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Having a Son or Daughter with Down Syndrome: Perspectives from Mothers and Fathers

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Abstract

This study asks parents who have children with Down syndrome how they feel about their lives so that such information could be shared with expectant couples during prenatal counseling sessions. A valid and reliable survey instrument was mailed to 4,924 households on the mailing lists of six non-profit Down syndrome organizations. Of the 2,044 respondents, 99% reported that they love their son or daughter; 97% were proud of them; 79% felt their outlook on life was more positive because of them; 5% felt embarrassed by them; and 4% regretted having them. The parents report that 95% of their sons or daughters without Down syndrome have good relationships with their siblings with Down syndrome. The overwhelming majority of parents surveyed report that they are happy with their decision to have their child with Down syndrome and indicate that their sons and daughters are great sources of love and pride.

Keywords

Down syndrome; prenatal diagnosis; parent; attitudes; trisomy 21

INTRODUCTION

When a new or expectant parent first learns about a diagnosis of Down syndrome (DS), a flashbulb memory is likely snapped—nearly always accurate, complete, and immune to forgetfulness [Brown and Kulik, 1977; Skotko, 2005b]. In one longitudinal prospective study, mothers who have children with Down syndrome were able to remember with 82% accuracy the original words that their physicians had used in describing the diagnosis some 21 years later [Carr, 1988]. The content of those words also matters; sometimes, a couple's decision to continue or terminate a pregnancy hinges on the information provided by their health care providers—however accurate or inaccurate, complete or incomplete, up-to-date or outdated. Powerful, then, is the information about DS conveyed by medical professionals.

In two recent evidence-based reviews, the Down Syndrome Diagnosis Study Group recommended that new and expectant parents receive accurate information that emphasizes in very practical terms what DS is, what causes the condition, and what it means to live with DS in today's society [Skotko, Capone, et al., 2009; Skotko, Kishnani, et al., 2009]. The

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American College of Obstetricians and Gynecologists (ACOG), American College of Medical Genetics (ACMG), National Society of Genetic Counselors (NSGC), National Down Syndrome Society (NDSS), and National Down Syndrome Congress (NDSC) also published a consensus document, stating that information and counseling about DS should be complete, consistent, nonjudgmental, and non-coercive [American College of Obstetricians and Gynecologists et al., 2009]. These recent calls for change were motivated, in part, by mothers who reported that the information they received about DS was oftentimes inaccurate, inadequate, and, in the worst cases, offensive [Skotko, 2005a; Skotko, 2005b; Skotko, Capone, et al., 2009; Skotko, Kishnani, et al., 2009].

What, then, should be said about DS? More specifically, what is the most accurate and up-to-date non-medical information that should be conveyed during the first discussion between a healthcare professional and a new or expectant couple? Popular books have provided a contemporary view of DS [Skotko and Kidder, 2001; Soper, 2009; Soper, 2007; Cohen et al., 2002; Groneberg, 2008; Berube, 1996; Beck, 1999; Zuckoff, 2002], but the anecdotal experiences are often singular, and most physicians have little time to digest these reading lists. National and local non-profit DS organizations have built extensive informative web pages, but, until now, their sections on parent attitudes have been limited to select families sharing their personal experiences.

Research exploring the parental impact of having a child with a disability have been challenged due to their methodological limitation of grouping together children with various disabilities [Dykens and Hodapp, 2001; Hodapp and Ly, 2005; Hodapp, 2007]. Drawing conclusions specific to families who have children with DS is difficult from these heterogeneous samples. In studies where children with DS are compared to children with other disabilities, researchers have noted a “Down syndrome advantage”: their families are described as warmer, closer, and more harmonious [Fidler et al., 2000; Thomas and Olsen, 1993; Seltzer et al., 1993; Abbeduto et al., 2004; Hoppes and Harris, 1990]; their parents experience less stress [Seltzer et al., 1993; Hodapp et al., 2001; Fisman et al., 1989; Dumas et al., 1991; Sanders and Morgan, 1997; Olsson and Hwang, 2003; Hodapp et al., 2003; Ricci and Hodapp, 2003; Piven et al., 1991; Most et al., 2006; Stores et al., 1998; Holroyd and McArthur, 1976; Ryde-Brandt, 1991] and fewer cases of depression [Abbeduto et al., 2004; Dumas et al., 1991; Ryde-Brandt, 1991; Wolf et al., 1989; Blacher and McIntyre, 2006]; their mothers and fathers are more confident about their parenting skills [Rodrigue et al., 1990] and claim that their children are easier to raise [Weinhouse et al., 1992; Marcovitch et al., 1986]; their parents report more marital satisfaction [Fisman et al., 1989; Rodrigue et al., 1990] with more satisfying networks of support [Seltzer et al., 1993; Hauser-Cram et al., 2001; Shonkoff et al., 1992]; their parents experience less overall pessimism [Fidler et al., 2000; Abbeduto et al., 2004] and report more positive experiences [Seltzer et al., 1993; Hoppes and Harris, 1990; Blacher and McIntyre, 2006; Goldberg et al., 1986] with fewer time restrictions [Olsson and Hwang, 2003]. Whether these difference are more attributable to higher parental incomes [Stoneman, 2007; Cahill and Glidden, 1996] or maturity that comes with advanced maternal age [Corrice and Glidden, 2009], both more likely among parents who have a son or daughter with DS, is unclear.

Yet during prenatal and postnatal counseling sessions, parents are not typically interested in how their situation is better than or different from having a child with another type of disability. Instead, they frequently ask how their family will compare to more “typical” families. In studies comparing families who have a child with DS to those who have a child without a disability, parents who have a child with DS report some comparative advantages: they are less likely to get divorced [Urbano and Hodapp, 2007; Cunningham, 1996]; their children are easier to raise [Marcovitch et al., 1987]; and parents experience greater satisfaction with support from friends and community groups [Erickson and Upshur, 1989].

