RESEARCH ARTICLE

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Family Perspectives About Down Syndrome

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National medical organizations recommend that during prenatal counseling sessions, healthcare providers discuss how having a child with Down syndrome (DS) might impact the family unit. Few studies, to date, have surveyed families about their life experiences. For this investigation, we examined 41 family attitudes, which were obtained from mailed questionnaires completed by 1,961 parents/guardians, 761 brothers/sisters, and 283 people with DS who were members of six DS non-profit organizations, chosen for their size, ethnic/racial diversities, and geographic distribution throughout the United States. About 83% of families reported to all being proud of the family member with DS, and 87% reported to all feeling love for the member with DS. Younger siblings (ages 9–11) were more likely to feel embarrassed by their sibling with DS if their parents/guardians also did. If one or more parents/guardians felt that their children without DS *did* have a good relationship with their child with DS, siblings were more likely to report that they loved and liked their brother/sister with DS. Overall, our data demonstrate that positive themes tend to dominate within modern-day families who have members with DS, although challenges were not insignificant for some. © 2015 Wiley Periodicals, Inc.

Key words: trisomy 21; prenatal tests; noninvasive prenatal screens; abortion; family attitudes; quality of life

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INTRODUCTION

As the popularity and availability of noninvasive prenatal screening (NIPS) expands, more expectant couples will contemplate pregnancy options after receiving a "positive" result for Down syndrome (DS). The American College of Medical Genetics and Genomics (ACMG) endorses "accurate, up-to-date, and balanced information about DS" as crucial to informed decision-making [Gregg et al., 2013]. The National Society of Genetic Counselors (NSGC) recommends that healthcare providers "specifically discuss how having a child with Down syndrome might affect the family as a unit, the parents' relationship as a couple, and other present or future children" [Sheets et al., 2011]. Meeting this charge, medical organizations collaborated with national DS organizations and The Joseph P. Kennedy Jr. Foundation to create *Understanding a Down Syndrome Diagnosis*, a booklet reviewing the evidence about DS intended for expectant couples who have received a prenatal diagnosis of DS but have not yet made a decision regarding their pregnancy options [Down Syndrome Consensus Group, 2008].

Brian G. Skotko and Susan P. Levine are co-first authors.

Conflict of interest: Brian Skotko serves in a non-paid capacity on the Medical and Science Advisory Board for the Massachusetts Down Syndrome Congress and the Board of Directors for Band of Angels Foundation, both non-profit organizations. He also serves on the Professional Advisory Committee for the National Center for Prenatal and Postnatal Down Syndrome Resources. Dr. Skotko occasionally gets remunerated from Down syndrome non-profit organizations for speaking engagements about Down syndrome. He has a sister with Down syndrome. He receives research support for clinical drug trials involving patients with Down syndrome from Hoffmann-La Roche, Inc. Sue Levine occasionally gets remunerated from Down syndrome non-profit organizations for speaking engagements about Down syndrome.

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Abbreviations: DS, Down syndrome; NIPS, noninvasive prenatal screening.

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While decades of studies have catalogued medical conditions that often accompany DS, few have surveyed families about their life experiences. Previously, we reported analyses of 3,056 parents, siblings (ages 9 and older), and people with DS (ages 12 and older) from across the United States [Skotko et al., 2011a,b,c]. From these questionnaires, a family portrait emerged. Nearly, all parents reported loving (99%) and being proud (97%) of their child with DS. About 79% said that their outlook on life was more positive because of their son/daughter with DS. Brothers and sisters also had a favorable perspective, with nearly all reporting love (97%) and pride (94%). Older siblings reported being a better person (88%) because of their brother/sister with DS. Yet, approximately 5% of parents reported general embarrassment, 4% regretted having their son or daughter with DS, and nearly 11% reported marital strain attributable to the child. Among siblings, 7% were embarrassed, and about 4% wished they could trade their sibling with DS in for a typically developing counterpart.

People with DS, themselves, were positive. Approximately 99% reported to be happy with their lives; 97% liked who they are; and 96% liked how they look. About 87% said that they could make friends easily, and only 4% felt sad about their lives, in general. Overall, mothers, fathers, brothers, sisters, and people with DS shared that they were satisfied—and even positive—about their lives, despite acknowledging the challenges that sometimes accompany DS.

