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Examining Courtesy Stigma in Siblings of People with Down Syndrome

THESIS

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by

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DEDICATION

Many thanks to

my husband and parents, for your love and support,

and to my thesis mentor and committee, for your guidance, patience, and insight

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ABSTRACT OF THE THESIS

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Master of Science in Genetic Counseling

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Professor Maureen Bocian, MS, MD, Chair

The purpose of this study was to determine whether siblings of people with Down syndrome face courtesy stigma, a stigma acquired as a result of an association with a person from a stigmatized group. The central hypothesis was that the majority of people who have a sibling with Down syndrome face courtesy stigma during both adolescence and adulthood. The data supports this hypothesis, showing that 76% of respondents reported courtesy stigma as adolescents and 62% reported courtesy stigma as adults. The levels of courtesy stigma reported were higher in adolescence than adulthood. However, the overall levels of courtesy stigma reported by all respondents were low, and participants reported strongly positive relationships with their siblings with Down syndrome despite experiencing courtesy stigma. Other findings of this study include a positive correlation between aberrant behaviors on the part of the individual with Down syndrome and higher levels of courtesy stigma reported by their siblings and an increase in courtesy stigma when the sibling with Down syndrome also has a comorbid condition, such as autism or a lack of verbal communication skills. Due to a small participant population, this study was unable to establish whether the amount of courtesy stigma experienced by siblings of people with Down syndrome has changed together with our society's attitudes toward people with intellectual disabilities over the past several decades.

INTRODUCTION

Courtesy Stigma

Stigma

Stigma: a set of negative and often unfair beliefs that a society or group of people have about something: a mark of shame or discredit: an identifying mark or characteristic: a specific diagnostic sign of a disease (meriam-webster.com). Stigmatization occurs though a combination of stereotyping, prejudice and discrimination in the presence of an imbalance of power between groups in a society. This imbalance in power may be fueled or underscored by social, economic, or political differences. Stigmatization is a process by which certain groups are marginalized and devalued by society because their values, characteristics or practices differ from those of the dominant cultural group (Rusch et al., 2005). Individuals with intellectual disabilities have been one of the most stigmatized and socially ostracized groups throughout the history of our society. The discrimination and disapproval that individuals with Down syndrome face in our culture due to their intellectual disabilities and distinctive facial features have been well studied and documented (Afia et al, 2012). The results can be far-reaching. When asked about the effects of stigmatization on their children with mental illness or intellectual disabilities, caregiving parents cited difficulty making and keeping friends, damage to their self-esteem, difficulty finding a job, and reluctance to admit to their condition (Wahl, 1989).

Family Stigma

While the individuals with mental illness or intellectual disabilities were the direct targets of the stigmatization, Wahl (1989) found that their caregiving relatives were also impacted by the stigmatization of their loved ones. The most commonly cited effects on the caregiver and family

unit were lowered self-esteem and damaged family relationships. Many studies support the idea that the effects of stigma are not limited to the stigmatized individuals but also often affect those who are closely associated with them, such as members of the family, friends, and even professionals who work with them. This is known as courtesy stigma (Birenbaum, 1992). Courtesy stigma may result in family members being teased, abused, blamed or considered responsible for their relative's disability (Larson & Corrigan, 2008). Family members may develop negative self-perceptions and emotions that may cause them to withdraw from social activities or conceal their perceived negative status from others. Previous research has shown that caregivers of people with intellectual disabilities, including parents and other older relatives, face courtesy stigma (Afia et al 2012). This meta-analysis reviewed thirty-seven previous studies concerning both the self-stigmatization faced by individuals with intellectual disabilities, including some that specifically examined Down syndrome, as well as the courtesy stigma faced by their caregiver relatives. Only one of these studies focused on the courtesy stigma experienced by siblings, and it did not focus specifically on siblings of people with Down syndrome. Separate studies have shown that siblings of individuals in other stigmatized groups, such as those with autism (Orsmund et al., 2009), mental illness (Corrigan & Miller 2004), and developmental delays (Seltzer et al., 2005), all experience courtesy stigma to varying degrees. It is known that individuals with Down syndrome are still a highly stigmatized group in our society and that stigmatization can carry over to their close relatives in the form of courtesy stigma. There is a gap in the existing research because no one has yet examined the possible courtesy stigma experienced by siblings of people with Down syndrome.

Down Syndrome

Introduction

Down syndrome is a chromosomal abnormality that is characterized by variable intellectual disabilities, typical and recognizable facial features, and often malformations of the heart or other organs. Down syndrome is the most common genetic cause of intellectual disability, also known as mental retardation. This syndrome is named for John Langdon Down, who first described its common features in a subset of children with intellectual disabilities in1866. It took over seventy years to determine the cause of Down syndrome once it had been characterized. Jérôme Lejeune, a French scientist, first announced the cause as a third copy of chromosome twenty-one in 1959.

Once the cause of this syndrome was firmly established, and with advances in technology in recent decades, scientists and medical professionals have been able to gain a much more thorough understanding of Down syndrome and to provide better care for people with this condition. Increasing social awareness of Down syndrome as well as the efforts of advocacy groups and legislative advances all have contributed to the better social treatment of people with Down syndrome. However, people with Down syndrome still face many challenges, both from their condition itself and from the prejudice they still face in our society (Ali et al., 2012).

Etiology

As Jérôme Lejeune discovered, Down syndrome is caused by the presence of additional material from chromosome twenty-one in many or all cells of the body. It is often referred to as trisomy twenty-one, which means that there is a third (tri-) copy of the twenty-first chromosome (-somy) present in the cell. The vast majority of individuals with Down syndrome, approximately

ninety-five percent, receive a complete extra copy of chromosome twenty-one via nondisjunction. This occurs due to an error in the separation of chromosomes during meiosis, the process of cellular division that leads to the production of gametes. During a normal meiosis, an originator cell contains forty-six chromosomes, two copies of each of the twenty-three human chromosomes. This cell then divides into two daughter cells, each containing a single copy of each chromosome. In Down syndrome, an error in this process called nondisjunction results in a sperm or egg cell that contains two copies of chromosome twenty-one instead of the usual single copy. When this sperm or egg cell pairs with the other normal complementary gamete containing a single copy of chromosome twenty-one, the resulting fertilized egg has three copies of chromosome twenty-one instead of the usual two. Meiotic nondisjunction occurs more often during maternal meiosis (92%) than during paternal meiosis (8%) (Ballesta et al., 1999). This type of meiotic error is also more likely to occur with advancing maternal age.

The five percent of cases of Down syndrome not caused by meiotic nondisjunction result from either post-zygotic nondisjunction or an inherited chromosome translocation involving all or part of chromosome twenty-one. Chromosome translocations occur when entire chromosomes, or parts of different chromosomes, are joined together. Unlike meiotic nondisjunction, chromosome translocations are inherited equally from maternal and paternal gametes and are not age-dependent. A complete third copy of chromosome twenty-one is not necessary for expression of the Down syndrome phenotype. Although most individuals with Down syndrome due to a translocation have a nearly entire additional copy of chromosome twenty-one, some individuals with translocation-caused Down syndrome only have a small additional piece of chromosome twenty-one in their cells and still meet clinical criteria for the condition. Individuals such as these

are referred to as having “partial” trisomy twenty-one. Studies of partial trisomy twenty-one cases have revealed certain genes that are important contributors to the Down syndrome phenotype (Ruparelia et al., 2010).

Post-zygotic nondisjunction can result in individuals who have an extra copy of chromosome twenty-one in some, but not all, of their cells. This condition is known as mosaic Down syndrome. Some studies suggest that people with mosaic Down syndrome may be less severely affected than people who have extra chromosomal material present in all of their cells (Leon et al., 2010). The ameliorated mosaic phenotype is dependent upon the level of mosaicism in various tissues, such as the brain, heart, and others, and on when in embryologic development the error occurred. It is difficult to measure the level of mosaicism in most tissue types, and the level of mosaicism found in lymphocytes, the most commonly analyzed cell type, may not accurately reflect the level of mosaicism in the brain, heart, or other types of tissue.

Chromosome twenty-one is the smallest of the human chromosomes. There are thought to be between 200 and 300 genes that encode proteins located on this chromosome (Hattori, et al. 2000). It is unclear exactly how the presence of extra material from chromosome twenty-one causes the clinical phenotype of Down syndrome. Conditions such as Down syndrome, in which the spectrum of clinical features results from more than one gene located along one chromosome, are called contiguous gene syndromes. One possible explanation for the effects of the extra material from chromosome twenty-one is that the imbalance in the amount of genetic material present in cells may disturb developmental processes that lead to the spectrum of phenotypic effects recognized in Down syndrome (Ruparelia et al., 2010).

Phenotype

John Langdon Down was the first person to describe the physical phenotype of Down syndrome. The typical findings include brachycephalic microcephaly, fine hair, upward slanting palpebral fissures, flattened midface, Brushfield (light-colored) spots on the irises, small and sometimes abnormally folded ears, macroglossia (large tongue), short and broad hands and feet, clinodactyly (lateral curving) of the fifth fingers, a single transverse palmar crease, and a larger than usual space between the first and second toes (Ahmed et al., 2005). None of these features by itself is enough to be diagnostic of Down syndrome, since most are found in some proportion of the general population. Rather, it is a combination of several of the aforementioned features in one individual that gives the typical appearance of Down syndrome.

People with Down syndrome are at higher risk for certain congenital anomalies and disorders involving various organ systems. Approximately fifty percent of babies with Down syndrome have congenital heart defects. The most common type of congenital heart abnormality is a ventricular septal defect (Ahmed et al., 2005). The severity of these defects ranges from mild to severe enough to require surgical intervention. Another condition more common in people with Down syndrome is thyroid dysfunction. Both hyper- and hypothyroidism are more common in people with Down syndrome than in the general population, especially hypothyroidism (King et al., 2014). Children with Down syndrome are also more prone to infections such as pneumonia. Ear infections are especially common in children with Down syndrome and lead to hearing loss in greater than forty percent of affected individuals. Hearing loss, if unidentified or untreated, can further impair children with Down syndrome once they reach school age. The higher incidence of ear infections is due to their often narrow ear canals as well as their relatively

weaker immune systems. The immune deficiency responsible for the increased frequency of various infections has also been proposed as a potential contributor to the nearly twenty-fold increased risk of leukemia in individuals with Down syndrome. Most of this increased risk occurs in the first few decades of life, with the highest incidence being in children less than five years of age (Ross et al., 2005). People with Down syndrome also have an increased frequency of congenital obstruction of the gastrointestinal tract, such as duodenal atresia and Hirschsprung disease, and of celiac disease.

In addition to their typical physical features and susceptibility to certain disorders, people with Down syndrome often exhibit a typical behavioral phenotype. Both children and adults with Down syndrome are often characterized by their outgoing, good-humored temperament and affectionate nature; however, individuals with Down syndrome can also tend toward stubbornness and obstinacy. Children with Down syndrome have been found to have greater empathic responses than either typically developing children or children with other types of intellectual disability (Kasari et al., 2003). On the other hand, children with Down syndrome are prone to acquiring new, unwanted behaviors due to their propensity for mimicry or imitation of the behaviors of others.