They also report similarities: they have similar levels of marital satisfaction [Rodrigue et al., 1990; Rodrigue et al., 1992; Van Riper et al., 1992; Noh et al., 1989]; they have no differences in stress levels [Dumas et al., 1991; Stores et al., 1998; Duis et al., 1997]; their families are just as cohesive, adaptable, and communicative [Thomas and Olsen, 1993]; and they have similar levels of confidence in their parenting skills [Rodrigue et al., 1990; Rodrigue et al., 1992]. These parents have also reported some comparative disadvantages: their children tend to have more behavioral problems [Stores et al., 1998; Cuskelly and Dadds, 1992; Gau et al., 2008]; more time is spent on caregiving activities [Erickson and Upshur, 1989; Barnett and Boyce, 1995; Pueschel et al., 1991]; and some parents experience more stress, anxiety, and depression [Sanders and Morgan, 1997; Noh et al., 1989; Gau et al., 2008; Roach et al., 1999; Padeliadu, 1998]. Across many studies, an increase in child behavioral problems was the most significant predictor of parental stress, if existing [Hodapp et al., 2003; Most et al., 2006; Stores et al., 1998; Hauser-Cram et al., 2001; Cunningham, 1996; Bourke et al., 2008; Sloper et al., 1991].

Yet, lacking from all of these previous studies is the perspective of contemporary families who have children with DS. Previous studies have already described how mothers receive a prenatal diagnosis and the decisions they make with such information [Skotko, 2005b; Skotko et al., 2009; Skotko and Bedia, 2005]. This study seeks to extend that research by answering some of the most commonly asked questions during prenatal and postnatal counseling sessions: What is life actually like for parents who have sons and daughters with DS? How many of them love their son or daughter with DS? How many of them regret having their child? In this largest study to date, parents from selected states across the country respond.

MATERIALS AND METHODS

Participants

This study was nested in a larger cross-cultural, epidemiologic research project on family attitudes toward persons with DS. (Attitudinal perspectives from siblings and people with DS will be published separately.) The project was approved as protocol H-26552 by the Institutional Review Board of Boston University Medical Center. Five families (7 parents), recruited through non-profit DS organizations by the first author, participated in focus group testing of the preliminary survey instruments (Phase I). For validity and reliability testing of the questionnaires, all 300 families associated with the Down Syndrome Society of Rhode Island were invited to participate (Phase II). Because there is no national registry of individuals who have DS in the United States, the final survey instruments were distributed to all of the 4,924 family members of 6 non-profit DS organizations, chosen for their size, cultural compositions, and geographic distribution throughout the United States: Down Syndrome Association of Atlanta (757 members), Massachusetts Down Syndrome Congress (1,143 members), Mile High Down Syndrome Association (Denver, CO) (877 members), Triangle Down Syndrome Network (Raleigh, NC) (280 members), Down Syndrome Association of Central Texas (371 members), and Down Syndrome Association of Los Angeles (1,574 members) (Phase III).

Survey Instrument

Phase I: Piloting—As no already existing survey instruments gathered the information that we were seeking, we created a 5-page questionnaire for parents and guardians, informed by previous research [Skotko, 2005a; Skotko, 2005b] with direct input from participant representatives. The surveys gathered both quantitative and qualitative information, using open-ended questions and a series of Likert statements on a scale of 1 to 7 (with “7” indicating strong agreement and “1” indicating strong disagreement). The questions

measured parent attitudes and their perceptions of their child's functional abilities, health condition, and educational challenges. As optional measures, parents were asked to report sociodemographic information (see Appendix online for full survey).

Phase II: Validation & Reliability Testing—Following each of the two focus group sessions, the research tools were edited for clarity. The revised questionnaire was then distributed along with an already validated, published survey—the Perception of the Impact of the Child's Chronic Illness on the Parents Questionnaire [Katz, 2002]—to all members of the Down Syndrome Society of Rhode Island for validity and reliability testing. Those who responded to the first mailing received an identical questionnaire 4 weeks later for test-retest reliability. In total, 82 responses (27%) were received after the first mailing, and 26% of these respondents completed the second mailing.

Construct validity was determined by assessing convergent and discriminant validity. Convergent validity was defined *a priori* as a Pearson's correlation between $|0.4|$ and $|0.6|$ between similar constructs, identified as the summative scores of questions 2, 9, and 10 on the already validated survey [Katz, 2002] and the summative scores of questions 12, 14, and 18 on our survey. The survey met our criteria for convergent validity ($r = -0.49$, $N = 56$). Discriminant validity was defined *a priori* as a Pearson's correlation between -0.4 and $+0.4$ between dissimilar constructs on our survey instrument, identified as the summative scores of questions 10 and 11 (positive construct) and questions 12 and 14 (negative construct). The survey met our criteria for discriminant validity ($r = -0.11$, $N = 80$).

Reliability was determined by measuring internal consistency reliability and test-retest reliability. Internal consistency reliability was defined *a priori* as a Cronbach's $\alpha \geq 0.6$ on questions measuring similar constructs. Questions 10 and 11 comprised the positive construct, with a Cronbach's $\alpha = 0.8$; questions 12 and 14 comprised the negative construct, with a Cronbach's $\alpha = 0.6$. As such, the survey met our criteria for internal consistency reliability. Test-retest reliability was assessed for each survey question with reliability defined *a priori* as $>80\%$ of participants responding ≥ 1 point difference on our 7-point Likert scale. Five questions failed to meet this reliability and were eliminated from our final survey instrument used for national distribution. The responses from our participants in our validity and reliability testing were not part of our final data analyses. The final survey had a Flesch-Kincaid grade level of 6.9; the survey was also translated into Spanish and checked for accuracy.

Phase III: Study—The final surveys were distributed to parents in 6 states using evidence-based best practices: We provided self-addressed stamped envelopes [Edwards et al., 2002]; nonrespondents were mailed a second copy of the questionnaire approximately 6 weeks after receiving the first copy [Edwards et al., 2002; Nakash et al., 2006]; the questionnaire packet was mailed on university stationery [Edwards et al., 2002]; and the survey was kept purposely short to encourage completion [Edwards et al., 2002; Nakash et al., 2006]. Two parent/guardian surveys were included in each packet, with additional copies available for download from a secure, password-protected web site when a person with DS had more than two parents/guardians. All of the contents in each mailing were labeled with a unique identifier number so that responses within family members could be linked for subsequent analyses. The survey packets were mailed to the non-profit DS organizations, who then forwarded the mailings to their members.