Until now, these data have not been examined at the family level as an interactive model. If a mother or father felt embarrassed by their son or daughter with DS, did siblings feel similarly? If one person thought their outlook on life was distinctly more positive because of the person with DS, did the other family members share in that positive assessment? If a person with DS was sad about his or her life, what was the perception of his/her other family members? Here, we examine relationships among attitudes within families of a person with DS.

MATERIALS AND METHODS

Participants

Survey respondents included parents/guardians, brothers and sisters (ages 9 and older), and people with Down syndrome (ages 12 and older) who were members of six DS non-profit organizations, chosen for their size, ethnic/racial diversities, and geographic distribution throughout the United States (California, Colorado, Georgia, Massachusetts, North Carolina, and Texas). Responses from 1,961 parents/guardians, 761 brothers/sisters, and 283 people with Down syndrome were analyzed. The research was approved as protocol H-26552 by the Institutional Review Board of Boston University Medical Center, where the authors were based at the time the surveys were conducted.

Survey Instruments

Separate questionnaires were developed for (i) parent/guardians; (ii) brothers/sisters, ages 9–11; (iii) brothers/sisters, ages 12 and older; and (iv) people with DS, ages 12 and older. (See online Supplementary Materials accompanying the original references.) The questions measured attitudes and perceptions about family life, gathering quantitative information with Likert statements and qualitative information using open-ended questions. Parents were asked to rate their child's functional abilities, health condition, and educational challenges. An age-adjusted composite functional activity score was calculated by summing assessments of 11 functional abilities (e.g., walking, preparing meals, going on dates), each scored 1 (no ability or not applicable) to 7 (performed very well), and dividing by the age in years of the person with DS. Participants were asked to report optional sociodemographic information. The focus group piloting, validation and reliability testing, and final survey distribution have been previously described [Skotko et al., 2011a,b,c].

Data Analyses

Forty-one family attitudes were considered (Table II). Algorithmic definitions of each attitude as calculated from the survey questions are available in an online supplement. A family unit is defined as all survey respondents who identified as being from the same family (surveys mailed to families were coded with unique identifiers). Some attitudes required data only from parents, some from both the parents and the family member with DS, and others responses from at least one sibling. The variable context of each attitude identified in Table II is specified in its description and led to variable sample sizes of families or family members endorsing a given attitude. The proportions of parents, siblings, individuals with DS, or families reporting a given attitude were calculated as simple ratios of the number endorsing the attitude among those for which data were available with exact Clopper–Pearson confidence intervals. Associations between attitudes of parents and siblings or parents and family members with DS were tested by Fisher's exact test for nominal variables and exact Cochran–Armitage trend tests for ordinal variables. Associations between attitudes and demographic characteristics of families were tested by logistic regression. Composite functional activity scores were log-transformed to stabilize variance. Stepwise logistic regression was used to select final models with thresholds of $P < 0.05$ for a term to enter the model and $P > 0.10$ to remove a term. The sample used for analysis was adjusted as terms were added or removed rather than using complete-case data over all possible predictors given the variable distribution of missing data.

RESULTS

Characteristics of Family Units

The majority of family units identified as White, with about 16% having one or more members identifying as Spanish/Hispanic/Latino (Table I). Most respondents were from California, Colorado, Georgia, and Massachusetts. The majority of family units were Catholic or Protestant, consisting of two married parents living together with a high school degree or higher. There were slightly more males with DS represented. Family members with DS were at varying stages in their education, had varying numbers of siblings, and had varying birth orders. The majority of people with DS were living with their parents. Most family units had a parent who did not feel their son/daughter with DS had health problems; although about 44% had a parent who felt learning problems were

TABLE I. Characteristics of Representatives From Family Units (N = 1,418)