Babies and young children with Down syndrome achieve the same motor developmental milestones as typical children, but they tend to do so at a slower pace. An average typical child will roll over at five months, sit alone at seven months, stand alone at eleven months, and begin walking at around one year of age. For an average child with Down syndrome, rolling over begins at eight months, sitting alone at nine months, standing alone at eighteen months, and walking usually begins around two years. Typical children can begin to speak their first

meaningful words as early as ten to fourteen months, and this usually happens at thirteen to eighteen months for children with Down syndrome (DSMIG, 2011).

Due to the mild to moderate intellectual disabilities of people with Down syndrome, they reach an average mental age of a nine-year-old typical child. The average intelligence quotient (IQ) for an adult with Down syndrome ranges from forty-five to forty-eight, and the upper limit for IQ in people with Down syndrome is approximately seventy (Handbook of Genetic Counseling, 2001). There is significant variability in the level of independence achieved by adults with Down syndrome. Some adults with Down syndrome are able to live in group homes with other adults with mild intellectual disabilities and to live relatively independent lives. These individuals may hold stable jobs in the community or at workshops designed to hone job skills for people with milder cognitive impairments. Other adults with Down syndrome may live at home with their parents and require more assistance with daily living tasks. Many individuals with Down syndrome are able to graduate from high school; some go on to attend college. This range of abilities is largely based on IQ and can also be influenced by the variation in educational opportunities and living environments of individuals with Down syndrome (Brown 2004).

Common Comorbid Conditions

Studies have shown an inverse relationship between IQ level and the prevalence of psychiatric disorders (Collacott et al., 1992). In keeping with this trend, people with Down syndrome have a higher than average susceptibility to various psychiatric illnesses, including depressive disorders, hyperkinetic disorders, obsessive compulsive disorder, autism, and dementia. Conversely, people with Down syndrome appear to be less prone to schizophrenia and personality disorders (Collacott et al., 1992).

Alzheimer disease, which is characterized histologically by the presence of beta-amyloid plaques and neurofibrillary tangles in the brain, is the most common form of dementia in the general population, affecting approximately eleven percent of adults over age sixty-five in the United States (Thies & Bleiler, 2011). The prevalence in Alzheimer disease in people with Down syndrome is significantly higher than in the general population, with approximately eighty percent of individuals over the age of fifty-five showing at least mild symptoms (Zigman, 2013). The prevalence of beta-amyloid plaque formation in the brains of people with Down syndrome is even more striking than it is in the general population, and this is likely due, at least in part, to the fact that the amyloid beta precursor protein gene is located on chromosome twenty-one.

Genetic Counseling

Genetic counseling related to Down syndrome can take place in either a prenatal or a pediatric setting and can cover a range of topics including explaining the full spectrum and range of the phenotype, understanding the etiology and how it occurs, helping parents or prospective parents set reasonable expectations for their child with Down syndrome, discussing recurrence risks, family planning, considering the potential impact on existing siblings, and many others. In the prenatal setting, many prospective parents wish to find out whether their fetus will have Down syndrome. Currently, several different screening and diagnostic testing options are available for individuals. One type of screening test uses the levels of several proteins identified in maternal blood during pregnancy, combined with an ultrasound measurement of the fetus' nuchal translucency (the thickness at t back of the fetus' neck), to determine the chance that the fetus has Down syndrome. This type of screening has approximately a ninety percent detection rate for Down syndrome but also has a false positive rate of approximately five percent

(Malone et al., 2005). A newer type of screen measures the amount fetal DNA fragments from chromosome twenty-one present in the maternal blood stream to determine the likelihood of there being three copies of this fetal chromosome rather than the typical two copies. This type of screen, known as cell-free fetal DNA screening or non-invasive prenatal screening, has up to a ninety-nine percent detection rate for Down syndrome (Chiu et al., 2011). More precise diagnostic data is obtained prenatally by performing either a chorionic villus sampling between ten and thirteen weeks or an amniocentesis after fifteen weeks of pregnancy. These procedures are used to obtain fetal cells, cultured from the placenta or amniotic fluid, respectively, that can be used to produce a karyotype, or organize profile of the fetus' full complement of chromosomes. Looking directly at the fetal chromosomes allows for a higher, diagnostic, level of accuracy; however, these procedures also carry a risk for complications that can include miscarriage. Navigating the complex plethora of screening and diagnostic testing options in the prenatal setting requires a close working relationship among genetic counselors, obstetricians, and prospective parents.

The recurrence risk for Down syndrome is dependent on the genetic form of the disorder. The recurrence risk to a couple with the classical form of Down syndrome due to nondisjunction is either approximately one percent or the maternal age-related risk, whichever is higher. In the translocation form of Down syndrome, the risk depends on whether the translocation was inherited from a parent or occurred *de novo*, for the first time in that individual child. The majority of cases of translocation Down syndrome occur *de novo*, and in these cases parents have a recurrence risk of less than one percent (Bansal et al., 2010). A parent who carries a translocation involving chromosome twenty-one is referred to as a "balanced translocation

carrier." In this circumstance, the carrier parent is asymptomatic and has a set of chromosomes containing the normal amount of chromosome material, but part or all of one copy of their twenty-first chromosome is attached to another chromosome. In this case, the genetic risk varies widely and depends upon the chromosome to which the whole or partial chromosome twenty-one is joined. The recurrence risk for the mosaic form of Down syndrome is similar to that of classic trisomy twenty-one and in some cases can be related to maternal age. Counseling for rare forms of Down syndrome can be quite complicated and should always involve the services of a professional genetic counselor or clinical geneticist.

Siblings of People with Down Syndrome

Several organizations as well as many individual researchers have focused on studying various aspects of the lives and experiences of individuals with Down syndrome, but their siblings have received less attention from the research community. The current study attempts to help fill this gap in our collective knowledge by examining one particular aspect of the experience these siblings share. From studies that have focused on these siblings' experiences, a wide range of emotions and perspectives have been reported; the majority of individuals have positive or relatively positive relationships with their siblings with Down syndrome. Cuskelly and Gunn (2003) conducted several studies on the childhood experiences of people who grew up with a sibling with Down syndrome. They found, via parental reports, that there was greater empathy and fewer unkind acts between sibling dyads in which one sibling has Down syndrome, compared to dyads of typically developing children. These researchers also examined the adjustment of children who grew up with a sibling with Down syndrome in comparison to children who grew up with typically developing siblings, and they found that there was no

difference between the groups with respect to adjustment measures. Siblings of children with Down syndrome performed equally well on parentally reported measures of externalizing and internalizing behaviors and social competence, as well as the siblings' perceptions of their own competence and self-worth (Cuskelly & Gunn, 2006). When compared to siblings of individuals with other chronic conditions, such as diabetes, autism, and orthopedic disorders, siblings of people with Down syndrome showed the most kindness and empathy toward their affected sibling (Nielsen et al., 2012). This finding may suggest that having a sibling with Down syndrome provides a unique experience and encourages even greater empathic responses than simply having a sibling with any type of disability or chronic health condition. Perhaps this effect on siblings could be explained by the fact that children with Down syndrome tend to show greater empathy, especially in response to the distress of others, than do typically developing children or children with other types of intellectual disability (Kasari et al., 2003). The effects of the early bonds formed between many individuals with Down syndrome and their siblings appear to persist into adolescence and adulthood. When compared to adult siblings of people with an autism spectrum disorder, adult siblings of people with Down syndrome tend to retain more emotional closeness, greater involvement in their siblings' lives, and fewer pessimistic thoughts about their siblings' future (Orsmond & Seltzer, 2007). Brian Skotko, well known for research in this field, has found that the majority of these individuals feel affection for their siblings with Down syndrome and report having positive sibling relationships (Skotko et al, 2011). However, not all attitudes were positive, and the study also found that nearly ten percent of respondents felt embarrassment because of their siblings with Down syndrome, and approximately five percent of siblings reported wishing that they could trade their brother or sister with Down syndrome for a

sibling without the condition. A separate study conducted by Skotko and Levine (2006) examined the experiences and feelings of siblings of people with Down syndrome. This study again uncovered a wide range of emotions reported by the brothers and sisters of people with Down syndrome as they experienced the disability community from a family perspective. Similar to their 2011 study, these researchers also found that the positive feelings and experiences that were reported far outnumbered the negative reports.

Given these previous findings, it would appear that, while there is a wide range of emotions experienced by siblings of people with Down syndrome, most siblings feel that their lives have been impacted in a positive way by their brothers or sisters with Down syndrome. No published studies appear to have delved deeper into the reasons that a small percentage of siblings have for reporting negative emotions about having a brother or sister with Down syndrome. Because it has been reported that siblings of people with various other stigmatized conditions experience a courtesy stigma, the goal of the current study is to see if siblings of people with Down syndrome experience a similar stigmatization by association. If this is found to be the case, this experience of a courtesy stigma could potentially account for some of the negative feelings reported by a minority of these siblings.

The Current Study

The purpose of the current study is to determine whether siblings of people with Down syndrome experience a courtesy stigma. I hypothesize that the majority face some level of courtesy stigma, or stigma by association with their stigmatized sibling. This hypothesis is based on published research showing that family members, including siblings, of relatives with various other stigmatized conditions face this type of stigma by association. Siblings of other stigmatized

groups, such as those with autism (Orsmund et al., 2009), mental illness (Corrigan & Miller 2004), and developmental disabilities (Seltzer et al., 2005) have been studied and found to face various challenges, including courtesy stigma.

The current study was designed to query adult siblings of people with Down syndrome about the level of courtesy stigma, if any, that they currently experience as well as whether they faced any courtesy stigma as adolescents. A secondary hypothesis of this study is that I expect participants to report more adolescent courtesy stigma than adult courtesy stigma. Adolescents tend to be more sensitive to the way that they feel others perceive them, so I expect people to be more vulnerable and sensitive to courtesy stigma during this period than at any point during their adulthood. The design of measuring both past and current courtesy stigma also allowed for an examination of the levels of courtesy stigma experienced by adolescent siblings of people with Down syndrome over the last several decades. A secondary hypothesis of the current study is that as our society has become relatively more open and accepting of individuals with Down syndrome over the past few decades, the level of courtesy stigma experienced by the adolescent siblings of people with Down syndrome will have decreased over this period of time. This secondary hypothesis could be supported by finding a difference by age in the proportion of siblings who report stigma, with older siblings reporting more stigma than siblings who are younger.

Another secondary hypothesis of this study posits that individuals may experience more courtesy stigma when their siblings with Down syndrome exhibit more socially undesirable behavior. This hypothesis is based on the idea that having a sibling with Down syndrome who may attract more negative attention in public situations through atypical and undesirable behaviors may

result in higher levels of courtesy stigma for our participants. For similar reasons, I hypothesize that participants whose siblings with Down syndrome have significant comorbid conditions, such as autism, Alzheimer disease, or others, will report experiencing more courtesy stigma. The final hypothesis of this study is that siblings who experience more courtesy stigma will choose to be less involved with their siblings with Down syndrome as adults. In order to investigate this hypothesis, participants were asked about their level of involvement with their siblings with Down syndrome through a report of how often they see and speak to them. I expect that participants who report higher levels of courtesy stigma will report lower levels of contact with their siblings with Down syndrome.

