ADDITIONAL CHILDREN IN FAMILIES OF CHILDREN WITH DISABILITIES: USING DEMOGRAPHIC DATA TO ANSWER CLINICAL QUESTIONS

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Thesis

Submitted to the Faculty of the

Graduate School of Vanderbilt University

in partial fulfillment of the requirements

for the degree of

MASTER OF SCIENCE

in

Special Education

May, 2009

Nashville, Tennessee

Approved:

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CHAPTER I

INTRODUCTION

Statement of the Issue

Upon giving birth to a newborn with disabilities, parents most often experience grief and denial (Solnit & Stark, 1961), but much less is known about how the child with disabilities influences the parents' subsequent reproductive decisions. Specifically, when parents have a child with disabilities, do they then proceed to have another child or do they stop?

Although several ideas have been put forward to explain subsequent reproductive choices among parents of children with disabilities, so far the most prominent theory has concerned the so-called "replacement child" (Cain & Cain, 1964). The replacement child theory discusses how a parent has another child as a substitute for a sibling who has died (Poznanski, 1972). This subsequent child is referred to as a "replacement child" (Cain & Cain, 1964), as parents mourn the absence of the child who died by trying to replace the child with a subsequent pregnancy. While the replacement child theory has been developed and discussed throughout the psychoanalytic literature, little quantitative evidence exists to support its validity.

Replacement child theory may also apply to parents of children with disabilities. Parents of children with disabilities feel the loss of a child who may never be able to live up to their expectations or ideals (Solnit & Stark, 1961). In combination with mourning, parents of children with disabilities may prepare for the future by having a subsequent

pregnancy. As siblings of individuals with developmental disabilities are frequently the future caregivers for their brothers and sisters (Hodapp, Gilden & Kaiser, 2005; Seltzer, Begun, Seltzer, & Krauss, 1991), parents of children with disabilities may be demonstrating foresight by having additional subsequent children who can help care for the individual with disabilities.

In applying replacement child theory to families of children with disabilities, there are many testable hypotheses. For example, if parents, in comparison to families of children without disabilities, are indeed "replacing" their children with disabilities, then they should more often have subsequent children. Having a subsequent child should especially occur among families in which the child with a disability is a first-born child. The percentages of such "replacement children" (i.e. the non-disabled child in the family) should decrease with second-born, third-born, and so on.

Overview of the Paper

While replacement child theory may answer some presently unanswered questions, the theory has rarely been tested among families of children with disabilities using a large-scale database. This study will use a large-scale database to apply replacement child theory to families of children with disabilities. First, I will examine the mourning process and subsequent replacement child theory discussed in the psychoanalytic literature. Next, I will discuss the application of replacement child theory to families of children with disabilities by focusing on the grieving process of the parents upon receiving the diagnosis of disability and the role of the sibling as a future caregiver. I will then discuss the arguments both for and against applying replacement child theory

to families of children with disabilities. Finally, I will discuss the advantages of using large-scale databases in trying to examine whether families of children with disabilities do indeed have a replacement child.

CHAPTER II

REVIEW OF THE LITERATURE

Mourning and Replacement Children

Among family members who have lost a loved one, mourning is a common reaction. Similarly, mourning can also occur after the loss of an abstract concept, after the parents learn that their child has a disability (Lehrman, 1956). Mourning or bereavement is a process with several stages, such as denial and anger, eventually leading to acceptance (Valeriote & Fine, 1987). Mourning is typified by the following characteristics: depression, loss of appetite, sleeplessness, anxiety, lethargy, and withdrawal (Valeriote & Fine, 1987; Zeenah, 1988). These feelings of guilt and anger occur regardless of whether the child died prenatally or postnatally (Peppers, & Knapp, 1980). Essentially, these parents mourn "what might have been".

One reaction after a child dies is to have another child or a "replacement" child. A replacement child is a child used by the parents as a substitute for this child who died (Poznanski, 1972). Physically, the subsequent child actually replaces or supplements the deceased child. Metaphorically, the replacement child replaces or fulfills the parent's lost dreams for the child who died. Parents of children who have passed away have a higher likelihood of having another child compared to parents of children who have not died (Legg, & Sherick, 1976; Johnson, 1984).

There are, however, positive and negative aspects of having a replacement child for both the parents and replacement children themselves. On the negative side, the

replacement child may disrupt and even prolong the mourning process for the parents (Valeriote & Fine, 1987). One reason for a longer mourning process may be that the replacement child then becomes the vessel for ongoing grief or even indefinite mourning regarding the lost child (Powell, 1995). Some professionals even explicitly tell parents not to have another child, as it will elongate their mourning process (Grout & Romanoff, 2000). Instead, parents are often counseled to complete the grieving process before having another child (Valeriote & Fine, 1987; Grout & Romanoff, 2000).

In addition to the parents, the replacement child may also have his or her own negative experiences. Beginning in early development, the replacement child may feel the "psychological weight" of existing mostly to fulfill the lost parental expectations of the sibling who died (Legg & Sherick, 1976). These feelings may continue as the child matures, affecting the child cognitively (Legg & Sherick, 1976; Valeriote & Fine, 1987), behaviorally (Valeriote & Fine, 1987) and emotionally (Legg & Sherick, 1976; Valeriote & Fine, 1987). As the child understands more about his compromised identity, the child may be more at risk for psychopathology (Grout & Romanoff, 2000; Johnson, 1984). Furthermore, parents who have not completed the grief cycle may have diminished parenting skills, leaving the replacement child essentially parentless (Poznanski, 1972; Valeriote & Fine, 1987). Alternatively, having already lost a child, the parents may become over-protective of the replacement child (Cain & Cain, 1964; Cornwell, Nurcombe, & Stevens, 1977; Zeenah, 1988).