Background variables	**%**
Race of all parent respondents within family (N = 1,279)	
White	86.3
Black or African American	2.5
Asian	1.8
American Indian and/or Alaska Native	0.5
Native Hawaiian and/or other Pacific Islander	0.2
Other	4.1
Mixed between parents/multiracial per parent	4.7
Race of all sibling respondents within family (N = 594)	
White	84.7
Black or African American	2.5
Asian	0.8
Other	6.9
Mixed between siblings/multiracial per sibling	5.1
Race of person with DS, self-report (N = 274)	
White	88.0
Black or African American	2.2
Asian	2.2
Other	5.5
Multiracial per person	2.2
Race of the all respondents within family (N = 1,372)	
White	83.9
Black or African American	2.4
Asian	1.5
American Indian and/or Alaska Native	0.2
Native Hawaiian and/or other Pacific Islander	0.1
Other	4.1
Mixed among family members/multiracial per person	7.7
Ethnicity of all parent respondents within family (N = 1,314)	
Spanish/Hispanic/Latino	11.8
Not Spanish/Hispanic/Latino	84.6
Mixed between parents	3.6
Ethnicity of all sibling respondents within family (N = 605)	
Spanish/Hispanic/Latino	13.9
Not Spanish/Hispanic/Latino	85.1
Mixed among siblings	1.0
Ethnicity of person with DS, self-report (N = 267)	
Spanish/Hispanic/Latino	10.5
Not Spanish/Hispanic/Latino	89.5
Ethnicity of all respondents within family (N = 1,407)	
Not Spanish/Hispanic/Latino	83.9
Other/mixed among family members	16.1
U.S. state where parents live (N = 1,325)	
CA	22.0
CO	19.5
GA	14.1
MA	23.3
NC	7.6
TX	6.5
Other, including mixed between parents	6.9
U.S. state where siblings live (N = 611)	
CA	20.9
CO	21.8
GA	12.3
MA	22.1
NC	7.7
TX	3.9
Other, including mixed among siblings	11.3
U.S. state where person with DS lives (N = 276)	
CA	29.3
CO	21.0
GA	8.0
MA	25.4

(*Continued*)

TABLE I. *(Continued)*

Background variables	%
NC	5.8
TX	4.0
Other	6.5
U.S. state where all respondents within family live (N = 1,412)	
CA	21.5
CO	19.3
GA	13.5
MA	23.0
NC	7.2
TX	6.2
Other/mixed among family members	9.2
Religion of all parents within family (N = 1,275)	
Catholic	32.8
Protestant	41.2
Mormon	2.4
Jewish	3.1
Atheist/none	7.3
Other, including mixed among family members	2.8
Multiple per parent/mixed between parents	10.4
Religion of all sibling respondents within family (N = 583)	
Catholic	34.1
Protestant	43.1
Mormon	2.7
Jewish	3.1
Atheist/none	12.2
Other	2.4
Multiple per sibling/mixed among siblings	2.4
Religion of person with DS, self-report (N = 263)	
Catholic	39.9
Protestant	41.1
Mormon	1.9
Jewish	5.7
Atheist/none	5.7
Other	5.7
Religion of all respondents within family (N = 1,374)	
Catholic	30.3
Protestant	39.0
Mormon	2.4
Jewish	2.8
Atheist/none	6.8
Other	2.1
Multiple per member/mixed among family members	16.5
Parental living situation (N = 1,321)	
Live together	92.3
Live apart	7.0
Discordant responses between parents	0.8
Parental marital status (N = 1,315)	
Married	84.9
Single, divorced, widowed, or unmarried with partner	15.1
Sex of person with DS, as reported by parents (N = 1,321)	
Female	44.7
Male	55.3
Sex of person with DS, by sibling report (N = 615)	
Female	42.4
Male	57.6
Sex of person with DS, self-report (N = 266)	
Female	45.9
Male	54.1
Sex of person with DS as reported by all family members (N = 1,403)	
Female	44.5
Male	55.5
Number of children, as reported by parents (N = 1,329)	
1	12.1
2	32.7
3	32.1

(*Continued*)

TABLE I. (*Continued*)

