62.5%	55.6%	50.0%	
		37.5%	44.4%	50.0%	
	Odds Ratio	Ref	1.33	1.67	
	95% CI	Ref	(.41,4.30)	(.52,5.35)	
	p-values	Ref	.63	.39	
Ocular Surgery N=108 df(2)	No	15 16.5%	41 45.1%	35 38.5%	$X^2 = 6.07$
	Yes	1 5.9%	4 23.5%	12 70.6%	
		93.8%	91.1%	74.5%	
		6.2%	8.9%	25.5%	
	Odds Ratio	Ref	1.46	5.14	
	95% CI	Ref	(.15,14.16)	(.61,43.18)	
	p-values	Ref	.74	.13	
Vision Problems within last year N=110 df(2)	No	15 15.8%	44 46.3%	36 37.9%	$X^2 = 7.23$
	Yes	2 13.3%	2 13.3%	11 73.3%	
		88.2%	95.7%	76.6%	
		11.8%	4.3%	23.4%	
	Odds Ratio	Ref	.34	2.29	
	95% CI	Ref	(.04,2.63)	(.45,11.61)	
	p-values	Ref	.30	.32	

In Table 4.8, the sample population has been reduced to only individuals that have a shunt. This univariate analyses model was constructed to investigate the relationship between each dependent variable and the numbers of shunt revisions the individuals with shunts had acquired; there were no significant relationships.

4.3 Multivariate Analyses of Dependent Variables Separately

The next evaluations focus on multivariate models that measure the relationship of each dependent variable (Visual Perception, Ocular Surgery, and Vision Problems) and the effects of confounders on each independent variable (Having a Shunt and Shunt Revisions).

Table 4.9 Multivariate Analyses for Dependent Variables (Visual Perception, Ocular Surgery, and Vision Problems) with Confounders versus Independent Variable 1 (Have a Shunt)

<i>Independent Variables with confounders</i>	<i>Dependent Variables</i>	
	<i>Visual Perception Problems OR, 95%CI, p-Values</i>	<i>Visual Problems Odd Ratios 95%CI p-Values</i>
Having a Shunt	4.29(1.42,12.95) .01	1.02(.25,4.13) .98
Age		
12 to 16 years old	Reference	Reference
17 to 20 years old	1.13(.37,3.48)	1.29(.21,7.98) .79
21 to 25 years old	.914(.29,2.89)	1.83(.32,10.44) .50
26 to 31 years old	1.83(.53,6.30)	1.29(.19,8.95) .80
Sex		
Male	Reference	Reference
Female	.41(.18,.90) .03	.50(.15,1.63) .25
Race		
White Non-Hispanic	Reference	Reference
Other	1.16(.41,3.30)	.64(.17,2.45) .52
Lesion Level		
Upper	Reference	Reference
Lower	1.04(.45,2.38)	.62(.19,2.00) .43
Household Income		
\$30,000 and below	Reference	Reference
\$30,000 and above	.88(.40,1.93) .75	.61(.20,1.90) .39

The first thing that stands out on Table 4.9 is the odds ratio for visual perception problems (OR: 4.29; CI: 1.42,12.95; $p < .05$), which reveals the likelihood of an individual having a shunt reporting visual perception problems increases 4.29 times or 329%; this finding suggests that confounders enhanced the relationship. Unfortunately, the small sample size distorted the results of “Ocular Surgery” in the multivariate model; however, the combined dependent variable model contains the responses to this question. The report of visual problems was not affected by the addition of the confounders.

Females, in this sample, seem to be 59% less likely to report visual issues.

Table 4.10 Multivariate Analyses for Dependent Variables (Visual Perception, Ocular Surgery, and Visual Problems) with Confounders versus Independent Variable 2 (Shunt Revisions)

Independent Variables with Confounders	Visual Perception Odds Ratio 95%CI P-Values	Ocular Surgery Odds Ratio 95%CI P-Values	Visual Problems Odds Ratio 95%CI P-Values
Revisions			
0 revisions	Reference	Reference	Reference
1 or 2 revisions	1.16(.26,5.23) .85	1.37(.10,18.79) .81	1.67(.12,22.89) .72
3 or more revisions	1.78(.39,8.00) .46	6.96(.56,86.84) .13	11.50(.87,151.38) .06
Age			
12 to 16 years old	Reference	Reference	Reference
17 to 20 years old	1.07(.31,3.72) .92	2.72(.27,27.19) .40	2.09(.19,23.42) .55
21 to 25 years old	1.12(.30,4.17) .87	3.40(.33,35.58) .31	4.13(.37,46.18) .25
26 to 31 years old	2.54(.53,12.17) .25	5.14(.35,75.39) .23	4.12(.24,70.96) .33
Sex			
Male	Reference	Reference	Reference
Female	.43(.18,1.05) .06	.51(.14,1.80) .30	.50(.12,2.14) .35
Race			
White	Reference	Reference	Reference
Other	1.10(.33,3.64) .88	1.16(.20,6.94) .87	.39(.07,2.23) .29
Lesion			
Upper	Reference	Reference	Reference
Lower	1.13(.45,2.84) .80	1.53(.44,5.30) .50	.71(.18,2.84) .63
Household Income			
\$30,000 and below	Reference	Reference	Reference
\$30,000 and above	.93(.38,2.26) .87	1.42(.42,4.80) .57	.25(.06,1.06) .06

Table 4.10 demonstrates that none of the multivariate analyses for the independent variable “Shunt Revisions” reveal any significant relationships.