In contrast to negative aspects, positive aspects of the replacement child have also been reported. Positive aspects include: having another child may help the family fill the sibling's void (Johnson, 1984); and the parents have a reason to live (Grout, & Romanoff,

2000) and to enjoy life again (Powell, 1995). Furthermore, Phipps (1985) disagrees that the subsequent child is a replacement child. In this study, the subsequent child was conceived because the parents realized they were running out of time to have subsequent children (Phipps, 1985). Having lost a child, though, does seem to affect the pregnancy of the subsequent child. According to one study, parents were more likely to refuse a baby shower, baby announcements, or any kind of publicity for the subsequent pregnancy (Phipps, 1985). Such behavior may imply a self-protective function of the couple to help guard against another public loss of a child.

Subsequent Children After a Child with Disabilities

The idea that a mother envisions an image of her unborn child also applies to mothers of children with disabilities. Prior to receiving a disability diagnosis, the mother develops a persona or abstraction of what her child will be like and what expectations the child will meet. Similar to parents whose children have died, parents of children with disabilities frequently experience mourning after learning that their children have disabilities and, therefore, may not fulfill their goals (Solnit & Stark, 1961). Similar to losing a child, the idealization of a certain child tends to be abruptly halted by the birth of a child with a disability (Solnit & Stark, 1961).

Many characteristics are shared by the mourning processes of losing a child and of having a child with a disability (Solnit & Stark, 1961). In both situations, the mothers experience a feeling of loss, resentment of losing the idealized child, and guilt. Furthermore, parents experience severe anxiety, denial, shock, and near chronic sadness (Olshansky, 1962; Wikler, Wasow, & Hatfield, 1981). These symptoms are also present

when losing a child. Additionally, mothers of infants who died and mothers of infants with disabilities may have similar durations of mourning, lasting from 4-6 weeks (Kennedy, 1970).

While parents of children who have died and parents of children with disabilities experience similar affective symptoms, parents of children with disabilities experience additional feelings related to their child's disability. For example, parents of children with disabilities may experience physical agitation, muscle tension, and fatigue (Epperson, 1971). Having a child with a disability also affects the behavior of the parents. For example, parents of children with disabilities seek information related to their child's disability (Fortier & Wanlass, 1984). While both parents of children with disabilities and parents of children who have died share similar mourning processes, parents of children with disabilities face additional unique experiences.

One area that may be either similar or unique concerns whether parents of children with disabilities are more likely to have subsequent children. First, I will discuss studies and theoretical constructs that support the premise that families of children with disabilities are indeed more likely to have subsequent children than families of children without disabilities. Then, I will discuss a few studies and other theories, which support why families of children with disabilities may not have subsequent children especially in relation to families of children without disabilities.

Argument for why Parents of Children with Disabilities Have Subsequent Children

There are several reasons why parents of children with disabilities might have subsequent children. This section lists the results of small-scale studies that imply that

parents of children with disabilities are more likely than parents of children without disabilities to have subsequent children. Furthermore, this section also details theories such as the role of the sibling and inclusive fitness, which also support the premise that parents of children with disabilities having subsequent children.

Small- scale studies.

To date, only a few studies exist regarding the replacement child in families of children with disabilities. For example, one study compared the pregnancy rates of mothers of 3,029 control infants to 4,918 infants with birth defects born between 1968 and 1980 (Davis, Khoury, & Erickson, 1995). In comparison to mothers of children who lived, mothers of children who died were more likely to have subsequent children thus providing evidence of replacement theory. Furthermore, the results suggested that, depending on the severity of the birth defect, mothers of children with certain birth defects would be more likely to have subsequent children than mothers of children without birth defects. For example, mothers of surviving children with surgically correctable conditions may be more likely to have a subsequent child in comparison to mothers of children without disabilities. This study suggests that mothers of children with certain disabilities may be more likely to have subsequent children than mothers of children without disabilities.

Small-scale studies have also applied replacement child theory to families of children with Down syndrome. For example, Fraser and Latour (1967) compared the reproduction rates in families of children without disabilities to 45 families of children with Down syndrome in Montreal. Their results indicated no decline in reproduction

among families of children with Down syndrome. While Fraser and Latour's findings do not explicitly support the replacement theory, they do counter the concept that reproduction halts after having a child with a disability.

The role of the sibling.

The presence of the subsequent non-disabled sibling may also help explain why parents of children with disabilities might have subsequent children. As persons with disabilities live longer lives in the community, their siblings are generally turned to as the future caregivers (Hodapp, Gilden & Kaiser, 2005; Seltzer, et. al., 1991). For example, in a study of mothers of individuals with disabilities, the non-disabled child was anticipated to fulfill a caregiving role for the child with a disability (Pruchno, Patrick, & Burant, 1996). Similarly, Greenberg, Seltzer, Orsmond, & Krauss (1999) also found that siblings of persons with intellectual disabilities are likely to provide both emotional support and future caregiving to their brothers and sisters.