Data Analyses

Means and standard deviations were calculated for each of our parental attitudes, measured on the 7-point Likert scale. To examine the relationships between parental attitudes, the

correlations were calculated with significance designated at p values of .05, .01, and .001. A composite functional activity score was calculated for each child by summing the 7-point Likert statements of each functional activity (e.g., walking, preparing meals, going on dates). Any response of “not applicable” was assigned a score of “1” with the composite functional activity score ranging between 7–77.

To investigate which variables might best predict the 11 different attitudinal responses of parents, we performed mixed stepwise, multivariate regression analyses. Variables were entered at the probability level of .05, and the standardized β and R^2 are reported. To determine significance of our models, ANOVA was performed and df , F , and p values for the models that achieved significance at .05 level are reported. The independent variables included the composite functional activity score divided by age of the child, the health conditions score, the educational challenges score, and sociodemographic variables (race, state, religion, living situation for the person with DS, parental income, parental age at survey completion, parental age at birth of son/daughter with DS, parental gender, number of children in family, age of son/daughter with DS, gender of son/daughter with DS, birth order of son/daughter with DS, marital status, and biological status of son/daughter with DS).

The responses to the two open-ended questions were coded by the first two authors using the Constant Comparative Method of Qualitative Analysis [Glaser and Strauss, 1967]. Thematic saturation was achieved after reviewing 40 responses for the first question and 50 responses for the second question. *De novo* themes were not subsequently identified. At least one author coded 997 random surveys for each question; approximately 10% were blindly coded by a second author with coding agreement achieved at 96%. Differences were discussed, and mutual agreement was obtained. We report those themes that were mentioned by at least 5% of respondents.

RESULTS

Respondents

Of the 4,924 families invited to participate in this study, we received 2,044 responses from parents and guardians, which represents 1,407 surveys from at least one parent or guardian in each household, a 29% response rate. Of the 2,044 responses, 54 declined to respond, and 1 was from a person living outside of the U.S., leaving 1,989 surveys for inclusion in our analyses. Of the 412 surveys translated into Spanish mailed to known Spanish-speaking families, we received 78 responses coming from 54 Spanish-speaking household, a 13% response rate.

The average age of the parent or guardian responding to the survey was 46.4 years ($N=1,939$; $SD=11.0$). The parents were, on average, 34.2 years old when their son or daughter was born ($N=1,931$; $SD=5.8$). At the time of survey completion, parents had, on average, 2.8 children ($N=1,978$; $SD=1.4$) with a mean gross household income of \$109,815 ($N=1,553$; $SD=89,570$). The median gross household income was \$100,000.

As can be seen in Table I, respondents were proportionally distributed by geography, and there was diversity among Hispanic origin, race, and religion. The majority of parents had received a college/university degree or higher. Most respondents were married mothers. Respondents had sons and daughters with DS of all ages, the majority of them still living at home.

Parental Feelings Toward Son or Daughter with Down Syndrome

The overwhelming majority of parents love their son or daughter with DS and are proud of them (Table II). Marital status had a significant association with the rating of love, but only to varying degrees of affirmation ($Love = 6.3 - 0.1 \text{ divorced} + 0.1 \text{ married, single, widowed, or with partner}$ ($R^2 = 0.01$, $F[0.05;2;1923] = 9.37$, $p < 0.001$)). Put another way, divorced parents would be predicted to respond, on average, with a 6.2 on the 7-point Likert scale, while parents of other marital status would be predicted to respond with a 6.4. The parental perceived level of learning difficulties was associated with the rating of pride, but only to varying degrees of affirmation ($Pride = 7.1 - 0.1 \text{ learning difficulties}$ ($R^2 = 0.04$, $F[0.05;4;1865] = 18.1$, $p < 0.001$)). Stated alternatively, parents who strongly agreed that their child had significant educational/learning difficulties would still be expected to respond with a 6.4, on average. Of all of our parents who responded, 52% agreed that their son or daughter had some degree of “significant educational/learning difficulties” ($N = 1,922$; $M = 4.5$; $SD = 1.6$). Parents who expressed love for their child were also likely to express pride (Table IV).

Having significant learning challenges as perceived by parents was associated with higher ratings of embarrassment ($Embarrassed = 1.2 + 0.1 \text{ learning difficulties}$ ($R^2 = 0.04$, $F[0.05;6;1819] = 12.8$, $p < 0.001$)). Yet, few parents/guardians felt embarrassed by their son or daughter, and fewer expressed regret for having their son or daughter with DS (Table II). Those who did express regret were more likely to have children with greater perceived health problems, learning difficulties, or were of Jewish background ($Regret = 0.9 + 0.1 \text{ health problems} + 0.1 \text{ learning difficulties} - 0.2 \text{ Catholic, Protestant, Mormon, atheist, or other religion} + 0.2 \text{ Jewish}$ ($R^2 = 0.07$, $F[0.05;5;1754] = 29.3$, $p < 0.001$)). Approximately 19% of all parents agreed that their son or daughter with DS had “significant health problems” ($N = 1,947$, $M = 2.8$; $SD = 1.8$). Parents who were embarrassed were more likely to express regret (Table IV). Of the parents who reported some degree of regret, the average age of their son or daughter was 15.1 years old ($N = 88$, $SD = 11.4$). Of these parents, 23% had children 5 to <10 years old; 24% had children 10 to <20 years old; and 31% had sons and daughters 20 years old.