No previous study has looked at the potential courtesy stigma facing siblings of people with Down syndrome, and I believe this to be a significant gap in the field of stigma research. If the primary hypothesis of this study—that siblings of people with Down syndrome face a courtesy stigma—is supported, this could be an important first step in recognizing this particular struggle in the lives of many such individuals. Ultimately, the goal of this research is to bring this possible stigma to light so that it can be addressed and alleviated in our society.

RESEARCH DESIGN AND METHODS

Participants

In order to participate in this study, we required that survey respondents be eighteen years of age or older. Participants also had to be the full-, half-, step-, or adoptive sibling of someone with Down syndrome in order to be included in this study. Thirty-one men and eighty-one women meeting the aforementioned criteria, as well as one participant who chose not to reveal his or her

gender, completed our survey, giving this study a total number of 113 participants. A demographic breakdown of the participant population of this study is provided in Table 1.

Instrument

The current study uses an online survey instrument to investigate the possible existence of a courtesy stigma faced by siblings of people with Down syndrome. This survey was approved by the University of California, Irvine's Institutional Review Board prior to its use, and can be viewed in Appendix A. The survey consisted of 116 questions and required approximately ten to fifteen minutes for completion. The survey included three initial questions asking for identifying data that were viewed only by an independent third party and not by the research team. Answers were used for the sole purpose of linking together respondents from the same families. Participants were able to complete the survey at their own convenience at any location that provided internet access.

The survey was constructed using UC Irvine's REDCap program, and consisted of three main sections, or domains (Harris et al., 2008). The first section is a demographic questionnaire that did not request any identifying information from participants but allowed the research team to investigate possible effects on the experience of courtesy stigma that may be related to variations among our respondents' backgrounds. The second section of our survey comprised a slightly modified version of the Affiliate Stigma Scale, designed and validated by Winnie Mak and Rebecca Cheung to assess courtesy stigma experienced by relatives of people with mental illness or intellectual disabilities (Mak & Cheung, 2008). While the original instrument was written in Chinese, Mak and Cheung used a backward-translation approach to develop an English version. Discrepancies between the Chinese and English versions were rectified, and the equivalence of

meaning on all items was ensured through consultation with several bilingual researchers (Mak & Cheung, 2008).

In the current study, the English version of the scale was modified to include the assessment of both past and present stigma, and the wording of the questions was changed only slightly to tailor them to siblings of people with Down syndrome rather than their original broad inclusion of any relative of someone with an intellectual disability or mental illness. For example, the original question, "The behavior of my relative with mental illness or intellectual disability makes me feel embarrassed," was modified to, "The behavior of my sibling with Down syndrome makes me feel embarrassed," in the current study. Winnie Mak, one of the original researchers who created the scale, was consulted directly to ask permission to use the instrument that she co-developed. She agreed to our use of her instrument and confirmed that it should be a valid and reliable instrument for assessing courtesy stigma in siblings of people with Down syndrome. She also approved the slight modification of the scale and did not feel that the minimal changes made would affect the validity of the instrument. The modified Affiliate Stigma Scale contained twenty-two items scored on a four-point Likert scale. Each item posed a scenario designed to measure courtesy stigma, and respondents were then asked to choose one of the following selections from the Likert scale: "Strongly disagree," "Disagree," "Agree," or "Strongly agree." To "Strongly disagree" represented a score of one point and a complete absence of courtesy stigma, while to "Strongly agree" represented a score of four points and showed the greatest amount of courtesy stigma. In other words, when the scores of all twenty-two items of the Affiliate Stigma Scale are averaged, an overall average score equal to one

represents an absence of courtesy stigma, and an average score greater than one indicates that some level of courtesy stigma was experienced.

The third section of the survey consisted of questions regarding the past and present health and behavior of the sibling with Down syndrome. Questions in this section of the survey were adapted from the Aberrant Behavior Checklist, which was designed to assess the behavior of individuals with intellectual disabilities (Aman et al., 1985). The research team was interested in gathering information about any comorbid medical conditions as well as any behavioral issues of the sibling with Down syndrome to assess whether these may have had an effect on the level of courtesy stigma experienced by the siblings of these individuals. The scores on each item of the Aberrant Behavior Checklist were summed to create a final behavior score for each sibling with Down syndrome. On the Aberrant behavior Checklist, twenty undesirable behaviors are listed, and for each, the respondent must choose whether the behavior is "No problem" (one point), "Slight problem" (two points), "Moderate problem" (three points), or "Severe problem" (four points). Higher sums of the scores from the twenty items indicate more overall undesirable behavior. Participants were asked to rate the past and present behavior of their sibling with Down syndrome separately on the Aberrant Behavior Scale so that the research team could test the effects of behavior on both adolescent and current courtesy stigma.

Recruitment methods

The research team reached out to Down syndrome associations and family support groups in all fifty states via email in the attempt to procure a geographically diverse respondent population. Numerous Down syndrome associations across the country, including the National Down Syndrome Society, the Down Syndrome Association of Orange County, Down Syndrome

Association of Los Angeles, San Diego's Down Syndrome Center for Research and Treatment, Down Syndrome Association of Southern Texas, Down Syndrome Association of Central Kentucky, Utah Down Syndrome Foundation, Down Syndrome Association of West Michigan, Down Syndrome Association of Central New Jersey, Rocky Mountain Down Syndrome Association, and many others agreed to share information about this study with their members. Several Down syndrome sibling and family support groups, including The Sibling Support Project, Down Syndrome Information Alliance, and FRIENDS (Family, Resource, Information, and Education Network for Down Syndrome) also agreed to share information about this study with their members. These organizations agreed to post a link to the online survey on their websites accompanied by an informational paragraph generated by the research team explaining the purpose of the study and the time commitment required, send information about our study to their members via email in their regular newsletters, and/or post a link to the survey and information about the study on their Facebook pages.

Two members of the research team were involved in clinics where people with Down syndrome are seen, and they passed out flyers containing information about our study to those among their patients who have adult siblings. The flyers contained a link to our online survey and provided information about the purpose of this study. The caregivers of these patients were also contacted via email to provide them with more information about the study as well as the link to the survey.

Data Analysis

Data from the REDCap survey were exported into an Excel spreadsheet by a member of UC Irvine's Institute for Clinical and Translational Science department. He also used the minimal set

of identifying data to link siblings from the same families together and assigned each family an anonymous numerical code prior to delivering the data to the research team. The de-identified data was then analyzed using Mystat, the free, downloadable statistical analysis software program (Systat Software, San Jose, CA). Since the levels of courtesy stigma reported by the participants in this study were not normally distributed but instead were skewed towards the lower end of the courtesy stigma rating scale, participants were separated into two groups based on whether they reported experiencing some level of courtesy stigma, shown by an average Affiliate Stigma Scale score greater than one, or none at all, shown by a an average Affiliate Stigma Scale score equal to one. The ages of our participants also failed to be normally distributed because we had far more participants in their twenties and thirties and fewer in older decade groups, so participants were once again split into two groups—those age thirty or younger and those over age thirty. Dividing our participants into these groups allowed for analyses using two-way tables and chi-square tests.

Along with the chi-square tests used to analyze the courtesy stigma data acquired in this study, descriptive data, including the information gathered from the demographic section of the questionnaire, are presented using appropriate descriptive statistics. Paired t-tests and McNemar's test were used to test the primary hypothesis that the majority of siblings of people with Down syndrome do experience some level of courtesy stigma, and that this experience of courtesy stigma will be more pronounced during adolescence than adulthood. Additionally, Spearman correlations were used to assess the relationship between aberrant behavior and courtesy stigma. Fisher's exact tests were used to test some two-way associations when warranted by small numbers of respondents.

RESULTS

Demographics

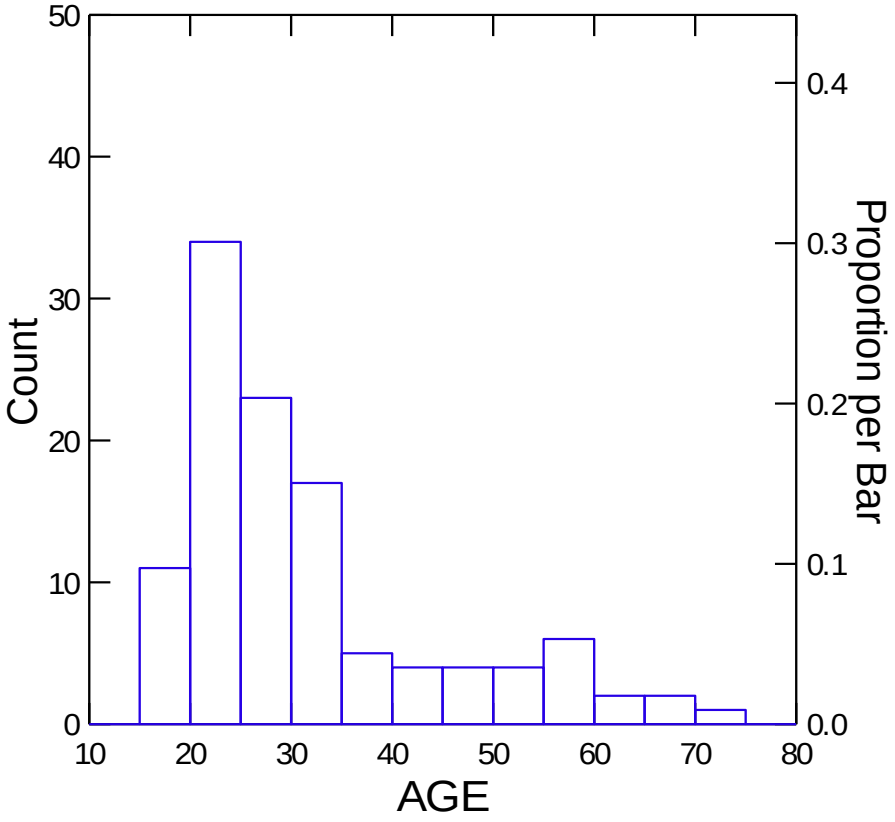
Table 1 shows the demographic makeup of the participant population. The participant population in this study contained more female respondents than males, a common occurrence since women tend to respond to surveys in greater numbers than men. Eighty-one respondents (72%) were female and thirty-one (27%) were male. White respondents were also overrepresented at 89% of the survey population as compared to the racial makeup of the United States. There was also an overrepresentation of young adult respondents to this survey, and fewer participants to represent older generations. The median age of the respondents to this study was twenty-eight, but the ages of the participants were not normally distributed. The age distribution can be seen in Figure 1. Respondents to this study were regionally diverse. All five regions of the United States- the Northeast (14%), Southeast (19%), Midwest (18%), Northwest (12%), and Southwest (35%)- were represented, and some respondents who now reside in the United States were born internationally. The participant population of this study was also well educated with 92% having attended at least some college, and the majority of participants had received either a Bachelor's or graduate degree.