Throughout the literature, it is acknowledged that female siblings most often fulfill future guardianship and caretaking roles for their brothers or sisters with disabilities (Krauss, Seltzer, Gordon & Friedman, 1996; Zetlin, 1986; Cook, Cohler, Pickett, & Beeler, 1997; Griffith & Unger, 1994). In study, for example, parents of children with disabilities reported that the non-disabled sisters were more likely than brothers to take on future caregiving roles (Griffith & Unger, 1994). Furthermore, for sisters, the gender of the sibling with a disability does not seem to matter in terms of future caregiving or closeness. In contrast, brothers judge themselves to have more positive emotion, less negative emotion, and less worry in relation to their brother (as

opposed to sister) with disabilities (see also Seltzer, Begun, Seltzer, & Krauss, 1991). According to a national sibling survey, compared to male siblings, female siblings spend more time with their siblings, are closer to their siblings, and feel more positive effects related to having a sibling with a disability (Hodapp, Urbano, & Burke, in submission).

Realizing that siblings fulfill future caregiving roles and responsibilities, national efforts have been made to provide additional support and knowledge to these siblings. For example, a national Sibling Leadership Network has developed "to provide siblings of individuals with disabilities the information, support, and tools to advocate for their brothers and sisters and to promote the issues important to them and their entire families" (Heller, Kaiser, Meyer, Fish, Kramer, & Dufresne, 2008, p. 5). The Sibling Leadership Network has an annual conference for adult siblings to learn more about their roles as future caregivers. After having a child with a disability, parents may consciously opt to have a subsequent child to fulfill these future caregiving roles. Considering the future needs of their children with disabilities in choosing to have a subsequent child reflects foresight on behalf of parents of children with disabilities.

Inclusive fitness.

In addition to wanting their children with disabilities to have siblings who can fulfill future caregiving roles, parents may also have subsequent children to carry on their progeny. To explain this concept, Hamilton (1975) developed the theory of inclusive fitness. Inclusive fitness is the theory that individuals want to procreate so that their genes can live on through their offspring. This theory applies not only to the parents but also to

their kin, including their children. Parents will also want to ensure that their children go on to have the next generation of children.

The concept of inclusive fitness is interesting in the context of children with disabilities. For example, few adults with Down syndrome go on to have children; there have only been three instances of a father with Down syndrome having a child (Pradhan, Dalal, Khan, & Agrawal, 2006). Furthermore, individuals with Down syndrome have a 50% chance of having a child with Down syndrome (Hsiang, Berkovitz, Bland, Migeon, Warren, et al., 1987). As such, according to the inclusive fitness principle, parents of children with Down syndrome should have additional (non-Down syndrome) children, as their children with Down syndrome are: (1) very unlikely to reproduce and (2) even if they do reproduce, they are likely to have another child with Down syndrome, who is in turn not likely to reproduce.

Reproduction rates are also lower for individuals with spina bifida (the other group in this study) than individuals without disabilities. Laurence and Beresford (1975) examined 51 adults with spina bifida, of whom 36% had children. This finding varies according to the marital status of the individual in that of the 22 individuals with spina bifida who were married, 81% of them had at least one child. The reproduction rates of individuals with spina bifida, though moderated by marital status, still are less than individuals without spina bifida.

The concept of inclusive fitness may also help explain why reproduction rates of parents of children with disabilities may differ according to the disability. For example, the parent of a child with a physical disability (e.g. spina bifida) may be less likely than the parent of a child with an intellectual disability (e.g. Down syndrome) to have a

subsequent child. But, according to inclusive fitness theory, both groups of parents would be more likely to have a subsequent child than parents of children without disabilities.

Arguments for why Parents of Children with Disabilities may not Have Subsequent Children

While there are several theories and a few studies, which support parents of children with disabilities having subsequent children, there are also some theories and small-scale studies which support the opposite. In comparison to parents of children without disabilities, parents of children with disabilities are less likely to have subsequent children. This section discusses the results of a few small-scale studies, which imply that parents of children with disabilities do not have subsequent children. This section offers a few reasons why parents of children with disabilities may opt not to have subsequent children.

Small-scale studies.

In direct contrast to the abovementioned reasons for having subsequent children after a child with a disability, several small-scale studies find that families of children with disabilities, in comparison to families of children without disabilities, do not go on to have subsequent children. For example, in a study of 24 women with previously high reproductive rates who then had a child with Down syndrome, reproduction stopped or sharply declined after having a child with Down syndrome (Tips, Smith, Perkins, Bergman, & Meyer, 1963).

Ando and Tsuda (1975) replicated Tips and colleagues' findings in Japan. Ando and Tsuda studied the reproductive practices across four groups: 119 families of children

with autism, 292 families of children with cerebral palsy, 146 families of children with Down syndrome, and 128 families of kindergarteners without disabilities. Parents of children with cerebral palsy and Down syndrome were more likely to stop having children than parents of children with autism or parents of children without disabilities. In a follow-up study of 146 children with Down syndrome in comparison to 128 kindergarteners without disabilities, children with Down syndrome were likely to be the "only-child" in the family for all mothers aged under 30 (Ando, 1978). Furthermore, children with Down syndrome were likely to be the last-born child for mothers under age 35 providing evidence that reproduction ceases after having a child with Down syndrome.

While these studies found that parents of children with Down syndrome did not continue to have children, these studies were compromised by several methodological issues. In all of these studies, the samples tended to be small and (possibly) biased in terms of who volunteered to participate. As such, the participants may not be representative of the population. Also, for both of Ando's studies, the participants were kindergarteners, allowing only a four-year window to determine if any subsequent children were born. Subsequent children born after this window were not included in the study. Furthermore, these studies date back to the 1960s and 1970s. Since then service delivery, quality of life issues, and policies have been created to provide equity and protection to individuals with disabilities and their families. As such, present-day families may not feel so overwhelmed by their child with a disability and may continue to have subsequent children.