Perceived Impact of Son or Daughter with DS on Family and Community

The majority of parents felt that their outlook on life is more positive because of their son or daughter with DS (Table III). Strongest endorsement for this statement came from parents who had more children and whose son or daughter had fewer medical problems and learning difficulties. Parents of Hispanic origin and/or of a lower educational background were also more likely to report highly positive outlooks. There were also associations between a parent’s outlook and his or her identified religion and demographic origin ($Positive outlook = 6.8 + 0.1 \text{ number of children} - 0.1 \text{ medical problems} - 0.2 \text{ learning difficulties} + 0.2 \text{ Spanish/Hispanic/Latino} - 0.1 \text{ educational level} - 0.2 \text{ Jewish or atheist} + 0.3 \text{ Protestant} + 0.2 \text{ Mormon or other religion, not Catholic} - 0.4 \text{ Texas} + 0.2 \text{ Massachusetts or North Carolina}$ ($R^2 = 0.08$, $F[0.05;11;1,679] = 13.7$, $p < 0.001$)).

A small number of parents reported that their sons or daughters were putting a strain on their marriage or partnership. Parents who perceived their children with DS to have significant medical conditions and learning difficulties were more likely to report marital strain ($DS strain on marriage = 1.0 + 0.1 \text{ medical conditions} + 0.2 \text{ learning difficulties} - 0.2 \text{ California, Colorado, Massachusetts, Texas} - 0.1 \text{ black/African American, American Indian/Alaska Native, Native Hawaiian, other, multiple races} + 0.1 \text{ white, Asian}$ ($R^2 = 0.08$, $F[0.05;6;1,684] = 25.8$, $p < 0.001$)), but an equal percentage of parents reported that their other sons and daughters without DS were putting a strain on their marriage or partnership ($Other children strain on marriage = 1.0 + 0.1 \text{ father responding} + 0.1 \text{ medical problems} +$

0.1 *learning difficulties* + 0.1 *educational level* ($R^2 = 0.05$, $F[0.05;7;1,511] = 13.0$, $p < 0.001$). Of the parents who agreed that their sons/daughters with DS were putting a strain on their marriage, 44% of these parents also agreed that their other children without DS were putting a strain on their marriage, with a significant correlation between the two responses (Table IV).

An overwhelming majority of parents feel that their sons and daughters without DS have a good relationship with their sibling who does have DS. The degree of the positive relationship was associated, to a small extent, with the degree of the learning difficulties that the parents perceived in their child with DS (*Good sibling relationship* = 5.2 – 0.1 *learning difficulties* ($R^2 = 0.06$, $F[0.05;5;1,561] = 20.9$, $p < 0.001$)). The majority of parents also feel that their sons and daughters without DS are more sensitive and caring because of their son/daughter with DS (*Caring siblings* = 5.3 – 0.1 *father responding* + 0.1 *number of children* – 0.2 *atheist or multiple religions* + 0.2 *Catholic, Protestant, Mormon, Jewish, other religion* + 0.2 *Live at home* ($R^2 = 0.04$, $F[0.05;6;1,504] = 11.9$, $p < 0.001$)). And, an overwhelming number of parents feel comfortable in responding to their children’s questions about DS. Race and parental gender were associated with varying levels of comfort (*Comfortable with answers* = 6.1 – 0.2 *black or African American* + 0.2 *all other races* – 0.1 *father responding* ($R^2 = 0.02$, $F[0.05;4;1,579] = 9.8$, $p < 0.001$)).

Finally, about half of parents and guardians agreed that non-profit DS organizations were helpful to them. Fathers, parents of Mormon, Jewish, or atheist backgrounds, and parents whose child with DS was the youngest were more likely to perceive the non-profit DS organizations as being less helpful (*Parent support groups* = 4.2 – 0.3 *fathers* + 0.3 *Spanish/Hispanic/Latino* + 0.1 *DS only child* – 0.2 *DS youngest child* – 0.2 *Mormon, Jewish, or atheist* + 0.2 *Catholic, Protestant, other religion, multiple religions* – 0.2 *live at home* ($R^2 = 0.09$, $F[0.05;9;1,594] = 17.6$, $p < 0.001$)). No geographic variances were significant in the parental perceptions of the non-profit DS organizations.

Perceived Life Lessons

Of the 997 random surveys qualitatively analyzed, 943 parents responded (95%) to the open-ended question asking them to share life lessons learned from their sons/daughters with DS (Table V). Chief among the responses were instructions on self-growth: “I’ve learned to live more in the present and worry less about the future”; “Life is all about attitude and perspective. Sometimes the people we think need the most help are actually the ones providing ...help to the rest of us”; “I’ve redefined the way I measure success in my life. It’s not based on material things, money, or power. It is based on family happiness, taking care of each other...”; “My definition of normal has changed”; “I look at people with less prejudice, but see the potential in everyone.”

Many parents described lessons in patience (e.g., “I’ve learned the good lessons of patience and that its rewards are a smile – and that is always enough”) and acceptance (e.g., “All people have something to offer”; “I’ve learned that a person’s worth is not measured by an IQ score”; “Simplicity can be astonishingly wonderful”). Many described coming to a greater understanding of love (e.g., “Love conquers all”; “Unconditional love is the best kind”; “Love and understanding as well as patience are the ingredients for happiness”; “Often the sweetest love comes from those that depend on us so much and cause us a little extra work and worry”) and joy (e.g., “Life’s enjoyment truly comes from celebrating the simple accomplishments...”; “Through reflux, eye surgeries ... using a walker every day for a year and a half, he greets every day with a smile. He loves everyone”; “...our son is the greatest joy and motivation of our lives”; “It’s amazing how I can feel such joy and elation even in the midst of harder frustrating times—he just makes it worth it”).

Some parents learned how to advocate (e.g., “You have to be very aware of your child’s rights and be an advocate for them”; “I’ve learned to be vocal and forget embarrassment”; “There is a tendency to be more protective of my son...he is more vulnerable to being injured or hurt by other kids”). Others learned to set higher expectations for their child (e.g., “Don’t limit his possibilities because his possibilities are limitless”; “Expect miracles”; “Keep your expectations of her [and all children’s] abilities very high. She amazes me every day with things she has learned that have not been directly taught to her”; “When you don’t expect much, you won’t get much”).

A small percentage of parents learned negative life lessons (e.g., “Life is not fair”; “People can surprise you. Some friends will really be there. Some will disappear”; “While most people are nice to him, some people still harbor old prejudices and will...not take the time to know him”).