Table 1. Demographic Makeup of Participants

	N	%
Gender		
Male	31	27
Female	81	72
Race		
Asian	3	3
African American	0	0
Pacific Islander	1	1
White	100	89
Other	8	7
Prefer not to specify	1	1
Ethnicity		
Hispanic/Latino	13	12
Not Hispanic/Latino	94	83
Prefer not to specify	5	4
Region Where Participant Grew Up		
Northeast	19	17
Southeast	14	12
Midwest	23	20
Northwest	9	8
Southwest	43	38
International	4	4
Region Where Participant Lives Now		
Northeast	16	14
Southeast	22	19
Midwest	20	18
Northwest	13	12
Southwest	40	35
Socio-economic Status		
Upper class	4	4
Upper-middle class	23	20
Middle class	66	58
Lower-middle class	19	17
Lower class	1	1
Education Level		
Did not complete high school	1	1
High school diploma/GED	8	7
Some college	32	28
Bachelor's degree	38	34
Graduate degree	33	29
Relationship to Sibling with Down Syndrome		
Full sibling	97	86
Half sibling	12	11
Step sibling	0	0

Adoptive sibling	3	3
Gender of Sibling with Down Syndrome		
Male	66	58
Female	47	42
Birth Order		
Older than sibling with Down syndrome	96	85
Younger than sibling with Down syndrome	17	15
	Mean, Median	Range, SD
Age	31, 28	18-71, 13
Total Number of Siblings	3, 3	1-9, 2
Years Spent in Same Home as Sibling with Down Syndrome	15, 15	0-52, 7

Figure 1. Age Distribution of Participants

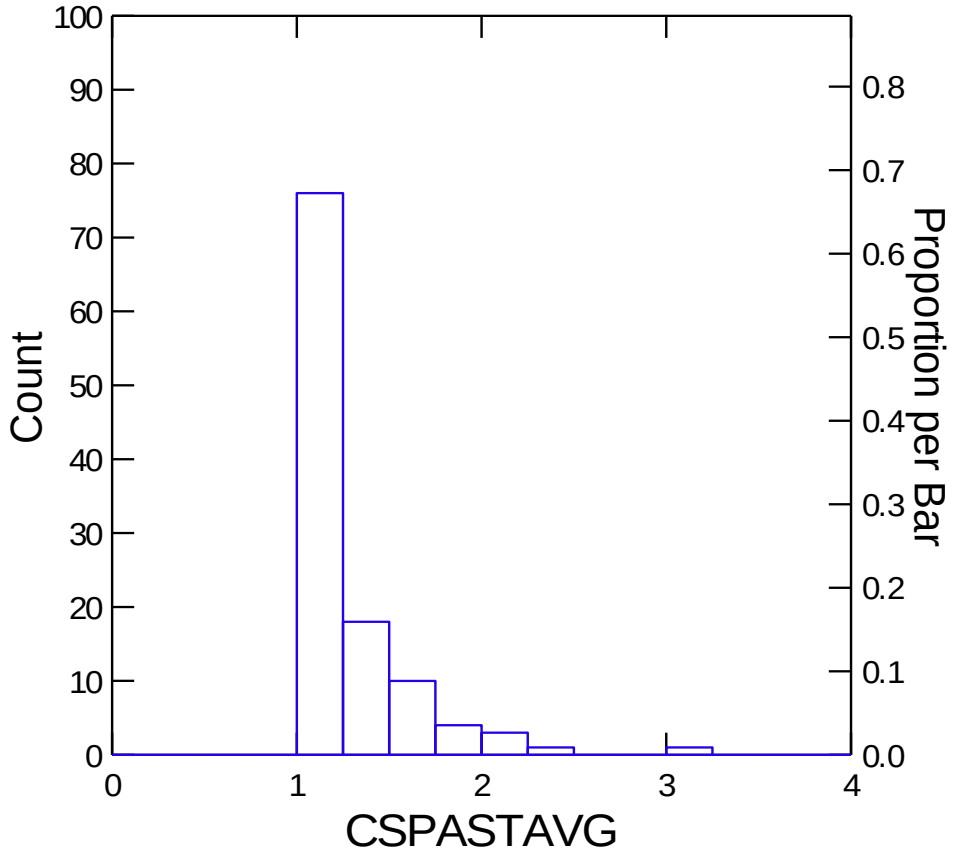


Primary Hypothesis

The primary hypothesis of this study—that the majority of siblings of people with Down

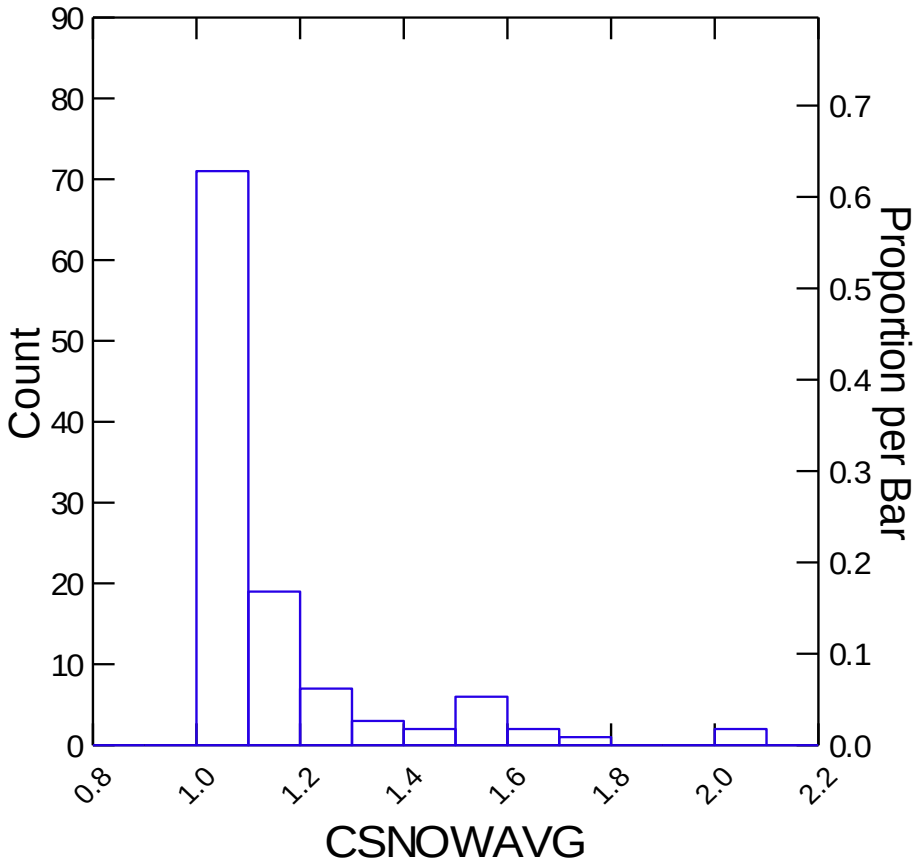
syndrome experience some level of courtesy stigma—was supported by the data gathered in this project. The majority (76%) of siblings who responded to our survey reported having felt at least a minimal level of courtesy stigma in the past, and 62% reported that they currently experience some level of courtesy stigma. A respondent was determined to have experienced courtesy stigma if their average score on the modified Affiliate Stigma Scale was greater than 1.0. One interest of the current study was to determine if there are differences in the levels of courtesy stigma experienced by siblings of people with Down syndrome during the adolescent and adult periods. The percentage of participants who reported adolescent courtesy stigma was significantly greater than the percentage of participants who reported that they currently experience courtesy stigma, as shown by a McNemar's test ($p < 0.05$). The average level of courtesy stigma experienced by our participants during their adolescent years (1.25) was also significantly greater than the average that they reported experiencing currently as adults (1.14), as shown by another paired t-test ($p < 0.05$). These findings support the idea that siblings experience more courtesy stigma during their adolescence than they do during their adult years. The average Affiliate Stigma Scale scores of the respondents for adolescent courtesy stigma are shown in Figure 2, and their averages for current adult courtesy stigma are shown in Figure 3.

Figure 2. Adolescent Affiliate Stigma Scale Average Scores



The mean of the adolescent courtesy stigma scores was1.25, the median was 1.14, and the range was 1.00-3.18.

Figure 3. Current (Adult) Affiliate Stigma Scale Average Scores



The mean of the adult courtesy stigma scores was 1.14, the median was 1.09, and the range was 1.00-2.00.

The research team was interested to determine whether the levels of courtesy stigma reported by our participants would vary depending on their demographic characteristics. Differences in the levels of courtesy stigma that siblings reported during their adolescence based on demographic characteristics are listed in Table 2. Respondents were divided into groups based on whether they expressed some courtesy stigma, defined by an average score greater than one on the modified

Affiliate Stigma Scale, or no courtesy stigma at all, defined by an average score equal to one on the modified Affiliate Stigma Scale.

Table 2. Adolescent Courtesy Stigma by Demographic Groups

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma		
	N	%	N	%	p value (chi-square)
Gender					0.467
Male	25	81	6	19	
Female	60	74	21	26	
Age					0.614
18-30	56	75	19	25	
31-71	30	79	8	21	
Race					0.190
White	78	78	22	22	
Non-White	8	62	5	38	
Ethnicity					0.029*
Hispanic/Latino	6	46	7	54	
Non Hispanic/Latino	75	80	19	20	
Region Where Participant Grew Up					0.372
Northeast	14	74	5	26	
Southeast	13	93	1	7	
Midwest	17	74	6	26	
Northwest	8	89	1	11	
Southwest	30	70	13	30	
International	4	100	0	0	
Socio-economic Status					0.435
Upper class	3	75	1	25	
Upper-middle class	19	83	4	17	
Middle class	50	76	16	24	
Middle-lower class	14	74	5	26	
Lower class	0	0	1	100	
Education Level					0.300
Did not complete high school	1	100	0	0	
High school diploma/GED	4	50	4	50	
Some college	24	75	8	25	
Bachelor's degree	32	84	6	16	
Graduate degree	24	73	9	27	
Relationship to Sibling with					0.369

Down Syndrome					
Full sibling	75	77	22	23	
Other (half or adopted sibling)	10	67	5	33	
Gender of Sibling with Down Syndrome					0.730
Male	51	77	15	23	
Female	35	74	12	26	
Birth Order					0.970
Older than sibling with Down syndrome	73	76	23	24	
Younger than sibling with Down syndrome	13	76	4	24	

*= Statistically significant value

Siblings of non-Hispanic or non-Latino ethnicity were significantly more likely to report a significant adolescent courtesy stigma compared to Hispanic or Latino siblings (80% vs. 46% respectively, p=0.029). Similarly, more participants who listed their race as "White" reported experiencing courtesy stigma (78%) than did their non-White counterparts (62%), but this difference did not reach statistical significance (p=0.190). Notably, no significant differences in the amount of adolescent courtesy stigma experienced were found based on the age or gender of our participants. Nor did the education level, socioeconomic status, region where our participants were raised, birth order of our participant and their sibling with Down syndrome, or gender of the sibling with Down syndrome significantly impact the amount of courtesy stigma experienced by our participants.

Along with the significant difference in the overall amount of courtesy stigma experienced during the adolescent and adult periods that was found by this study, some differences in the demographic factors impacting adolescent and adult courtesy stigma were also noted. Differences in the levels of courtesy stigma that siblings reported experiencing in the present based on demographic characteristics are listed in Table 3.