Higher cost in caring for children with disabilities.

While services for individuals with disabilities and their families have improved since the 1970s, families of children with disabilities are still subject to more frequent hospitalizations, more stress, and higher fiscal costs than families of children without disabilities. For example, 50% of children with Down syndrome are hospitalized (i.e. non-birth hospitalizations) at least once from the age of 0 until the age of 3 (Hodapp, Urbano, & So, 2008). Furthermore, in comparison to individuals without Down syndrome, individuals with Down syndrome are more likely to have congenital heart defects, leukemia, obesity, hearing and/or vision problems, dementia and seizures (Roizen & Patterson, 2003). Facing this plethora of medical issues, families may feel high levels of stress and incur greater fiscal costs, which may dissuade them from having another child.

In addition to the parental stress caused by the medical conditions children with disabilities face, families of children with disabilities incur other sources of stress. For example, Singer (2006) conducted a meta-analysis of 18 studies comparing levels of depression of mothers of children with disabilities to mothers of children without disabilities. Somewhat higher levels of depression were noted for mothers of children with disabilities. Furthermore, it seems that maternal distress decreases as the child with a disability reaches adulthood (Glidden & Schoolcraft, 2003). The highest levels of maternal distress may occur during the mother's childbearing years, providing another reason for families of children with disabilities to stop having children. Facing such a great deal of stress, families of children with disabilities may feel that having another child would simply add more stress to their households.

Finally, families of children with disabilities incur higher fiscal costs than families of children without disabilities. For families of children with intellectual disabilities, the poverty cycle is twofold (Emerson, 2007). First, families who live in poverty are more likely to have a child with an intellectual disability. In comparison to families of children living above the poverty level, families living in poverty are more likely to face environmental toxins and worse medical care, which result in an increased likelihood of having a child with a disability. Second, caring for a child with an intellectual disability imposes a large fiscal burden upon the family. Spending money on services, childcare, medical care and other resources necessary for a child with a disability are likely to impose a financial burden on the family. Realizing the extra costs in caring for a child with a disability, parents may refrain from having another pregnancy as the resources are not present to care for another child.

Databases

Across the literature, virtually every article about replacement children and parent mourning is derived from the psychoanalytic research. None of these studies rely on large, quantitative databases to test the replacement child theory. To determine whether parents of children who have died are in fact having a subsequent child, it may be necessary to rely on a large database. Such databases, for example, might include all of the state's births, or include information from a large-scale national survey.

There are many advantages to using such large-scale databases. First, these databases allow researchers either to study an entire population or at least to have an almost entirely representative sample of the population. Birth records are not samples of

convenience, but rather are multi-source datasets allowing researchers to track subjects over a longer period of time and examine family issues (Tu & Mason, 2006). In contrast, a sample of convenience involves a datasource that may be biased toward a particular group. For example, using parent support groups as participants may be biased in favor of actively involved parents. Furthermore, samples of convenience are usually too small to look at multiple variables. Additionally, small samples are more likely to be skewed and not reflect the general population. Unfortunately, samples of convenience are used in most studies and are typically geared toward a specific group. Relying on samples of convenience skews the data and may yield unrepresentative findings. In contrast, by relying on birth records, the researcher is more likely to have an unbiased pool of subjects. The sheer number of people in a large-scale database should improve the likelihood that the sample is more representative of the entire population.

Furthermore, the number of people in the database allows a more comprehensive understanding of the family dynamic over a span of years (Tu & Mason, 2006). Being able to look at the family dynamic over a long time span is especially helpful for the study of genetic syndromes in families. For example, the Utah Population Database Project tracks the initial Latter Day Saints and their followers by linking their births, deaths, and cancer registries (Tu & Mason, 2006). The Utah Population Database has been used to study preeclampsia (Esplin, Fausett, Fraser, Kerber, Mineau, et al., 2001), cancer (Boucher & Kerber, 2001), and familial melanoma (Florell, Boucher, Astle, Kerber, Mineau, Wiggins, et al., 2005). By having large-scale databases, families can be examined using psychosocial risk factors along with health and demographic variables.

An additional advantage of large-scale databases is the increased likelihood that the number and accuracy of a disability diagnosis is correct. With any disability, including Down syndrome and spina bifida, there is potential for a participant to be mislabeled or missed by the records. However, by linking multiple records, mislabeling a disability can be avoided. For example, Urbano and colleagues (2007) linked the official Tennessee Birth Records with State's Hospital Discharge records. By linking across datasets, they were able to confirm most every participant's diagnosis of Down syndrome. Linking across multiple datasources is just one way in which disability diagnoses can be validated.

The Israeli National Down syndrome Birth Registry provides another example of a large-scale population database (Sadetzki, Chetrit, Akstein, Luxenburg, Keinan, Litvak, et al., 1999). Sadetzki and colleagues used multiple datasources to confirm the percentage of children with Down syndrome. Using three different sources, they confirmed the diagnosis of Down syndrome for 82% of cases. Sadetzki and colleagues then used cytogenetic analysis to confirm 91% of the diagnoses. This is one example of a large-scale database and the use of multiple datasource to increase the accuracy in diagnoses of disabilities.

Related to the accuracy of the diagnosis, another benefit of large-scale databases is their longitudinal nature. By spanning a long period of time, records can be linked together, thereby developing a complete medical history for each individual over a substantial period of time. In one study, Frid, Annerin, Rassmussen, Sundelin, and Drott (2002) used three different registries to examine the medical histories of 211 children with Down syndrome born between 1973 and 1980. Having multiple databases across a

longer time period allowed them to examine various correlates related to the child and sicknesses across time. The longitudinal nature of these databases allows for a much more complete understanding regarding each individual's medical history.