Advice for Other Parents

Of the 997 random surveys qualitatively analyzed, 970 parents responded (97%) to the open-ended question asking them about advice they would offer to a couple expecting a child with DS (Table VI). A lot of parents indicated that they would describe the joys and rewards of raising a child with DS (e.g., “He lights up the room with his joy and sense of delight”; “Each milestone is a thrilling occurrence”; “The joy their child will bring will be just like the joy that any child brings to a family”; “Your life is about to change in an interesting and wonderful way!”). Many affirmed that the expectant parents would love their child (e.g., “Your life is going to be filled with joy, love, and lots of great surprises”; “Our love is the greatest gift we can give our daughter—she has provided that and more in return”; “You will love this child desperately, and your life will have purpose”). Yet, nearly an equal number said that they would also describe some of the struggles and challenges (e.g., “It will exhaust you. It will challenge you...you will take nothing for granted”; “They are about to have the hardest job they’ve ever loved”; “You will begin a lifelong journey filled with joys and triumphs. Challenges and hardships too, but they are more than outweighed by the overwhelming love you will feel for this baby”). Many others would describe how life would go on and things would become okay (e.g., “Relax. Take it one day at a time. Your life will be so much richer for having this child in your life”; “Your child will take you to familiar and unfamiliar places, but you will be a better person because of this journey”; “What at first appears to be the worst possible thing that could be happening, can turn into the best possible thing”; “Don’t sweat it. It will be alright”; “The day will come when Down syndrome will not consume you”).

On a practical level, many parents recommended that expectant parents seek out resources and support groups (e.g., “Contact the local Down syndrome parent association for information, resources and support. Find an early intervention program”; “I learned more from [talking to] other parents than from reading books, etc.”; “Be involved in the local DS group to get information and be hooked up with other parents”; “Do take advantage of every conference, seminar, workshop regarding advancements for children with DS”; “Support groups and associations are helpful to prepare yourself as your child’s advocate”). Others mentioned the importance of finding a good physician when their child is born (e.g., “Find positive thinking doctors...”; “Look for the pediatrician who has a good knowledge of Down syndrome”). Expectant parents would also learn how to advocate and teach (e.g., “Recognize the importance and the challenge of providing a strongly stimulating environment and an active schedule for your child...”; “When asking for support services in school for our child, we never take no for an answer”; “It’s up to you, the parent, to be in charge of every aspect of your child’s life and be proactive”).

Parenting a child with DS will take patience (e.g., “The first [and hardest] lesson to learn was patience. My child has taught me to slow down”; “Accept the slow pace at which change takes place—compared to your other children—and be sensitive to the steady progress your child is making, despite the many barriers she faces”; “Be patient. Don’t overreact”), laughter (e.g., “Learn to laugh early on”; “You will cry and laugh daily, and you will learn as you go”; “Laugh at the funny things rather than be embarrassed), and commitment (e.g., “It will be a lifelong commitment”; “Both [parents] must be willing to accept the hard work that lies ahead and be strongly committed”).

A small percentage of parents would share negative feelings (“Life is very hard generally, and with a Down syndrome kid, it’s harder”; “It’s hard for me to say everything will be okay. Not true with a kid with Down syndrome/autism”; “Can we all please stop pretending that we’re all ‘doing great’ all the time and actually say ‘I’m miserable’ sometimes and then get on with our day anyway?”; “Your child will have challenges that will likely challenge your family”; “It will be harder on your marriage, but you will find much more joy and happiness.”; “It will exhaust you. It will challenge you. You will have to fight school systems and insurance companies. You will face discrimination, and no one will care”). A few encouraged termination (“Terminate the pregnancy if still in the first six weeks”; “Abort it”; “Think it over carefully”).

DISCUSSION

Overall Results

Clinicians should include up-to-date, accurate, and complete information about DS in their counseling to new or expectant parents, according to recent evidence-based guidelines and a national consensus statement [Skotko, Capone, et al., 2009; Skotko, Kishnani, et al., 2009; American College of Obstetricians and Gynecologists et al., 2009]. This large study of more than 2,000 parents and guardians from 6 different states provides perspective on the experience of having a son or daughter with DS.

Nearly all of them responded that they love their child and are proud of his or her accomplishments. Parents cited lessons in patience, acceptance, and flexibility. They learned to be more tolerant, kind, and empathetic. Mothers and fathers noted that their perspectives on what is important in life changed, and they gained an appreciation and respect for differences in other persons. Laughter, joy, and celebration were frequently mentioned as part of their family experience.

While marital status and the degree of the child’s learning challenges were associated, to a small extent, with the degree of love and pride felt by the parent, striking were the variables that were not. The child’s functional activities (e.g., speaking, reading, grooming, living independently) were not predictors for how much love or pride was felt by the parent. In short, parents who identified their children as having limited functional abilities reported as much love and pride as parents who perceived their children to be high-functioning.

Previous survey research has shown that global ratings of health provide an accurate measurement of health status [Idler and Benyamini, 1997]. We found that the parents’ perceptions of their child’s health were not associated with their ratings of love or pride. Further, parents equally reported to love their son or daughter with DS regardless of race, educational level, income level, or religion.

An overwhelming majority of parents also believed that siblings were more caring and sensitive because they had a brother or sister with DS. The relationship between the children was almost uniformly described as positive, not associated with the degree of medical

problems in the child with DS and not related to his or her level of functional activities. In short, parents whose children with DS had complex medical needs and/or limited functional abilities felt their other children were still as caring and sensitive as those with siblings who were higher-functioning or healthier. In a separate study, we will be analyzing whether the siblings perceived this to be true, as well.

Of course, not all parents believed that their lives were enhanced. About 11% of parents felt that their son or daughter with DS was uniquely putting a strain on their marriage (Table III). On the whole, however, parents who have children with DS are less likely to get divorced when compared to parents who do not have children with DS [Urbano and Hodapp, 2007]. Only 4% of parents regretted having their child altogether (Table II). These parents were more likely to report complex health problems and significant learning difficulties in their son or daughter. And, while not surveyed, we suspect that many of these families might also have reported significantly difficult-to-manage behavioral problems in their son or daughter, which have been described as a source of parental stress [Hauser-Cram et al., 2001]. Of course, the parents might also have regretted just having a child, in general, regardless of his or her disability.