Table 3. Current Courtesy Stigma by Demographic groups

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma		
	N	%	N	%	p value (chi-square)
Gender					0.090
Male	23	74	8	26	
Female	46	57	35	43	
Age					0.745
18-30	45	60	30	40	
31-71	24	63	14	37	
Race					0.970
White	61	61	39	39	
Non-White	8	62	5	38	
Ethnicity					0.998
Hispanic/Latino	8	62	5	38	
Non Hispanic/Latino	57	61	37	39	
Region Where Participant Grew Up					0.041*
Northeast	13	68	6	31	
Southeast	11	79	3	21	
Midwest	8	35	15	65	
Northwest	5	56	4	44	
Southwest	27	63	16	37	
International	4	100	0	0	
Region Where Participant Lives Now					0.148
Northeast	10	63	6	38	
Southeast	14	64	8	36	
Midwest	7	35	13	65	
Northwest	9	69	4	31	
Southwest	27	68	13	33	
Socio-economic Status					0.537
Upper class	3	75	1	25	
Upper-middle class	12	52	11	48	
Middle class	41	62	25	38	
Middle-lower class	13	68	6	32	
Lower class	0	0	1	100	
Education Level					0.611
Did not complete high school	1	100	0	0	
High school diploma/GED	3	38	5	63	
Some college	19	59	13	41	

Bachelor's degree	24	63	14	37	
Graduate degree	21	64	12	36	
Relationship to Sibling with Down Syndrome					0.064
Full sibling	63	65	34	35	
Other (half or adopted sibling)	6	40	9	60	
Gender of Sibling with Down Syndrome					0.148
Male	44	67	22	33	
Female	25	53	22	47	
Birth Order					0.837
Older than sibling with Down syndrome	59	61	37	39	
Younger than sibling with Down syndrome	10	59	7	41	

*= Statistically significant value

The geographical regions in which our participants were raised significantly impacted the amount of courtesy stigma that they reported experiencing as adults (p=0.041). Participants who grew up in the Midwest were the only group whose majority (65%) reported currently experiencing no courtesy stigma. The majority of participants from all other regions of the United States and abroad reported that they do currently experience some level of courtesy stigma, as listed in Table 3. An association between the region in which the participant currently lives and courtesy stigma also approached significance (p=0.148). Again, a majority of participants (65%) from the Midwest region reported currently experiencing no courtesy stigma, while the majority of participants from all other regions reported that they do experience some level of courtesy stigma.

Both the gender of our participants and the gender of their siblings with Down syndrome appear to play larger roles in the level of courtesy stigma they reported as adults than either variable did in their adolescent experience of courtesy stigma. More male participants (74%) reported

currently experiencing courtesy stigma than female participants (57%), an association that approached statistical significance (p=0.090). More participants who have a male sibling with Down syndrome (67%) reported experiencing courtesy stigma than those with a female sibling with Down syndrome (53%), although this association did not reach statistical significance (p=0.148). Another association (p=0.064) showed that the majority of participants who are full siblings to their sibling with Down syndrome reported currently experiencing courtesy stigma (65%) while a minority of half- and adoptive-siblings reported current courtesy stigma (40%). The lower stigma reported by Hispanic or Latino siblings in adolescence was not observed when looking at stigma in adulthood (p=0.998). Similarly, no significant difference in adult courtesy stigma was noted between racial groups (p=0.970). The age, education level, socio-economic status, and birth order of our participants remained insignificant in terms of the amount of courtesy stigma they reported experiencing currently.

Change in Courtesy Stigma Over Time

Another secondary hypothesis of this study was that the level of courtesy stigma experienced by siblings of people with Down syndrome would have decreased over the past several decades as our society became more accepting of people with intellectual disabilities. The data did not support this hypothesis for either adolescent courtesy stigma, as shown in Table 4, or current courtesy stigma, as shown in Table 5. While it is worth noting that the age distribution of our participant population was skewed, with notably more participants in the younger decade groups, no association between older age and greater courtesy stigma was found.

Table 4. Adolescent Courtesy Stigma by Age Groups

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma	
Age	N	%	N	%
18-20	15	79	4	21
21-30	41	73	15	27
31-40	11	73	4	27
41-71	19	83	4	17

The overall p value for the effect of age, by decade, on adolescent courtesy stigma is 0.938.

Table 5. Current Courtesy Stigma by Age Groups

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma	
Age	N	%	N	%
18-20	13	68	6	32
21-30	32	57	24	43
31-40	9	60	6	40
41-71	15	65	8	35

The overall p value for the effect of age, by decade, on current courtesy stigma is 0.852.

Effects of Aberrant Behavior on Courtesy Stigma

The final secondary hypothesis of this study was that higher levels of courtesy stigma would be reported when our participants' siblings with Down syndrome exhibited more socially undesirable behaviors.

Spearman correlation tests were calculated to estimate the associations between aberrant behavior in the past or present and adolescent and current courtesy stigma as well as the effects of both past and present behavior on current courtesy stigma. The results of these analyses are listed in Table 6.

Table 6. Spearman Correlations of Aberrant Behavior and Courtesy Stigma

	Past Aberrant Behavior	Current Aberrant Behavior
Adolescent Courtesy Stigma	r =0.451	N/A
Current Courtesy Stigma	r =0.414	r =0.484

Results show that greater adolescent courtesy stigma is moderately correlated (r=0.451) with higher past aberrant behavior scores, and greater current courtesy stigma is also moderately correlated (r=0.484) with higher current aberrant behavior scores. The relationship between past aberrant behavior with current courtesy stigma was slightly weaker (r=0.414) but still positively correlated. Overall, the hypothesis that higher levels of aberrant, or socially undesirable, behavior would be correlated with higher levels of courtesy stigma was supported by the data collected in this study. Stronger correlations may have been found if this study had recruited larger number of participants.

Courtesy Stigma and Comorbid Conditions

Mak and Cheung's 2008 study using the Chinese version of the survey used in the current study found a significant increase in courtesy stigma for caregivers of people with both an intellectual disability and autism over those who were caring for someone with an intellectual disability

only. In the current study, participants were asked whether their sibling with Down syndrome had also been diagnosed with autism as well as several other possible comorbid conditions. The impact of these various comorbid conditions on participants' adolescent courtesy stigma is listed in Table 7, and their impact on participants' current courtesy stigma is listed in Table 8.

Table 7. Impact of Comorbid Conditions on Adolescent Courtesy Stigma

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma		
Comorbid Condition	N	%	N	%	P value (Fisher's exact test)
Alzheimer Disease					1.000
Yes	4	80	1	20	
No	82	77	25	23	
Autism					0.571
Yes	4	100	0	0	
No	81	75	27	25	
Depression					1.000
Yes	9	75	3	25	
No	75	76	24	24	
Epilepsy					0.592
Yes	3	60	2	40	
No	82	77	25	23	
Lacks Verbal Communication					0.452
Yes	10	91	1	9	
No	76	75	25	25	
Obesity					1.000
Yes	16	76	5	24	
No	70	76	22	24	
Obsessive Compulsive Disorder					0.437
Yes	6	67	3	33	
No	75	77	22	23	
Uses Physical Aids for Mobility					0.671
Yes	5	71	2	29	
No	81	76	25	24	

None of the comorbid conditions assessed had a statistically significant impact on the levels of adolescent courtesy stigma experienced by our participants. Only four participants reported having a sibling diagnosed with both Down syndrome and autism. While 100% of these participants reported experiencing adolescent courtesy stigma compared to 75% of participants whose siblings had not been diagnosed with autism, the number of participants with autistic siblings was too small for this difference to reach statistical significance (p=0.571). Similarly, participants whose siblings with Down syndrome are unable to communicate verbally reported more adolescent courtesy stigma (91%) compared to their counterparts with verbal siblings with Down syndrome (76%), but again due to the small number of participants who have nonverbal siblings, this association failed to reach statistical significance (p=0.452).

Table 8. Impact of Comorbid Conditions on Current Courtesy Stigma

	Experienced Courtesy Stigma		Did Not Experience Courtesy Stigma		
Comorbid Condition	N	%	N	%	P value (Fisher's exact test)
Alzheimer Disease					0.077
Yes	4	80	1	20	
No	67	63	40	37	
Autism					0.153
Yes	4	100	0	0	
No	64	59	44	41	
Depression					0.761
Yes	8	67	4	33	
No	59	60	40	40	
Epilepsy					0.379
Yes	2	40	3	60	
No	66	62	41	38	
Lacks Verbal Communication					0.048*
Yes	10	91	1	9	
No	58	57	43	43	

Obesity					0.141
Yes	16	76	5	24	
No	53	58	39	42	
Obsessive Compulsive Disorder					1.000
Yes	6	67	3	33	
No	58	60	39	40	
Uses Physical Aids for Mobility					0.704
Yes	5	71	2	29	
No	64	60	42	40	

*=Statistically significant value

The only comorbid condition that reached statistical significance with respect to current courtesy stigma was verbal communication in the sibling with Down syndrome. Participants with nonverbal siblings reported more current courtesy stigma (91%) than did their counterparts with verbal siblings (57%) (p=0.048). As with adolescent courtesy stigma, having a sibling with both Down syndrome and autism led to courtesy stigma in adulthood for 100% of the four participants whose siblings have this diagnosis compared to 59% of participants who reported adult courtesy stigma with a non-autistic sibling. Again, because of the small number of participants with siblings with comorbid autism, this difference did not quite reach statistical significance (p=0.153).

Courtesy Stigma and Involvement

A secondary hypothesis of this study was that the level of courtesy stigma experienced by siblings during their adolescent years would be negatively correlated with their involvement with their siblings with Down syndrome in adulthood. Our initial plan was to be able to examine the trends of courtesy stigma and involvement within families, but we did not obtain enough sibling pairs or groups to allow for an effective analysis in this manner. Instead, associations across our

entire sample of participants were observed, and the results from these analyses are listed in Table 9 below.

Table 9. Effects of Adolescent Courtesy Stigma on Current Involvement

	Daily Contact [N (%)]	Weekly or Monthly Contact [N (%)]	Contact on Holidays or No Contact [N (%)]
In-Person (p=0.065)			
Experienced Adolescent Courtesy Stigma	18 (67)	40 (87)	26 (68)
Did Not Experience Adolescent Courtesy Stigma	9 (33)	6 (13)	12 (32)
By Phone (p=0.097)			
Experienced Adolescent Courtesy Stigma	16 (64)	50 (76)	20 (91)
Did Not Experience Adolescent Courtesy Stigma	9 (36)	16 (24)	2 (9)

The data did not support this particular hypothesis but rather showed mixed results. For in-person contact, the percentage of respondents who experienced adolescent courtesy stigma and maintain daily contact with their siblings with Down syndrome (67%) is nearly identical to the percentage of respondents who experienced adolescent courtesy stigma and rarely or never see their siblings with Down syndrome (68%). A somewhat higher percentage of the respondents who maintain weekly or monthly contact with their siblings with Down syndrome reported experiencing adolescent courtesy stigma (87%). These results appear to show that the level of contact between our respondents and their siblings with Down syndrome was not significantly affected by courtesy stigma experienced in adolescence.