Table 4.11 Univariate Analysis for Combined Dependent Variables (Visual Complaints) versus Independent Variable 1 (Have a Shunt)

		Have a Shunt		
<i>Combined Variables</i>		<i>No</i>	<i>Yes</i>	<i>Significance</i>
Vision Complaints	No	24	50	$X^2= 9.37$
		33%	68%	p=.00
				OR=3.98
N = 139	Yes	7	58	95%CI
		11%	89%	(1.58,10.01)

Table 4.11 displays the findings of a univariate model that combined all three dependent variables (*Visual Perception*, *Ocular Surgery*, and *Vision Problems*) into one variable versus the independent variable of *Have a Shunt*. The odds ratio for this model (OR: 3.98; CI: 1.58, 10.01; $p < .05$) suggests a 3.98 time or 298% increased likelihood of participants reporting some form of visual complaint if they have a shunt. Note that by combining the variables the 95% CI has a smaller range and has higher significance than the variables alone.

Table 4.12 Univariate Analysis for Combined Dependent Variables – Visual Complaints versus Independent Variable 2 - Shunt Revisions

Shunt Revisions

<i>Combined Variables</i>		<i>0</i>	<i>1 or 2</i>	<i>3 or more</i>	<i>Chi Square</i>
Vision Complaints	No	9	24	16	$X^2 = 3.98$
	Yes	7	21	30	
N= 107		18%	49%	33%	
df(2)		56%	53%	35%	
		12%	36%	52%	
		44%	47%	65%	
Odds Ratio		Ref	1.125	2.41	
95% CI		Ref	(.36,3.56)	(.76,7.68)	
p-values		Ref	.84	.14	

Table 4.12 presents the univariate model for evaluating combined dependent variables with the independent variable *Shunt Revisions*. Individuals with 1 to 2 shunt revisions were the largest group, but the p-values did not show any significance.

Table 4.13 Multivariate Analysis with Combined Dependent Variables (Visual Complaints) with Confounders versus Independent Variable 1 (Have a Shunt)

Combined “Visual Complaints”

<i>Independent Variable with confounders</i>	<i>Odds Ratio</i>	<i>95% CI</i>	<i>p-values</i>
Having a Shunt (Yes=1)	5.33	(1.76-16.16)	.003
Age			
12 to 16 years old (0)	Reference	Reference	.65
17 to 20 years old (1)	1.49	(.48-4.61)	.49
21 to 25 years old (2)	1.06	(.33-3.39)	.92
26 to 31 years old (3)	1.97	(.56-6.87)	.29
Sex			
Male	Reference	Reference	
Female	.39	(.18-.87)	.02
Race			
White/ Non-Hispanic	Reference	Reference	
Other	1.07	(.37-3.08)	.90
Lesion Level			
Upper	Reference	Reference	
Lower	.64	(.28-1.47)	.29
Household Income			
\$30,000 and below	Reference	Reference	
\$30,000 and above	1.20	(.54-2.65)	.65

This analysis follows the trend of the previous *Having a Shunt* multivariate models with an increased likelihood of 5.33 times or 433% in reporting vision issues when the confounding variables are added to the model, (OR: 5.33; CI: 1.76-16.16; $p < .05$). Females again show a 61% decreased likelihood of reporting an issue, (OR: .39; CI: .18-.87; $p < .05$).

Table 4.14 Multivariate Analysis with Combined Dependent Variables – Visual Complaints versus Independent Variable 2 - Shunt Revisions

Combined Vision Complaints

<i>Independent Variable with confounders</i>	<i>Odds Ratio</i>	<i>95%CI</i>	<i>p-values</i>
Revisions			
Yes to Shunt (0)	Reference	Reference	.07
1 to 2 revisions (1)	1.02	(.22 – 4.79)	.98
3 or more revisions (2)	3.06	(.64-14.57)	.16
Age			
12 to 16 years old (0)	Reference	Reference	.65
17 to 20 years old (1)	1.26	(.35-4.59)	.72
21 to 25 years old (2)	1.12	(.29-4.38)	.87
26 to 31 years old (3)	2.62	(.52-13.33)	.25
Sex			
Male	Reference	Reference	
Female	.40	(.16-1.00)	.05
Race			
White/ Non-Hispanic	Reference	Reference	
Other	.83	(.23-2.93)	.77
Lesion Level			
Upper	Reference	Reference	
Lower	.67	(.26-1.75)	.41
Household Income			
\$30,000 and below	Reference	Reference	
\$30,000 and above	1.44	(.57-3.65)	.45

Table 4.14 illustrates that all of the variables and co-variables (except for *Sex*) fail to meet the reliability criteria set for the p-values and 95% CI. Females again appear to be 60% less likely to report a vision complaint.