Interestingly, the majority of parents who expressed regret had older children with DS. Children with DS born today have many more social, educational, and vocational opportunities than their previous counterparts. Support structures are more developed, and parent resources more numerous. That fewer new parents express regret may be due to greater available social supports. Another explanation is that new parents experience more prenatal diagnostic opportunities than before, with the possibility that these parents who received a prenatal diagnosis of DS and choose to continue their pregnancy might be more resolute in the parenting of their son or daughter. Alternatively, some parents might not develop regret until their children get older, when complex behaviors, medical conditions, and learning difficulties emerge and when issues of lifelong dependence are clearer. The parent of an older child with DS may face greater frustrations with inflexible school systems or scant employment opportunities. Whatever the reason(s), this finding underscores an important charge for all clinicians and non-profit DS organizations who work with parents who have children with DS: a small, albeit real, number of parents have regrets and may need specific outreach and supports.

While all of our parents and guardians were sampled from the mailing lists of non-profit DS organizations, just more than half of the respondents stated that these organizations were helpful. One possibility is that some of these parents on the mailing lists might not take advantage of the opportunities because of lack of interest or scheduling conflicts. Others, however, might actively participate and just not find the opportunities beneficial.

The survey participants who did find the organizations most helpful were mothers (rather than fathers), parents of Catholic, Protestant, “multiple” faiths, or “other” religions (rather than Mormon, Jewish, or atheist backgrounds), parents who identified themselves as Spanish/Hispanic/Latino, and parents whose child with DS was the only child. One explanation could be that these sub-populations more regularly participated in the non-profit organizations. For example, more mothers than fathers might participate in the non-profit organizational activities, making them more likely to appreciate the programs and services. The non-profit DS organizations are also increasingly providing information and resources online. Research has shown that women are more likely than men to gather health information online [Fox, 2011]. Additionally or alternatively, some of these sub-populations might desire—or need—more assistance than others. For example, parents who have an only child with DS might appreciate more parenting support than experienced parents who already have many children, the youngest of whom has DS. Similarly, parents who identify

themselves as Spanish/Hispanic/Latino might appreciate the assistance from non-profit DS organizations if access to healthcare and resources is limited by language barriers. Still another explanation on why certain sub-populations valued the non-profit DS organizations more could be that these organizations do a better job in providing programs and services for certain subpopulations—or have members that are more homogenous in certain sociodemographic characteristics. Regardless, this finding is an important charge to non-profit DS organizations: more polling is needed to better satisfy and serve the needs of parent and guardian members. Already, many organizations are beginning to recognize that their members' needs are varied, as programs such as the “Dads Appreciating Down Syndrome” are being created (<http://www.dadsnational.org/>).

Limitations of Current Study

This study is subject to selection bias. Only parents who were on the membership lists of non-profit DS organizations were sampled, making it possible that their views are not representative of all parents who have sons and daughters with DS. While being on the mailing list of a non-profit DS organization does not mean that the person attends meetings or subscribes to the views of the majority of its members, there may be some parents and guardians with differing perspectives beyond the reach of such organizations. Unfortunately, no national population-based registry exists for people with DS, although legislative efforts are in place to create one, according to the National Down Syndrome Society Policy Center (www.ndss.org). Until such time occurs, the most robust way of surveying parents and guardians is through non-profit DS organizations, as has been done with similar published research [Skotko, 2005a; Skotko, 2005b]. We purposely invited every member of the non-profit DS organizations to participate in our research, so as not to further restrict the selection bias within the organizations. We also took particular care to invite only those members of the sampled non-profit DS organizations, so as to not allow unfettered selection bias that might have occurred, for example, with an open-invitation web-based survey. In the end, though, more than 2,000 parents and guardians participated in this research, making the results powerful, although limited by the diversity of respondents.

This study is also subject to non-response bias. Our response rate of 29%, however, is high by research standards, with the nationally respected Pew Research Center citing between 15–25% response rates on their studies [The Pew Research Center for the People & the Press, 2010]. Emerging research shows that the response rates on national surveys have been declining over time and that lowered response rates do not necessarily reflect lower survey accuracy [Singer, 2006]. The non-profit DS organizations do not collect reliable and robust demographics on their members, so we were unable to know how representative our participants were of their members, at large.

Our results are also limited by the lack of diversity of our respondents, which did not include many black/African American, Asian, American Indian, or Alaska Native Americans. Our results also suggest that the average respondents were from middle- to upper-class families. Our median gross household income of \$100,000 was significantly higher than the national median gross household income of \$49,777 reported in the last Census income publication in 2009 [DeNavas-Walt et al., 2010]. Until non-profit DS organizations diversify their memberships or until a population-based DS registry is created, family epidemiologists will have participants with largely homogenous cultural compositions. Our results did, however, appropriately represent Spanish/Hispanic/Latino Americans. Approximately 14% of our respondents identified themselves as Spanish/Hispanic/Latino, which compares to the 16% of Americans who did so in the last U.S. Census [Humes et al., 2010]. We purposely oversampled in areas of the country with higher numbers of Spanish/Hispanic/Latino Americans, and we also made all of our survey materials available in Spanish. To this

extent, the diversity of our respondents represents an improvement over previously published studies [Skotko, 2005a; Skotko, 2005b].

Future Research

While this study remains the largest of its kind in scope, the results are representative of only the sampled populations. Research from the United Kingdom, Scotland, Ireland, Spain, Sweden, and the Netherlands all indicate that new and expectant parents are dissatisfied with the quality of information that they have received about DS from their healthcare providers [Skotko, Capone, et al., 2009a; Skotko, Kishnani, et al., 2009b]. Additional research could explore parental attitudes in these countries so as to better inform new and expectant parents.

This study purposely did not compare the attitudes of families who have children with DS to matched families, so we cannot assume that their responses are similar to or different from “typical” families. To the extent that others are interested in knowing, for example, if parents of children with DS love their children more or regret them less compared to “typical” counterparts, additional research would need to be done. The singular aim of this research was to define the attitudes of parents who have children with DS, as a start.