The percentage of respondents who experienced adolescent courtesy stigma increased as the frequency of their phone contact with their siblings with Down syndrome decreased. Sixty-four

percent of respondents who maintain daily phone contact reported experiencing adolescent courtesy stigma, and this percentage increased to 76% in respondents who maintain weekly or monthly phone contact and 91% in respondents who rarely or never call their siblings with Down syndrome. While this association was notable, it failed to reach statistical significance (p=0.097). These results could indicate that the courtesy stigma experienced by some of our participants did not have a large enough impact on their relationships with their siblings with Down syndrome to significantly decrease the frequency of their involvement with their siblings as adults. Given the relatively low average courtesy stigma (1.25) experienced by the participants in this study, it is possible that this small amount of courtesy stigma does not have a large effect on their adult relationships with their siblings with Down syndrome.

DISCUSSION

Conclusions

The primary hypothesis of this study—that the majority of siblings of people with Down syndrome do experience courtesy stigma—and some of the secondary hypotheses, including the idea that aberrant behavior and higher levels of courtesy stigma are associated, were supported by the data. Other secondary hypotheses, such as the idea that courtesy stigma faced by the siblings of people with Down syndrome in our society would have decreased over time, failed to attain statistical support. The research team found that the majority of the siblings who participated in this study reported experiencing courtesy stigma both as adolescents and as adults. The percentage of participants who reported courtesy stigma, as well as the levels of courtesy stigma that they reported, were both higher in adolescence than adulthood. The higher

levels of courtesy stigma in adolescence compared to adulthood could be due to increased sensitivity to social pressures during this period or to the fact that more of our participants lived in the same home and had closer contact with their siblings with Down syndrome during this time in their lives.

While the majority of the siblings who responded to this survey did report experiencing courtesy stigma both currently and during their adolescence, the levels of courtesy stigma reported were lower than those found by Mak and Cheung when they used the same questionnaire to survey mothers of people with intellectual disabilities in Hong Kong in 2008. The scale developed and validated by Mak and Cheung in 2008 consists of twenty-two questions which are all scored on a four-point Likert scale where "Strongly disagree" equals one point, "Disagree" equals two points, "Agree" equals three points, and "Strongly agree" equals four points. The average level of courtesy stigma reported in the 2008 study performed by Mak and Cheung was 2.16, while the average in the current study was 1.25 for adolescent courtesy stigma and 1.14 for current courtesy stigma. The higher levels of courtesy stigma found by Mak and Cheung in 2008 could be due to the fact that they sampled mothers who were the primary caregivers for their children with intellectual disabilities and who were, therefore, all in daily contact with their affected children and responsible for meeting their daily needs. The siblings in this study were varied in their level of involvement with their affected siblings and were often not the primary caregivers who were responsible for their daily needs. Another possible explanation for the lower averages in the current study is that the 2008 study was conducted in a Chinese population. Cultural differences may play a role in how people perceive courtesy stigma as well as how comfortable they feel accurately reporting the level of this type of stigma that they have experienced. It may

be that courtesy stigma is a less acknowledged or accepted concept in the United States, and this bias could have influenced participants in the current study to report lower levels of courtesy stigma than their Chinese counterparts. Additional studies would be necessary to determine if either of these explanations may be the true cause of the lower courtesy stigma averages reported in the current study.

While there are several important variables differentiating the participant population of this study from the original cohort, it is encouraging to note that siblings in our society are reporting low levels of courtesy stigma. Along these lines, it was also heartening to note that experiencing courtesy stigma in adolescence did not appear to significantly reduce the amount of contact our participants maintain with their siblings with Down syndrome as adults. Unfortunately, due to a relatively small number of participants with a skewed age distribution, this study was not able to show a significant downward trend in the levels of courtesy stigma experienced by siblings of people with Down syndrome over time in our society.

Some associations with respect to the effects of various demographic factors on courtesy stigma were also found during data analysis. It is interesting to note that the region where our participants grew up did not appear to have a significant impact on whether they experienced courtesy stigma during their adolescence, but it did impact whether they reported experiencing courtesy stigma as an adult. This could represent a delayed effect of regionally varied upbringing on the perception of courtesy stigma, or it could be an inconsistency due to the relatively small number of respondents in this study. It was also noted that a larger percentage of full siblings of people with Down syndrome reported experiencing courtesy stigma than their half- or adoptive-sibling counterparts. This could be due to the fact that full siblings feel a closer association or

relationship to their siblings with Down syndrome than non-full siblings and, therefore, may be more sensitive to experiencing courtesy stigma. Additional studies would be needed to determine the true cause of this association.

A moderate correlation between aberrant behavior and higher levels of courtesy stigma, both in adolescence and adulthood, was found. Similarly, the comorbid conditions, such as autism, lack of verbal communication, or Alzheimer disease, that can be associated with aberrant, atypical, or socially undesirable behavior had the strongest effects on increasing the levels of courtesy stigma faced by our participants. These effects supported some of the secondary hypotheses of this study, since the research team expected that behaviors that tend to draw unwanted attention would increase the level of courtesy stigma experienced by siblings above the baseline level for Down syndrome. Previous studies, including Mak and Cheung's 2008 study, found that autism is a particularly stigmatizing condition for close relatives. This is likely due to the lack of verbal communication and the atypical or antisocial behaviors that many autistic individuals exhibit, all of which may attract negative and unwanted attention from others in public and increase the courtesy stigma felt by siblings and other close relatives. This could also explain the association towards more courtesy stigma in the siblings of nonverbal individuals. The other comorbid conditions may not have been present during adolescence, in the case of Alzheimer disease, or may have been less noticeable in public settings, where courtesy stigma is most relevant.

Overall, many associations were noted that failed to meet statistical significance in the small participant population of the current study but that may have done so in a larger population. The significant findings that were noted support the idea that, while many siblings of people with Down syndrome experience courtesy stigma in our society, they appear to be facing low enough

levels that their relationships with their siblings with Down syndrome are not significantly harmed.

This study has been educational in more ways than were anticipated at its conception. The data obtained from respondents were informative regarding the current and past experiences of siblings of people with Down syndrome with respect to courtesy stigma. In addition, through the comments written by respondents at the end of the survey as well as numerous correspondences with siblings and parents of people with Down syndrome via phone and email, I have learned a great deal about the strong bonds and loving familial relationships that these people share with their relatives with Down syndrome. It was unfortunate that some family members misconstrued the intent of the current study; however, it was also heartwarming to see how emphatically these people rushed to the defense of their children or siblings with Down syndrome when they perceived a bias against them. After speaking or exchanging emails with these individuals and explaining the true intent of the study, including the necessity of using a validated survey instrument, the research team was able to correct their misperceptions and assuage their concerns to the general satisfaction of all parties involved. This study has truly been a learning experience, not only about courtesy stigma but also about the strength of the bonds that parents and siblings share with individuals with Down syndrome.

Limitations

Sample Size

A total of 113 eligible participants completed our online survey. While some interesting trends were noted during the analysis of the survey data, few of these findings reached statistical significance. This could be due at least in part to the relatively small number of respondents who

completed the survey. Having a smaller number of total participants reduces the power of any statistical analyses performed, so it would be recommended for future studies of courtesy stigma in siblings to query larger numbers of siblings in order to be able to find significant differences between and among subgroups. Obtaining participants for this study was a challenge for the study team. Potential participants were reached through numerous Down syndrome associations and family support groups across the United States. It is more common for parents, rather than siblings, to be actively involved in the groups we contacted. It is also more common for parents of younger children with Down syndrome to be more actively involved in support groups and local associations, so these parents may not have any children old enough to be eligible for our study. Also, since we initially reached mostly parents of people with Down syndrome, we relied on these parents to pass information about our survey along to their eligible children and then on those individuals to choose whether or not to complete the survey. Being farther removed from our potential participants likely led to a lower response rate than we would have had if we had been able to reach eligible siblings directly. The same concept applied to the participants reached through UC Irvine's Down syndrome clinics. In many families, it was the parent of the individual with Down syndrome who attended the clinic, so we relied on these parents to pass information about our study on to their children. A larger sample size would have been ideal in this study and might have allowed the research team to obtain more findings of statistical significance.

Ascertainment Bias

Because participants were recruited through Down syndrome associations and support groups, the individuals who completed the survey may not be representative of the overall adult sibling

population in this country. Individuals and families who are involved in these groups may be more highly motivated and more closely bonded than those who are not involved. Therefore, this study was limited by a participation bias. In order for our study to reach eligible participants, these adult siblings must either be directly involved in a Down syndrome association or support group, be the primary caretaker of one of the patients in the UC Irvine Down syndrome clinic, or be in touch with their family members who meet one of the aforementioned criteria. This leads to a biased population of siblings who are more involved with their families and who likely share a closer relationship with their siblings with Down syndrome.

Our participant population was also skewed toward younger adult siblings. The mean age of our participants was 30.97 years, and the majority of our participants were age thirty or younger. This overrepresentation of younger individuals could be due to their greater affinity for internet use, which would have allowed for easier access to our online survey. Mailing out paper surveys to older siblings may have allowed us to reach a greater number of older respondents. Older siblings may also be more difficult to reach if their siblings with Down syndrome have passed away and they are no longer involved in support organizations. A participant population that was more representative of the actual population in our society in terms of age might have provided a more accurate representation of the levels of courtesy stigma siblings of people with Down syndrome have faced over the last several decades in our society.

Use of a Validated Instrument

Another limitation of this study was the necessary use of a validated instrument to measure courtesy stigma. The only available, validated instrument to measure courtesy stigma in family members of people affected with stigmatizing medical conditions was Mak and Cheung's

Affiliate Stigma Scale (2008). This scale was originally used in an Asian population to measure courtesy stigma, also referred to as affiliate stigma, in parents and caregivers of people with mental illness or intellectual disabilities. In order to retain the validity of this scale, the wording of the questions was changed only minimally to tailor the questions to siblings of people with Down syndrome. Many of the twenty-two questions that make up this scale are worded in a somewhat negative manner, and this unexpectedly raised concerns among many of the respondents to the survey. The questions of the original Affiliate Stigma Scale may have been worded in this negative way to reflect the fact that courtesy stigma is in itself a negative concept. The original wording may have also been more appropriate for the Asian population in which this instrument was originally validated. Ten participants in the current study voiced concerns about the negative wording of the survey questions in the Comments section provided at the end of the survey.

Participant Comments

A total of thirty-one participants made use of the Comments section at the end of this survey to communicate with the research team. Of the comments given, nine were positive remarks about the study, twelve were neutral, and ten voiced concerns about the negative wording used in the courtesy stigma section of the survey and generally regarded this study in a negative manner. The ten participants who commented on the negative wording of the courtesy stigma questions listed a range of underlying causes for their distress. Some respondents worried that the negatively worded questions reflected an underlying bias of the research team that people with Down syndrome are a detriment to their siblings or families and thought that the results of this study would be published in the hope of proving this idea. Such an idea of the research team's

attitude toward people with Down syndrome could not be farther from the truth; this study was intended to determine whether siblings of people with Down syndrome experience courtesy stigma. The outcome of this study does not comment on the value of people with Down syndrome within their families. Rather, this study was designed to comment on the level of courtesy stigma experienced by our respondents as an indirect measure of our society's acceptance of people with Down syndrome. In fact, it has become clear through the process of conducting this study and reaching out to numerous siblings and other relatives of people with Down syndrome that these individuals are highly valued and well loved by their families.