To achieve a more complete view of families using medical records, it may also be necessary to use second-order record linkage. Second-order linkages are organized around family structures, with linkage accomplished by matching records based on a set of identifying fields or characteristics (Tu & Mason, 2006). For example, using birth records, the researcher first creates families by linking records together using the social security number of the mother to collect a complete picture of all of the children the mother gave birth to within a specified time period. Researchers can then compare births over time or designate a target child and compare each target child and the subsequent and/or prior children.

This Study

This study hypothesizes that, compared to families of children without disabilities, families of children with disabilities are more likely to have a subsequent child after having a child with a disability. As such, families of children with disabilities (in this study, spina bifida and Down syndrome) are predicted to be more likely to have larger families regardless of the race, marital status, age, and educational attainment of the mother.

Using a large-scale database, this study addresses the issue of families having subsequent-or "replacement"-children in five ways:

1. This study looks at the descriptive differences across groups (i.e., families of children with spina bifida, families of children with Down syndrome, and families of children without disabilities).
2. This study examines at whether the family has a subsequent child after having a child with disabilities (Down syndrome or spina bifida) versus having a child without disabilities.
3. This study examines the family size across families of children with and without disabilities. Specifically, how does the total number of children compare in families of children without disabilities, have spina bifida, or have Down syndrome?
4. The study looks at the potential influence on having subsequent children or of other parent-family variables, including maternal education, age, and race, and
5. This study examines the influence of parent-family variables on family size across families of children with and without disabilities.

This study looks at three different groups: families of children without disabilities, families of children with spina bifida, and families of children with Down syndrome. Among these three groups, it is predicted that families of children with Down syndrome are more likely to have a subsequent child than families of children with spina bifida. Furthermore, families of children with spina bifida are more likely to have a subsequent child than families of children without disabilities. This prediction is grounded in evolutionary psychology, as individuals with spina bifida are more likely to reproduce than individuals with Down syndrome, but not as likely to reproduce as individuals

without disabilities (Laurence & Beresford, 1975; Hsiang et al., 1987). Furthermore, in comparison to individuals with spina bifida and individuals without disabilities, it is more likely that individuals with Down syndrome will require future caregiving due to their intellectual disabilities. As such, families of children with Down syndrome are expected to be more likely to have a subsequent child than these other two groups so the child can fulfill future caregiving roles.

CHAPTER III

METHOD

This study compared the birth records of children in Tennessee born between 1990 and 2006 across three groups: children without disabilities; children with spina bifida; and children with Down syndrome. This section begins by providing an overall description of the subjects. I discuss descriptive information regarding the first-born children across these three groups before discussing descriptive information regarding the second thru fifth born children across the three groups. Finally, I describe how these subjects were derived and the study's general procedures.

Participants

All families of children with Down syndrome, with spina bifida, and with no disabilities.

This study includes 728,957 families born in Tennessee between 1990 and 2006. Within this dataset, 727,563 (99.8%) families had all children who did not have an identified disability, 302 (<.01%) families had a child with spina bifida, and 1,092 (.2%) families had a child with Down syndrome. Table 1 shows the demographic information for each group.

Table 1. Family Demographics for all families

	Without disabilities (n=727,563)	Spina Bifida (n=302)	Down syndrome (n=1,092)	X^2	p
Mother Race: White	661,191(79.2%)	266(88.96%)	910 (84.34%)	34.93	.000
Non-white	174,092(20.8%)	33(11.04%)	169(15.66%)		
Mother marital status: Married	563,557(65.9%)	213(70.5%)	777(71.2%)	3.13	.209
Not married	291,981(34.1%)	89(29.5%)	391(28.8%)		
Child: Male	438,073(51.2%)	158(52.3%)	599(52.5%)	.874	.646
Female	417,635(48.8%)	144(47.7%)	543(47.5%)		

Families for which the target child was the 1st born.

Part 1 of this first study includes a total of 352,672 families. All families were of first-born children born in Tennessee between 1990 and the beginning of 2006. Within the 352,672 families, 124 families had children with spina bifida, 385 families had children with Down syndrome, and the remaining 352,163 families had children with no identifiable disability. The families of children without disabilities were randomly selected from the larger dataset to be included in this study. Approximately 78.6% (277,221) of the mothers of first-born children were White and18.8% (66,275) of the mothers were non-Caucasian. Regarding first-born children, 48.8% (172,101) of the children were female and 51.2% (180,569) were male (See Table 2).

Table 2. First-born Demographics

	Without disabilities (n=352,163)	Spina Bifida (n=124)	Down syndrome (n=385)	X^2	p
Mother Race: White	276,789 (80.7%)	108 (88.5%)	324 (85.0%)	9.398	.009
Non-white	66,204 (19.3%)	14 (11.5%)	57 (15.0%)		
Marital Status: Married	209,500 (59.5%)	77 (62.1%)	244 (63.4%)	2.743	.254
Not married	142,597 (40.5%)	47 (37.9%)	141 (36.6%)		
Child: Male	180,288 (51.2%)	64 (51.6%)	217 (56.4%)	4.124	.127
Female	171,873 (48.8%)	60 (48.4%)	168 (43.6%)		

Families for which the target child was 2nd thru 5th born.