In some cases, two or more parents/guardians from the same household responded with separate surveys. For this analysis, we chose to treat all parents/guardians equally, as we did not want to assume that parents/guardians would respond with similar answers. Additionally, we did not want to limit the families to just one response, so we chose to invite and include all respondents. In forthcoming analysis, we intend to do a full “family” analysis where we take a look at the data from those families with full responses: parents, siblings, and the self-advocate. This will provide an opportunity to explore variances within households.

Lastly, in as much as the attitudes of parents who have children with DS are captured, further attempts should be made to capture the attitudes of parents who chose not to continue their pregnancies after receiving a prenatal diagnosis of DS. Parallel questions could be asked—e.g., Did you think you would love a child with DS should you have continued your pregnancy? Did you think you would have regrets had you continued the pregnancy? Of course, such questions are highly sensitive, and only one such study has been performed, to date [Korenromp et al., 2007].

Implications

New research promises a definitive noninvasive prenatal test for DS [Chiu et al., 2011; Ehrich et al., 2011; Fan et al., 2008; Lo, 2009], and some have speculated that when that day arrives, nearly all women will learn of a DS diagnosis prenatally [Skotko, 2009]. Now, and increasingly so in the future, clinicians will need to provide expectant parents with accurate information about family life when a member has DS. Extensive training is needed, but, thus far, the education of healthcare professionals has placed scant focus on what to say about DS to new and expectant parents. When 2,500 medical school deans, students, and residency directors were surveyed in 2005, 81% of medical students reported that they “are not getting any clinical training regarding individuals with intellectual disabilities,” and 58% of medical school deans said such training is not a high priority [Special Olympics, 2005]. When 532 ACOG fellows and junior fellows were questioned in 2004, 45% rated their training regarding prenatal diagnosis as “barely adequate or nonexistent,” and only 28% felt “well qualified” in general prenatal genetic counseling [Cleary-Goldman et al., 2006]. When a separate group of 507 ACOG fellows and junior fellows were questioned some four years later, little had changed—approximately 40% thought their training was “less than

adequate,” and only 36% felt “well qualified” in counseling an expectant mother whose prenatal screen suggests a high chance for DS [Driscoll et al., 2009].

A consensus statement, written in 2009, from ACOG, ACMG, NDSGC, NDSS, and NDSC calls for four areas of collaborative change in anticipation of these forthcoming noninvasive prenatal diagnostic tests [American College of Obstetricians and Gynecologists et al., 2009]: (1) A “gold-standard” packet of information should be developed for all expectant parents who receive a prenatal diagnosis of DS. The booklet, “Understanding a Diagnosis of Down Syndrome Diagnosis” has now been created with assistance from all of the organizations and is available for dissemination from the organization, Lettercase, Inc. (www.lettercase.org). (2) Practice guidelines should be written on how best to deliver a prenatal diagnosis of DS. Since this recommendation, a 29-member Down Syndrome Diagnosis Study Group has now published an evidence-based review, which can serve as a blueprint for the academic societies as they develop their own practice guidelines [Skotko, Kishnani, et al., 2009]. (3) A public awareness campaign should be initiated to educate pre-pregnant couples about life with DS. Since this recommendation, the NDSS has started the “My Great Story” media campaign (www.ndss.org), and the NDSC has initiated the “More Alike than Different” campaign (www.ndscenter.org). (4) Quality training should be developed for healthcare professionals on how to deliver a prenatal diagnosis of DS. Addressing this urgency is a new online patient simulation, available for free, with evaluation already published in peer-reviewed journals (www.brighter-tomorrows.org) [Ferguson et al., 2006].

At the core of the recommendations in the consensus statement is the need for a more informed understanding of family life when a member has DS. This study provides further information that can be incorporated into informational booklets, public awareness campaigns, and professional trainings. Additionally, the study provides evidence-based information from our sampled population that can now be shared with expectant couples during prenatal counseling, whether done in the offices of obstetricians, family practitioners, geneticists, and genetic counselors or in one of the many DS clinics across the country (www.ndss.org):

- The overwhelming majority of parents who have children with DS report that they love their son or daughter and are proud of them.
- The overwhelming majority of parents who have children with DS report that their outlook on life is more positive because of their son or daughter with DS.
- Parents who have children with DS mention that while there are struggles and challenges, their children with DS bring them much joy and many rewards. They cite life lessons in acceptance, patience, and purpose.
- The overwhelming majority of parents who have children with DS say that their other children have good relationships with their brothers and sisters with DS.
- The majority of parents who have children with DS report that their other children are more caring and sensitive, as a result.
- A very small percentage of parents who have children with DS say that they are embarrassed by their son or daughter or even regret having them altogether. The majority of these parents had children with significant medical and learning challenges.
- Slightly more than half of parents who have children with DS say that they have found non-profit DS organizations helpful.

Delivering a diagnosis of DS will remain difficult for providers and parents alike, but clinicians now have even more tools to deliver such news in a more complete and accurate manner.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Table ICharacteristics of Parent Respondents ($N=1,989$).

Background Variables	%
Parent role ($N=1,955$)	
Mother	63
Father	37
Marriage status ($N=1,958$)	
Married	88
Divorced	4
Unmarried but with partner	3
Widowed	2
Single	2
Age of son/daughter with DS ($N=1,953$)	
< 5	33
5 and < 10	22
10 and <15	14
15 and <20	10
20 and <25	8
25 and <30	5
30 and <35	3
35 and <40	2
40	2
Gender of son/daughter with DS ($N=1,973$)	
Male	55
Female	45
Birth order of son/daughter with DS ($N=1,921$)	
Only	12
Youngest	52
Between youngest and oldest	16
Oldest	20
Biological parent ($N=1,969$)	
Yes	97
No	3
Spanish/Hispanic/Latino ($N=1,939$)	
Yes	14
No	86
Race ($N=1,886$)	
White	89
Black or African American	2
Asian	2
American Indian or Alaska Native	1
Other	5

Background Variables	%
Multiple	1
U.S. State (<i>N</i> = 1,966)	
Massachusetts	22
California	21
Colorado	20
Georgia	15
North Carolina	8
Texas	7
Other	7
Religious Affiliation (<i>N</i> = 1,859)	
Protestant	44
Catholic	35
Atheist	10
Jewish	4
Mormon	3
Other	3
Multiple	1
Educational Level (<i>N</i> = 1,940)	
Not graduated from 8 th grade	1
Graduated from 8 th grade	2
Graduated from high school	24
Graduated from college/university	47
Received a master's degree	20
Received a doctorate	6
Living Situation (<i>N</i> = 1,953)	
Lives with child with DS	93
Does not live with child with DS	7

Table II

Parental Feelings Toward Son or Daughter with Down Syndrome.