Another concern that was voiced by a few respondents concerned my position as a genetic counseling graduate student. Those who commented on my future profession, in the context of the negatively slanted wording of the survey questions, worried that it was my intent to use this study to influence potential future patients in the prenatal setting to abort babies found to have Down syndrome. One of the most important tenets of the genetic counseling profession, and one that is emphasized in the UC Irvine genetic counseling graduate program and to which I am committed, is to maintain a nondirective, unbiased role with our patients and their families. The intention of this study was not to comment on the value of people with Down syndrome but rather to measure the amount of courtesy stigma their siblings face in our society so that it can be addressed and alleviated.

The comments that regarded this study in a positive light included an appreciation for our use of person-first language throughout the survey, interest and excitement for the topic we were covering, interest to hear about our findings, an appreciation for taking an interest in siblings, and others. Many of the neutral comments consisted of brief descriptions of the participants'

siblings with Down syndrome with the apparent intention of providing the research team with a more intimate picture of the people with whom our participants grew up and the impact they have had on our participants' lives. After reading these comments, it is clear that our participants love their siblings with Down syndrome dearly and, whether or not they experienced courtesy stigma due to their association with these siblings, they clearly share a strong bond.

Likert Scale

The questions adapted from the Affiliate Stigma Scale (Mak & Cheung, 2008) were scored on a four-point Likert scale with choices including, "Strongly disagree," "Disagree," "Agree," and "Strongly agree." Unlike many typical Likert scales, this scale did not offer a neutral option. Likert scales have an advantage in that they collect quantitative data by asking for a degree of agreement or opinion, rather than a simple yes or no answer. However, the accuracy of Likert scales, or any measure of attitude, can be compromised by the influence of social desirability. Respondents may not answer survey questions entirely truthfully if they feel that their answers are socially unacceptable or would portray them in a negative light. In an attempt to combat the effect of social desirability, we assured all respondents that they would remain anonymous. We did not ask for any identifying information about the respondents themselves, and we included multiple reassurances that the identifying details that we did request (the first initial, last name, and birth date of their sibling with Down syndrome) would never be seen by the research team or linked to their other survey answers. However, even with the assurances we made regarding the respondents' anonymity, some social desirability bias may have been present in their answers to questions regarding the sensitive subject of courtesy stigma and their relationships with their

siblings with Down syndrome. Given the rather negative wording of the questions about courtesy stigma, respondents may have felt uncomfortable agreeing with such negative statements.

In addition, some researchers have found a central tendency bias, or an inclination of respondents to avoid the more extreme answers at either end of the Likert scale. In the current study, this bias does not appear to be present, since respondents chose "Strongly disagree," the most extreme option on the negative side of the scale, more often than any other choice. This propensity of respondents to overwhelmingly choose this extreme option may reflect the bias introduced by the negative wording of the questions. In the case of this study, it appears that the respondents' bias against the negative wording could have outweighed the central tendency bias of the Likert scale. It is also possible that the actual attitudes of the majority of our respondents was reflected in their consistent choice of the "Strongly disagree" option to the courtesy stigma questions because they truly did not experience this phenomenon.

Future Directions

Given the finding that the negative wording of the adapted Affiliate Stigma Scale questions used in this study offended some respondents, courtesy stigma should be reassessed in siblings of people with Down syndrome using a new scale that is created and validated in an American population. The new survey should contain a mixture of positively and negatively worded questions in order to properly measure courtesy stigma, a negative concept, in a more balanced way that does not offend and, therefore, does not bias respondents. It is a concern of the research team that the negative wording used in this study may have caused participants to respond with more strong disagreement to the questions than may accurately reflect their experiences.

In addition, once a more balanced tool for assessing courtesy stigma in siblings has been created,

it could be used to assess the experiences of siblings of individuals from various other stigmatized groups. If siblings of individuals with a range of different stigmatized conditions were queried, the levels of courtesy stigma they experienced could be compared among the various stigmatized conditions. This could provide a commentary on our society with respect to which conditions are most stigmatizing for the people close to affected individuals.

Another suggestion for the future direction of research in the realm of sibling courtesy stigma would be to include a control group. This control group would consist of people with typical siblings, or those unaffected by any stigmatizing conditions, and their responses to the courtesy stigma questionnaire could be used as a baseline against which to compare the responses of siblings of stigmatized individuals. This could help clarify whether the courtesy stigma reported by siblings of affected individuals is truly due to the stigma attached to the affected individual's condition.

One issue that was uncovered in some of the comments written by respondents and that deserves further study is the concern that my position as a genetic counseling graduate student, or future practicing genetic counselor, may have introduced a bias or underlying agenda into my study. It could be illuminating to delve further into the attitudes of relatives of people with Down syndrome, the most common chromosome abnormality discussed in prenatal genetic counseling sessions, toward genetic counselors. While attitudes and practices have improved over recent decades, the medical community has an unfortunate history of frequently breaking the news of a Down syndrome diagnosis to parents in a negative way (Skotko et al., 2006). In the prenatal setting, parents have at times been told by their obstetricians that they should abort a pregnancy when found to carry a child with Down syndrome, with no attempt to explore their feelings about

the situation or to educate them in an unbiased manner about the disorder (Skotko et al., 2009). In our current healthcare system, it is often genetic counselors and medical geneticists who do the majority of education for parents of babies with Down syndrome in pre- and postnatal settings. Both genetic counselors and medical geneticists are trained to educate parents about Down syndrome in an unbiased manner in the prenatal setting and in a positive manner in postnatal situations. Parents of babies found to have Down syndrome were sometimes told about the diagnosis in a way that devalued their new child (Skotko et al., 2009). These early experiences are often ingrained in parents' minds and, in some cases, create a distrust or dislike of medical professionals. Future research could assess the attitudes of both parents and siblings of people with Down syndrome toward genetic counselors as well as other types of medical professional. Such a study could examine whether there has been a change in attitudes over time. If this were true, relatives of younger individuals with Down syndrome may have more positive attitudes toward healthcare providers than relatives of older individuals with Down syndrome. Also, it would be interesting to compare the attitudes of parents versus siblings of people with Down syndrome toward genetic counselors and other medical professionals to see if their differing experiences and roles within the family affect their perception of healthcare professionals. One might hypothesize that parents would be more wary of medical professionals due to these negative experiences early in the life of their child with Down syndrome, as opposed to siblings who may not have been present for, or aware of, these situations.

REFERENCES

Ahmed, I., Ghafoor, T., Samore, N., & Chattha, M. (2005) Down syndrome: clinical and cytogenetic analysis. *Journal of the College of Physicians and Surgeons- Pakistan*. 15(7):426-429.

Ali, A., Hassiotis, A., Strydom, A., & King, M. (2012) Self stigma in people with intellectual disabilities and courtesy stigma in family carers: a systematic review. *Research in Developmental Disabilities.* 33:2122-2140.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985) The aberrant behavior checklist: a behavior rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency*. 89(5):485-491.

Ballesta, F., Queralt, R. Gomez, D., Solsona, E., Guitart, M., Ezquerra, M., Moreno, J., & Oliva, R. (1999) Parental origin and meiotic stage of non-disjunction in 139 cases of trisomy 21. *Annales de Genetique*. 42(1):11-15.

Bansal, V., Suresh, S., Suresh, I., Jagadeesh, S., & Fazal, G. (2010) Genetic counseling in chromosomal abnormalities. *Journal of Prenatal Diagnosis and Therapy.* 1(1):14-19.

Birenbaum, A. (1992) Courtesy stigma revisited. *Mental Retardation*. 30(5):265-268.

Brown, R. (2004). Life for adults with Down syndrome- an overview. *Down Syndrome Issues and Information*. Retrieved May 20, 2014, from http://www.downsyndrome.org/information/development/adults/.

Chiu, W. K., Akolekar, R., Zheng, Y., et al. (2011). Non-invasive prenatal assessment of trisomy 21 by multiplexed maternal plasma DNA sequencing: large scale validity study. *BMJ: British Medical Journal*. 342.

Collacott, R. A., Cooper, S. A., & McGrother, C. (1992) Differential rates of psychiatric disorders in adults with Down's syndrome compared with other mentally handicapped adults. *The British Journal of Psychiatry: The Journal of Mental Science*.161:671-674.

Corrigan, P., & Miller, F. (2004) Shame, blame, and contamination: a review of the impact of mental illness stigma on family members. *Journal of Mental Health.* 13(6):537-548.

Cuskelly, M. & Gunn, P. (2003) Sibling relationships of children with Down syndrome: perspectives of mothers, fathers, and siblings. *American Journal of Mental Retardation*. 108(4):234-244.

Cuskelly, M. & Gunn, P. (2006) Adjustment of children who have a sibling with Down syndrome: perspectives of mothers, fathers, and children. *Journal of Intellectual Disability Research*. 50(12):917-925.

Down, J. (1866). Observations on an ethnic classification of idiots. *Clinical Lecture Reports, London Hospital*. 3:259-262.

Down Syndrome Medical Interest Group. (2011). Down syndrome developmental milestones.

Goffman, E. (1963). *Embarrassment and Social Organization.* Hoboken, NJ: John Wiley & Sons Inc.

Handbook of Genetic Counseling: Down Syndrome/Trisomy 21. Retrieved May 20, 2014, from http://en.wikibooks.org/wiki/Handbook_of_Genetic_Counseling/Down_Syndrome_-_Trisomy_21-1.

Harris, P A., Thielke, R., Taylor, R., Payne, J., Gonzalez, N., &Conde, J. G.(2008). Research Electronic Data Capture (REDCap) - A metadata-driven methodology and workflow process for providing translational research informatics support. Journal of Biomedical Informatics, (doi:10.1016/j.jbi.2008.08.010).

Hattori, M., Fujiyama, A., Taylor, T.,Watanabe, H., Yada, T., Park, H., Toyoda, A., Ishii, K., Totoki, Y., Choi, D., Soeda, E., Ohki, M., Takagi, T., et al. (2003) Empathy and response to distress in children with Down syndrome. *Journal of Chlid Psychology and Psychiatry, and Allied Disciplines*. 44(3):424-431.

King, K., O'Gorman, C., & Gallagher, S. (2014) Thyroid dysfunction in children with Down syndrome: a literature review. *Irish Journal of Medical Science*. 183(1):1-6.

Larson, J., & Corrigan, P. (2008) The stigma of families with mental illness. *Academic Psychiatry*. 32 (2):87-91.

Lejeune, J., Gauthier, M, & Turpin, R. (1959). Les chromosomes humains en culture de tissus. *CR Hebd Seances Acad Sci*. 248: 602-603.

Leon, E., Zou, Y. S., & Milunsky, J. M. (2010) Mosaic Down syndrome in a patient with low-level mosaicism detected by microarray. *American Journal of Medical Genetics, Part A.* 152A(12):3154-3156.