This study also included a total of 376,285 families of second through fifth born children born in Tennessee between 1990 and the beginning of 2006. Within the 376,285 families, 178 families had a child with spina bifida, 707 families had a child with Down syndrome, and the remaining 375,400 families had children without identifiable disabilities. Among these non-first-born children, 32% (235,837) of the target children were the second-born, 13% (98,899) of the children with the third-born, 4% (31,330) of the children were the fourth-born and the remaining 1% (10,219) were the fifth-born child in their families. Similar to the first-born children, the children without disabilities were randomly selected from the larger dataset to be included in this study (see Table 3).

Table 3. Post First-Born Demographics

	Without Disabilities (n=375,400)	Spina Bifida (n=178)	Down syndrome (n=707)	X^2	p
Mother Race: White	384,402 (78.1%)	158 (89.3%)	586 (84.0%)	26.95	.000
Non-white	107,888 (21.9%)	19 (10.7%)	112 (16.0%)		
Marital status: Married	354,057 (70.3%)	136 (76.4%)	533 (75.4%)	11.81	.003
Not married	149,384 (29.7%)	42 (23.6%)	174 (24.6%)		
Child: Male	257,785 (51.2%)	94 (52.8%)	382 (50.5%)	.348	.840
Female	245,762 (48.8%)	84 (47.2%)	375 (49.5%)		

Source of Data

For this study, I used the official Birth Records from the state of Tennessee. These records include children born from 1990 until 2006.

The Birth record of each child born in Tennessee contains nearly 150 variables. These data come from two sources: self-report by the mother and information collected by a birth clerk trained by the Tennessee Department of Health. The birth records include the following information:

- Maternal factors: age; race; marital status; education; home address; county; state; prior live births; inter-delivery interval from last birth;
- Prenatal practices: mother's weight gain; number of doctor's visits; months during pregnancy when prenatal visits began; alcohol use;

- Newborn characteristics: gender; birthweight; gestational age; APGAR scores at 1 and 5 minutes; birth complications; abnormal conditions; and congenital abnormalities.

The maternal factors come from the questionnaire the mother completes. The birth clerk's information comes from the hospital records. These records include information about: prenatal care, risk factors and complications during pregnancy, details about the delivery, and newborn characteristics. Within the birth clerk's information is information regarding whether the child has spina bifida or Down syndrome.

Hospital discharge records were minimally used in this study. These state records record any in- or out-patient hospitalization in Tennessee between 1997 and 2005. The records include the diagnosis, procedures, demographics, duration of stay, and insurance of the patient. Of the 9 potential diagnoses to be listed, one of the diagnoses can be "Down syndrome". In another study (Urbano, Hodapp, & So, 2008) these records were used to confirm that the individuals had Down syndrome and spina bifida to be included in this study.

Procedures

Prior to beginning this study, the State of Tennessee Department of Health and the Vanderbilt Kennedy Center entered into a contract to gain access to the health records for the subjects in this study. Furthermore, an application was submitted and approved by the Vanderbilt University Institutional Review Board. Within this application, necessary procedures were included to protect the safety of the data and storage of the data.

According to the official Tennessee Birth Records, over 1.3 million children were born in Tennessee between 1990 and 2006. For each child, the mother completed a questionnaire. Recorders used this questionnaire along with hospital records to complete a birth record, which details maternal factors, prenatal health practices, and infant descriptives. This study relied on these birth records to identify the participants.

Using second-order linkage based on the mother's social security number, Dr. Urbano formed families of children born between 1990 and 2006. From the initial 1.3 million children, 728,957 families of the children born in Tennessee between 1990 and the beginning of 2006 were controls. By having a side-by-side linkage of children born in the same families, we have a more complete view of family demographics during this time period. As such, we were able to determine how many of these children were first-born, second-born, and so on.

In addition to knowing the birth order of these children, we also identified whether the child had Down syndrome, spina bifida, or no identifiable disability. To identify whether the child had either of these disabilities, we looked at the Birth Record variable labeled "Congenital anomalies of the newborn". This variable lists 13 anomalies, including both Down syndrome and spina bifida. The absence of a diagnosis in these codes indicated that the child does not have a disability. From the families of children without disabilities, a target child was randomly selected for this study.

For this study, if a child had Down syndrome or spina bifida, regardless of the birth sequence of the child, the child was included in this study. The family of this child was then included in this study with the target child being the child with the disability and the target child having the birth order of the child with disabilities. If the family did not

have a child with an identifiable disability, then we randomly chose a target child. In single-child families, the first child was the target child. In two-children families, either the first or second child was the target child, and so on.

Given that both older mothers and mothers giving birth later in the study period both have less "time" to have subsequent children, we ran separate analyses for these groups. All of the analyses were run by looking at the groups overall, looking at mothers older than 35 (Old Ms) and mothers younger than 35 (Young Ms), children born prior to 2000 (Datayr ≤2000) and children born after 2000 (Datayr >2000), and combining maternal age and the year of the child's birth (Old Ms, early; Young Ms, early; Old Ms, late; Young Ms, late). By looking at the groups overall, we were able to detect if there were significant differences. We then divided the groups by maternal age as mothers above the age of 35 have less time to have a subsequent child than mothers younger than 35. We also divided the birth years of the child as mothers of children born prior to 2000 had more time, within our dataset, to have a subsequent child. For children born after 2000, these mothers have less time to have a subsequent child included in our dataset.

Once the linked dataset was complete, all identifying information or Protected Health information was removed (PHI). This de-identified dataset was then given to the principal investigator who ensured the accuracy of the data and cleaned the data. For example, the intervals between the births of the individuals were converted into years instead of days. Furthermore, any absence of a disability label for the children in the dataset were re-coded to be labeled as "non-disabled". This final, de-identified and cleaned dataset was then used for all of the analyses in SPSS.