Statements	<i>N</i>	<i>M</i> [*]	<i>SD</i>	% Agree [†]
I am proud of my son or daughter with DS.	1,977	6.8	0.8	97
I love my son or daughter with DS.	1,979	6.9	0.6	99
In general, I am embarrassed by my son or daughter with DS.	1,958	1.5	1.2	5
Overall, I regret having my son or daughter with DS.	1,966	1.5	1.2	4

* Parents were asked to rate their level of agreement with the statements on a Likert scale of 1 to 7 with “1” indicating “strongly disagree,” “4” being “neutral,” and “7” indicating “strongly agree.”

[†] Percentage of parents who circled “5,” “6,” or “7” on Likert scale for that statement.

Table III

Parental Perception of the Impact their Son or Daughter with DS has on Family and Community.

Statements	<i>N</i>	<i>M</i> [*]	<i>SD</i>	% Agree [†]
I feel that my outlook on life is more positive because of my son or daughter with DS.	1,967	5.9	1.4	79
My child(ren) without DS have a good relationship with my son or daughter with DS.	1,715	6.4	1.0	95
On the whole, I believe that my other children are more caring and sensitive to others because of my son or daughter with DS.	1,668	6.0	1.3	84
I feel comfortable answering my children's questions about DS.	1,723	6.6	0.9	95
Right now, my son or daughter with DS is putting a strain on my marriage/partnership.	1,881	1.8	1.5	11
Right now, my children without DS are putting a strain on my marriage/partnership.	1,656	2.0	1.6	11
DS parent support groups are helpful to me.	1,867	4.9	1.6	55

* Parents were asked to rate their level of agreement with the statements on a Likert scale of 1 to 7 with "1" indicating "strongly disagree," "4" being "neutral," and "7" indicating "strongly agree."

† Percentage of parents who circled "5," "6," or "7" on Likert scale for that statement.

Table IV

Correlation Among Parents' Responses on Survey Statements.

	A	B	C	D	E	F	G	H	I	J	K
A	—										
B	0.67*	—									
C	-0.34*	-0.35*	—								
D	0.38*	0.29*	-0.24*	—							
E	-0.43*	-0.37*	0.37*	-0.35*	—						
F	0.11*	0.08*	-0.05 [‡]	0.15*	-0.06 [‡]	—					
G	0.41*	0.38*	-0.25*	0.30*	-0.28*	0.15*	—				
H	0.27*	0.26*	-0.13*	0.30*	-0.16*	0.13*	0.40*	—			
I	-0.25*	-0.15*	0.23*	-0.30*	0.28*	-0.06 [‡]	-0.27*	-0.15*	—		
J	-0.15*	-0.16*	0.17*	-0.17*	0.20*	-0.07 [‡]	-0.23*	-0.16*	0.56*	—	
K	0.33*	0.38*	-0.19*	0.24*	-0.18*	0.14*	0.27*	0.28*	-0.16*	-0.15*	—

The statements were as follows: A: I am proud of my son or daughter with DS; B: I love my son or daughter with DS; C: In general, I am embarrassed by my son or daughter with DS; D: I feel that my outlook on life is more positive because of my son or daughter with DS; E: Overall, I regret having my son or daughter with DS; F: DS parent support groups are helpful to me; G: My child(ren) without DS have a good relationship with my son or daughter with DS; H: On the whole, I believe that my other children are more caring and sensitive to others because of my son or daughter with DS; I: Right now, my son or daughter with DS is putting a strain on my marriage/partnership; J: Right now, my children without DS are putting a strain on my marriage/partnership; K: I feel comfortable answering my children's questions about DS.

* $p < 0.001$.
[‡] $p < 0.01$.
[‡] $p < 0.05$.

Table V

What life lessons have you learned from your son or daughter with DS? ($N = 997$).

Category	Total*	%
personal self-growth	435	48%
patience	318	35%
acceptance / respect	216	24%
love	215	24%
joy	115	13%
everyone has gifts / we're all more alike than different	107	12%
lessons on blessings / faith / God	99	11%
don't take anything for granted / appreciation	73	8%
kindness / empathy	70	8%
perseverance	62	7%
learning how to advocate	55	6%
learning how to set higher expectations for others	47	5%
learning how to be positive	46	5%
tolerance	45	5%

* Indicates number of parents who incorporated this category in their response to this question; percentages will not add to 100%, as responses might contain more than one category. Only categories with at least 5% response have been included.

Table VI

If a couple were expecting to have a child with DS, what would you like to tell them? ($N=997$).

Category	Total*	%
you will experience joy/rewards	392	39%
there will be struggles/challenges	319	32%
you will experience love	261	26%
important to identify good support group/resources	246	25%
children with DS are more alike than different from typically developing children	245	25%
life goes on / things will be okay	244	24%
you will experience personal growth / perspective	193	19%
you will learn acceptance	155	16%
comment about blessing/ spirituality / faith / God	141	14%
you will learn how to advocate / teach	85	9%
children with DS have a good quality or life	84	8%
keep expectations high	75	8%
congratulations / you are lucky to have this child	58	6%
life will change / never be the same	50	5%
be patient	48	5%
be positive	46	5%

* Indicates number of parents who incorporated this category in their response to this question; percentages will not add to 100%, as responses might contain more than one category. Only categories with at least 5% response rate have been included.