Malone, F.D., Canick, J.A., Ball, R.H., et al., First- and Second-Trimester Evaluation of Risk (FASTER) Research Consortium. (2005). First-trimester or second-trimester screening, or both, for Down's syndrome. *New England Journal of Medicine*. 353(19):2001-2011.

Mak, W., & Cheung, R. (2008) Affiliate stigma among caregivers of people with intellectual disability or mental illness. *Journal of Applied Research in Intellectual Disabilities*. 21:532-545.

Nielsen, K. M., Mandleco, B., Roper, S. O., Cox, A., Dyches, T., & Marshall, E. S. (2012) Parental perceptions of sibling relationships in families rearing a child with a chronic condition. *Journal of Pediatric Nursing.* 27(1):34-43.

Orsmond, G. I., Kuo, H. Y., & Seltzer, M. M. (2009) Siblings of individuals with an autism spectrum disorder: sibling relationships and wellbeing in adolescence and adulthood. *Autism.* 13(1):59-80.

Orsmond, G. I. & Seltzer, M. M. (2007) Siblings with autism or Down syndrome: effects on adult lives. *Journal of Intellectual Disability Research*. 51(9):682-696.

Presson, A. P., Partyka, G., Jensen, K. M., Devine, O. J., Rasmussen, S. A., McCabe, L. L., & McCabe, E. R. (2013) Current estimate of Down syndrome population prevalence in the United States. *Journal of Pediatrics*. 163(4):1163-1168.

Ross, J. A., Spector, L. G., Robinson, L. L., & Olshan, A. F. (2005) Epidemiology of leukemia in children with Down syndrome. *Pediaric Blood & Cancer*. 44(1):8-12.

Ruparelia, A., Wiseman, F., Sheppard, O., Tybulewicz, V., & Fisher, E. (2010) Down syndrome and the molecular pathogenesis resulting from trisomy of human chromosome 21. *Journal of Biomedical Research*. 24(2):87-99.

Rusch N., Angermeyer, M., & Corrigan, P. (2005) Mental illness stigma: concepts, consequences, and initiatives to reduce stigma. *European Psychiatry.* 20(8):529-539.

Seltzer, M. M., Greenberg, J., Orsmond, G., & Lounds, J. (2005) Life course studies of siblings of individuals with developmental disabilities. *Mental Retardation.* 43(5):354-359.

Skotko, B., Capone, G., & Kishnani, P. (2009) Postnatal diagnosis of Down syndrome: synthesis of the evidence on how best to deliver the news. *Pediatrics*. 124(4):751-758.

Skotko, B., & Levine, S. (2006) What the other children are thinking: brothers and sisters of persons with Down syndrome. *American Journal of Medical Genetics*. 142(3):180-186.

Skoko, B., Levine, S., & Goldstein, R. (2011) Having a brother or sister with Down syndrome: perspectives from siblings. *American Journal of Medical Genetics*. 155:2348-2359.

stigma. 2014. In *Merriam-Webster.com*. Retrieved May 12, 2014, from http://www.merriam webster.com/dictionary/stigma.

Thies, W., & Bleiler, L. (2011). 2011 Alzherimer's disease facts and figures. *Alzheimer's and Dementia: The Journal of the Alzheimer's Association.* 7(2):208-244.

Wahl, O. (1989) Family views of stigma. *Schizophrenia Bulletin*. 15(1):131-139.

Zigman, W. B. (2013) Atypical aging in Down syndrome. *Developmental Disability Research Reviews*. 18(1):51-67.

APPENDIX A

Examining Courtesy Stigma in Siblings of People with Down Syndrome

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• You are being asked to participate in a research study to examine your thoughts and feelings about having a brother or sister with Down syndrome.
• You are eligible to participate in this study if you are 18 years of age or older and are the full-, half-, step-, or adoptive sibling of someone with Down syndrome.
• The research procedure involves an online survey that will take approximately 15 minutes to complete. You may complete this survey at your earliest convenience on any computer with internet access.
• Possible risks/discomforts associated with this study are minimal. Researchers do not expect the completion of this survey to cause physical or psychological distress.
• There are no direct benefits to you from participation in this study. However, this study may uncover and raise awareness about the courtesy stigma being experienced by siblings of people with Down syndrome.
• You will not be compensated for your participation in this research study.
• All research data collected will be stored securely and confidentially by UC Irvine's Institute for Clinical and Translational Sciences group. Once identifying data has been removed and anonymous codes have been assigned to subjects, then the UC Irvine research team will store the data in private files on UC Irvine's secure computer network.
• The research team and authorized UC Irvine personnel may have access to your study records to protect your safety and welfare. Any information derived from this research project that personally identifies you will not be voluntarily released or disclosed by these entities without your separate consent, except as specifically required by law.
• If you have any comments, concerns, or questions regarding the conduct of this research please contact the researchers listed at the top of this form.
• Please contact UC Irvine's Office of Research by phone, (949) 824-6662, by e-mail at IRB@research.uci.edu or by mail at 5171 California Avenue, Suite 150, Irvine, CA 92617 if you are unable to reach the researchers listed at the top of the form and have general questions, have

concerns or complaints about the research, have questions about your rights as a research subject, or have general comments or suggestions.
• Participation in this study is voluntary. There is no cost to you for participating. You may choose to skip a question or a study procedure. You may refuse to participate or discontinue your involvement at any time without penalty. You are free to withdraw from this study at any time.

Do you wish to participate in this study after reading the above information?
Yes No

The following 3 questions ask for identifying information about your sibling (brother or sister) with Down syndrome. This information will be kept confidential and ONLY used to link siblings from the same family together. This information will ONLY be seen by one independent UC Irvine employee and NOT by the research team.

First initial of your sibling with Down syndrome _____
Last name of your sibling with Down syndrome _____
Date of birth of your sibling with Down syndrome _____

Demographics

What is your age? _____
What is your gender?
- Male
- Female
- Prefer not to specify

What is your race?
- American Indian or Alaska Native
- Asian
- African American
- Pacific Islander
- White
- Other
- Prefer not to specify

What is your ethnicity?
- Hispanic or Latino
- Not Hispanic or Latino
- Prefer not to specify

In which region did you grow up?
- Northeast
- Southeast
- Midwest

Northwest
Southwest
Mexico
Canada
Other- International

In which region do you currently live?

Northeast
Southeast
Midwest
Northwest
Southwest

How would you describe the household you grew up in?

Upper class
Upper-middle class
Middle class
Lower middle class
Lower class

What is your level of education?

Did not complete high school
High school diploma or GED
Some college
Bachelor's degree
Graduate degree

How many total siblings (full, half, step, etc.) do you have? _____

How are you related to your sibling with Down syndrome?

Full sibling
Half sibling
Step sibling
Adopted sibling

How old is your sibling with Down syndrome? _____

What is the gender of your sibling with Down syndrome?

Male
Female

For how many years have you known your sibling with Down syndrome? _____

For how many years have you lived (or did you live) in the same home as your sibling with Down syndrome? _____

How often did/do you see your sibling with Down syndrome?

Currently
- Daily
- Weekly
- Monthly
- On holidays
- Rarely/never

When you were aged 0-12
- Daily

Weekly
Monthly
On holidays
Rarely/never

When you were aged 12-18
Daily
Weekly
Monthly
On holidays
Rarely/never

When you were aged 18-25
Daily
Weekly
Monthly
On holidays
Rarely/never

How often do you speak with your sibling with Down syndrome by phone?
Daily
Weekly
Monthly
On holidays
Rarely/never

Instructions: Below are some sentences related to your past and present life as a sibling of a person with Down syndrome. There are no right or wrong answers. Please read each sentence and choose the option which best represents your opinion.

I feel inferior because I have a sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I avoid communicating with my sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

Other people would discriminate against me if I am with my sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I feel emotionally disturbed because I have a sibling with Down syndrome.

Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I dare not tell others that I have a sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

My reputation is damaged because I have a sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

The behavior of my sibling with Down syndrome makes me feel embarrassed.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I reduce (or limit) going out with my sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

People's attitude towards me turns bad when I am together with my sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I feel helpless for having a sibling with Down syndrome.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

Given that I have a sibling with Down syndrome, I reduce contact with my friends and relatives.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

Having a sibling with Down syndrome imposes a negative impact on me.
Currently
Strongly disagree Disagree Agree Strongly agree
During childhood/adolescence
Strongly disagree Disagree Agree Strongly agree

I feel sad because I have a sibling with Down syndrome.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

When I am with my sibling with Down syndrome, I would keep a relatively low profile.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

Having a sibling with Down syndrome makes me think that I am incompetent compared to other people.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

I worry that other people would know I have a sibling with Down syndrome.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

I reduce interacting with my sibling with Down syndrome.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

Having a sibling with Down syndrome makes me think that I am lesser to others.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

I feel that I am under great pressure because I have a sibling with Down syndrome.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

I dare not participate in activities related to Down syndrome lest other people would suspect I have a sibling with Down syndrome.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

Having a sibling with Down syndrome makes me lose face (or lose respect).

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

Given that I have a sibling with Down syndrome, I reduce contact with the neighbors.

Currently

Strongly disagree Disagree Agree Strongly agree

During childhood/adolescence

Strongly disagree Disagree Agree Strongly agree

Please answer the following questions about your sibling with Down syndrome.

Does your sibling communicate verbally (with speech)?

Yes

No

Does your sibling use aids (e.g. cane, walker, wheelchair) for mobility and/or walk with a noticeable limp?

Yes

No

Is your sibling obese (very overweight)?

Yes

No

Has your sibling with Down syndrome been diagnosed with any of the following conditions?

Alzheimer disease

Yes

No

I am not sure

Autism

Yes

No

I am not sure

Depression

Yes

No

I am not sure

Obsessive Compulsive Disorder (OCD)

Yes

No

I am not sure

Schizophrenia

Yes

No

I am not sure

Epilepsy (seizures)

Yes

No
I am not sure

Please rate the past and present behavior of your sibling with Down syndrome. For each question, indicate whether the behavior was/is a problem, and choose the appropriate response.

My sibling is excessively active at home, school, work, or elsewhere
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling injures himself/herself on purpose
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling is listless, sluggish, inactive
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling is aggressive to other children or adults (verbally or physically)
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling seeks isolation (stays away) from others
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling makes meaningless, repeated body movements
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling is boisterous (inappropriately noisy and rough)
Present
No problem Slight problem Moderate problem Severe problem
Past
No problem Slight problem Moderate problem Severe problem
My sibling screams inappropriately
Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling talks excessively (too much)

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling has temper tantrums/outbursts

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling repeats the same abnormal behavior or movements over and over

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is preoccupied; stares into space

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is impulsive (acts without thinking)

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is irritable and whiny

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is restless, unable to sit still

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is withdrawn; prefers solitary activities

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling is odd; has bizarre or strange behavior

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling does not obey and is difficult to control

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling yells at inappropriate times

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

My sibling doesn't smile or frown often and doesn't respond to other people's emotions

Present

No problem Slight problem Moderate problem Severe problem

Past

No problem Slight problem Moderate problem Severe problem

Thank you very much for completing our survey.

If you have any questions or concerns, please contact Kelly Fulk at klharris@uci.edu.