CHAPTER IV

RESULTS

The results section is divided into five parts. The first subsection discusses the descriptive differences across groups of children without disabilities, families of children with spina bifida and families of children with Down syndrome. The second subsection examines the subsequent children among these different groups for the first, second, third, and fourth-born child. The third subsection examines the total number of children across families of children without disabilities, families of children with spina bifida, and families of children with Down syndrome. The fourth subsection examines the influence of other parent-family variables upon subsequent children across the three groups. Finally, the fifth subsection examines the parent-family variables upon the family size across groups.

I. Descriptive Differences Across Groups

Family size.

There were significant differences in the family size of families of children with disabilities in comparison to families of children without disabilities, $X^2(2, N=728,957) = 36.88$, $p<.001$. In comparison to families of children without disabilities (59.6%), proportionally fewer families of children with Down syndrome (42.5%) and spina bifida (48.0%) were single-child families.

Maternal age.

Across the three groups, there were significant differences in the mother's age at the birth of each child. As Table 4 illustrates, mothers of children with Down syndrome were on average older than mothers of children with spina bifida and mothers of children without disabilities. The increased maternal age of mothers of children with Down syndrome held true regardless of the birth order of the child (see Table 4).

Table 4. Maternal Age by Group

	Without disabilities		SB		DS		F	p
	Mean	sd	Mean	Sd	Mean	sd		
Child 1	23.88	5.66	23.63	5.42	26.69	7.92	47.41	.000
Child 2	26.81	5.46	27.10	6.03	29.67	6.65	51.51	.000
Child 3	28.23	5.45	28.79	5.57	32.47	6.80	62.52	.000
Child 4	29.20	5.52	27.86	4.85	34.01	6.84	36.84	.000
Child 5	30.13	5.53	31.40	5.48	35.07	6.39	11.02	.000

Mother's marital status.

The marital status of the mother did not significantly differ across the groups regardless of the birth order of the child (see Table 5).

Table 5. Marital Status by Group

	Without disabilities N(%)	SB N(%)	DS N(%)	X^2	p
Child 1: Married	59.5% (209,500)	62.1% (77)	63.4% (244)	2.74	.254
Not Married	40.5% (142,597)	37.9% (47)	36.6% (141)		
Child 2: Married	74.5% (175,387)	77.2% (78)	78.2% (295)	3.13	.209
Not Married	25.5% (59,935)	22.8% (23)	21.8% (82)		
Child 3: Married	71.7% (70,696)	78.4% (40)	73.7% (151)	1.53	.465
Not Married	28.3% (27,915)	21.6% (11)	26.3% (54)		
Child 4: Married	65.7% (20,495)	57.1% (8)	69.5% (66)	1.06	.588
Not Married	34.3% (10,717)	42.9% (6)	30.5% (29)		
Child 5: Married	58.3% (5,937)	-----	68.0% (17)	.961	.418
Not Married	41.7% (4,243)	-----	32.0% (8)		

Note: In the spina bifida group, there were sometimes too few subjects to perform analyses (N<10); in these cases, chi-squares compare the families of children without disabilities to the DS groups.

Birth intervals.

Across the groups, birth intervals between each of the children (e.g. the time between the first-born child's birth and the second-born child's birth) across the groups did not significantly differ regardless of the birth order of the target child (see Table 6). Across all groups and children of each birth order, mothers had their subsequent child roughly 2-4 years after their prior child.

Table 6. Birth Intervals in Years

	Without disabilities Years (sd)	SB Years (sd)	DS Years (sd)	F	p
Child 1	3.15 (2.09)	2.80 (2.49)	3.05 (2.06)	.855	.425
Child 2	2.96 (2.16)	2.84 (2.15)	2.58 (1.87)	1.66	.190
Child 3	2.58 (2.04)	2.25 (.957)	2.42 (1.63)	.176	.839
Child 4	2.36 (1.98)	3.00 (1.73)	2.69 (1.98)	.374	.688

II. Subsequent Children

Regardless of maternal age or year the child was born, families of children with disabilities were more likely to have a subsequent child than families of children without disabilities (see Table 7). Furthermore, when mothers were younger than 35, families of children with Down syndrome were significantly more likely to have a subsequent child than families of children with spina bifida, $X^2(1, n=424) = 8.22$, $p<.005$.

Table 7. First-born child: Subsequent children across groups

	Without disabilities %(n)	SB % (n)	DS %(n)	X^2	p
Overall	28.8% (101,316)	37.1% (46)	45.7% (176)	58.03	.000
Old Ms (≥35)	12.4% (2,631)	----------	20.0% (16)	4.20	.059
Young Ms (<35)	29.8% (98,685)	37.0% (44)	52.5% (160)	77.52	.000
Datayr (≤2000)	40.0% (75,249)	45.9% (39)	57.7% (135)	31.62	.000
Datayr (>2000)	15.9% (26,067)	----------	27.2% (41)	14.33	.000
Old Ms, early	15.5% (1,778)	----------	26.7% (12)	4.284	.059
Young Ms, early	41.6% (73,471)	45.8% (42)	65.1% (123)	43.36	.000
Old Ms, late	8.8% (853)	----------	11.4% (4)	0.300	.545
Young Ms, late	16.3% (25,214)	----------	31.9% (37)	20.55	.000

Note: In the spina bifida group, there were sometimes too few subjects to perform analyses (N<10); in these cases, chi-squares compare families of children without disabilities to the DS groups.