CHAPTER V DISCUSSION AND CONCLUSION

5.1 Discussion of Research Questions

This study supports the common idea that the procedure of VP shunting may cause problems with vision. (Gaston et al., 2005; Arling et al., 2007) The magnitude of the outcomes were surprising with the results of having a shunt increasing the likelihood of an individual in the sample having visual complaints by a factor of 298% and this relationship increased to 333% when confounders were added to the evaluation. Nevertheless, shunt revisions did not appear to have a significant relationship with vision complaints. Given the highly complex neuro-anatomy of the visual processing center of the brain and the nature of VP shunting as a procedure, perhaps these results are to be expected. (Macintyre-Beon et al., 2010) Conceivably, once a VP shunt is introduced subsequent shunt revisions make little difference on the vision complaints, which would suggest that the major impact to the visual processing center occurs with the initial procedure. These findings are among the first in the literature to demonstrate this relationship on the scale of a state-level sample.

5.2 Study Strengths and Limitations

The main strength of the study was the design of the Arkansas Spina Bifida Questionnaire. The questionnaire was set up over a 12-year period and was consistent in the participation of the children with spina bifida and their parents. The data set had very detailed questions about vision and possible vision complaints, which made it ideal for this type of investigation. The design of this study also makes it easy to reproduce and compare with future related research. Cross sectional designs as a whole have the ability

to be used as a descriptive tool or analytical tool. This cross sectional study was more of a descriptive tool and could be used to direct a cohort study with the information provided in the results section.

The main limitation of the project was the small sample size, which limited the power of the computations and made this endeavor more of a descriptive tool.

Throughout the investigation the 95% CIs were relatively large and p-values were generally not significant. Nevertheless, this study still serves a purpose by implying a direction for further investigation.

5.3 Implications of Findings

The major implications of the findings imply that there may be some relationship between shunts and vision issues among patients with the co-morbidities of spina bifida and hydrocephalus. The significance of this relationship is very important to the medical, social, and public health professionals treating individuals with spina bifida and hydrocephalus.(Caines et al., 2007; Aring et al, 2007; Macintyre-Beon et al, 2010; Holgrove et al., 2009) Having this information may result in more of a wait and see response or more attention to the visual functionality of individuals with shunts. (Margaron et al, 2010; Matson et al., 2005) For example, it may be effective to coordinate vision care before, during, and after treatment of hydrocephalus.(Bowen & McLone, 2010) So far, only one article suggested the need of a visual specialist to co-manage patients with hydrocephalus on a routine basis and in synergy with the treating neurosurgeon. (Caines et al., 2007) If this relationship can be substantiated, then the next move would support a need to research current and future treatments for hydrocephalus. Finding a better and less physician-dependent treatment plan for treating hydrocephalus

could improve patient mobility, self-image, and overall decrease medical expenditures.(Garton et al., 2002) On the other hand, reducing the build up of fluid in the brain is mandatory to save a person's life. (Bowen & McLone, 2010) So, the answer to this question is very complex and more research is needed to determine what constitutes optimal treatment.

5.4 Recommendations for Future Research

After reviewing the available literature on shunts, it is apparent that the evidence of the actual causes of visual compromise is still unknown. It is very hard to distinguish if visual problems are caused by hydrocephalus or the addition of the shunts. Directionality of the cause in this scenario is also an issue. Only a few research teams (Altintas, et al., 2005; Gaston et al., 1991; Rabinowicz et al., 1976) have addressed the issue of directionality by using pre- and post- surgical visual examinations. None of the research papers reviewed had a comparison of visual complaints before having hydrocephalus, while suffering from the conditions, and comparisons of the complaints after VP shunt procedures versus endoscopic third ventriculostomy. Ideally, a cohort study design would help evaluate which of these aspects is the cause, the exasperator, or the cure.

5.5 Conclusion

The increase of visual complaints after the introduction of a VP shunts and the increase of shunt revisions has not been fully investigated and larger more detailed studies are needed to truly investigate the link between visual complaints and shunts. Nevertheless, if and when the side effects of shunts and the increased likelihood of revisions can be proven to limit the quality of life for individuals with spina bifida and hydrocephalus, then more research should go into fine-tuning less invasive procedures to

address the issue. On the other hand, if further research on the VP shunts reveals that it is the safest and effective way to alleviate the build of spinal fluid in the brain, then attention should go into providing vision services and having a vision specialist on staff at clinics that service patients with spina bifida and hydrocephalus.

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