In comparison to families of children where the second-born child did not have a disability, families of second-born children with disabilities were significantly more likely to have another child, $X^2(2, n=235,837) = 40.30$, $p<.001$. Families of third-born children with Down syndrome were also more likely than families of third-born children without disabilities to have subsequent children, $X^2(1, n=98,848) = 6.22$, $p<.014$. This trend stopped at the fourth-born child where there was no significant difference between the groups, $X^2(1, n=31,316) = 2.40$, $p<.148$.

III. Family Size Across Groups

In comparison to families of children without disabilities, families of children with disabilities had larger families, as indicated by the mean number of children in their families. Table 8 shows that families of children with Down syndrome and spina bifida

tended to be bigger than families of children without disabilities. This finding was particularly true for mothers younger than 35 (Young Ms).

Table 8. Family size across groups

	Without disabilities Mean(sd)	SB Mean(sd)	DS Mean(sd)	F	P
Overall	1.24(.534)	1.52(.108)	1.41(.651)	16.79	.000
Young Ms (<35)	1.24 (.537)	1.54(.77)	1.45(.667)	19.76	.000
Old Ms (≥35)	1.09(.325)	1.00(.000)	1.06(.243)	.135	.874
Datayr (≤2000)	1.29(.581)	1.62(.795)	1.48(.058)	14.87	.874
Datayr (>2000)	1.10(.326)	1.00(.000)	1.19(.401)	2.39	.091
Old Ms, early	1.09(.336)	1.00(.000)	1.08(.277)	.057	.945
Young Ms, early	1.29(.585)	1.63(.799)	1.52(.716)	17.08	.000
Old Ms, late	1.07(.302)	1.00(.000)	1.00(.000)	.141	.868
Young Ms, late	1.10(.326)	1.00(.000)	1.21(.415)	2.90	.055

Across the remaining second-born and third-born children, families of children with disabilities were again more likely than families of children without disabilities to have bigger families (see Table 9). This trend, however, stopped at the fourth-born child in which there were no significant differences in family size across the groups.

Table 9. Family size across groups for subsequent children

	Without disabilities Mean(sd)	SB Mean(sd)	DS Mean(sd)	F	P
Child 2	2.20 (.504)	2.32 (.582)	2.39 (.684)	28.093	.001
Child 3	3.15 (.415)	3.08 (.337)	3.21 (.475)	3.003	.050
Child 4	4.18 (.322)	4.21 (.426)	4.17 (.376)	1.834	.160

IV. Subsequent Children and the Influence of Parent-Family Variables

Marital status.

Regardless of marital status, families of children with disabilities were more likely to have more than one child in comparison to families of children without disabilities. Among unmarried mothers, 44.0% of families of first-born children with Down syndrome had one or more subsequent children, in comparison to 31.9% of families of first-born children with spina bifida and 26.6% of families of first-born children without disabilities, $X^2(2, n=142,785) = 22.48$, $p<.001$. Unmarried mothers of first-born children with disabilities were more likely to have a subsequent child in comparison to unmarried mothers of first-born children without disabilities when the mother was younger than 35, $X^2(2, n=139,435) = 85.10$, $p<.001$, the child was born before 2000, $X^2(2, n=70,326) = 13.59$, $p<.001$, and the mother was younger than 35 and the child was born before 2000, $X^2(2, n=68,490) = 16.87$, $p<.001$.

Married mothers of first-born children with disabilities were also more likely to have subsequent children than married mothers of first-born children without disabilities. Overall, 46.7% of married mothers of first-born children with Down syndrome, in comparison to 40.3% of married mothers of first-born children with spina bifida, in comparison to 30.3% of married mothers of first-born children without disabilities had a subsequent child, $X^2(2, n=209,821) = 34.91$, $p<.001$.

For second-born children, younger, unmarried mothers and married mothers (regardless of age) of children with disabilities were more likely to have subsequent children than mothers of children without disabilities. As shown by Table 10, when the

mother was unmarried, there was not a significant difference in having a third child regardless of whether the second-born did or did not have a disability among the groups. However, if the mother was both unmarried and under 35, 31.1% of mothers of children with Down syndrome in comparison to 15.2% of mothers of children without disabilities had subsequent children, $X^2(1, n=152,997) = 41.60$, $p<.001$. For married mothers there were significant differences in having a third child depending on whether the second-born had a disability across the groups regardless of the age of the mother (mothers younger than 35, $X^2(2, n=153,064) = 50.53$, $p<.001$; mothers at or older than 35, $X^2(2, n=22,696) = 13.46$, $p<.001$) and the birth year of the child (before 2000, $X^2(2, n=112,718) = 35.90$, $p<.000$; at or after 2000, $X^2(2, n=63,010) = 10.31$, $p<.001$).

Table 10. Marital status: Subsequent children after the second-born across groups

	Without disabilities % (N)	SB % (N)	DS % (N)	X^2	p
Married	14.0% (24,469)	24.4% (19)	26.4% (78)	45.20	.000
Not Married	21.3% (12,782)	----------	29.3% (24)	3.077	.100

Note: In the spina bifida group, there were sometimes too few subjects to perform analyses (N<10); in these cases, chi-squares compare families of children without disabilities to the DS group.

Table 11 shows the differences in having subsequent children depending on whether the third-born child had a disability across the groups when considering the marital status of the mother. In comparison to married mothers of children without disabilities, married mothers of children with Down syndrome were more likely to have subsequent children.