Background variables	**%**
4	12.9
5	4.1
>5	3.7
Discordant responses between parents	2.4
Birth order of persons with DS, as reported by parents (N = 1,299)	
1	31.8
2	32.2
3	19.9
4	8.3
5	2.5
>5	2.3
Discordant responses between parents	3.0
Number of parents, reported by person with DS (N = 273)	
0	0.7
1	5.9
2	87.2
3	5.1
4	1.1
Number of parents, by sibling report (N = 617)	
1	3.1
2	83.3
3	6.6
4	4.1
Discordant responses among siblings	2.9
Number of siblings, reported by person with DS (N = 254)	
0	6.3
1	33.1
2	35.8
3	13.4
>3	11.4
Health problems for person with DS (minimum parental score) (N = 1,308)	
1 (Not a problem)	36.9
2	22.9
3	9.0
4 (Neutral)	15.6
5	7.3
6	4.4
7 (Very much a problem)	3.9
Learning problems for person with DS (minimum parental score) (N = 1,300)	
1 (Not a problem)	6.5
2	8.7
3	10.9
4 (Neutral)	28.8
5	19.8
6	16.1
7 (Very much a problem)	9.1
Educational level of parent (minimum, if >1 family respondent) (N = 1,316)	
<8th grade	1.8
8–11th grade	2.0
High school graduate	29.8
College graduate	47.8
Master's degree	15.4
Doctorate degree	3.2
Educational level of parent (maximum, if >1 family respondent) (N = 1,316)	
<8th grade	0.8
8–11th grade	1.6
High school graduate	19.5
College graduate	47.2
Master's degree	23.6
Doctorate degree	7.3
Educational level of sibling (minimum, if >1 family respondent) (N = 490)	
<8th grade	25.3
8–11th grade	29.2
High school graduate	23.7
College graduate	16.1

(*Continued*)

TABLE I. (*Continued*)

Background variables	**%**
Master's degree	4.7
Doctorate degree	1.0
Educational level of sibling (maximum, if >1 family respondent) (N = 490)	
<8th grade	23.3
8–11th grade	29.2
High school graduate	24.7
College graduate	16.1
Master's degree	5.3
Doctorate degree	1.4
Educational level of person with DS, self-report (N = 268)	
1st–8th grade	16.8
In high school	27.6
High school graduate	47.4
In college	6.0
College graduate	2.2
Working situation for person with DS, self-report (N = 268)	
In school	50.4
Out of school and working	35.8
Out of school and not working	13.8
Living situation for persons with DS, self-report (N = 277)	
Living with one or both parents/guardians	84.1
Living by myself in apartment or home	5.8
Living with roommates in apartment or home	4.7
Living in a group home	2.5
Living in another place	2.9
Living situation of all sibling respondents within family (N = 607)	
Live with sibling with DS	74.1
Do not live with sibling with DS	24.2
Discordant report among siblings	1.6
Living situation of all respondents in family (N = 1,411)	
Living together	84.1
Other, including discordant report among family members	15.9
Biological status of parent respondent (N = 1,324)	
Biological parent	96.1
Not biological parent	1.6
Discordant report between parents	2.3
Sibling with DS a step- or half-sibling? (per sibling report) (N = 608)	
Yes	11.7
No	86.0
Discordant report among siblings	2.3
Biological status of person with DS as reported by all family members (N = 1,398)	
Biological family member	91.1
Other, including discordant report among family members	8.9

a concern. Mean composite functional activity scores were 33.5 (SD = 13.3, median = 31.5) before age adjustment and 6.4/year after age adjustment, with wide variance (SD = 14.2/year, median = 3.8/year). The majority of parent respondents were biologically related to the member with DS; about 12% of brothers and sisters included step- or half-siblings.

Shared Perspectives and Attitudes

Of the 172 family units that had at least one parent, one sibling, and one person with DS responding, 83% reported to all being proud of the family member with DS (Table II). This was more likely in families in which siblings were better educated ($P = 0.001$) (Table III). Approximately, 87% of family units had all respondents expressing love for the member with DS (Table II). This was more likely if the person with DS was biologically related to the other family members ($P = 0.020$) (Table III). The functional skills of the person with DS, as reported by their parents, were not associated with any of the examined attitudes in the final multiple regressions.

Younger siblings (ages 9–11) were more likely to feel embarrassed by their sibling with DS if their parents/guardians also did ($P = 0.04$) (Table II). This was particularly true if the child with DS was older ($P = 0.002$) (Table III), but less so in families with White parents ($P = 0.024$) (Table III). If all of the parents/guardians in a family unit expressed regret in having the child with DS, older siblings were more likely to want to trade their sibling with DS for one that did not have DS ($P = 0.03$) (Table II). This was more likely among siblings with less education ($P = 0.088$) and if the sibling with DS was born earlier in the birth order ($P = 0.008$) (Table III). Again, the functional skills of the person with DS, as reported by their parents, were not associated with any of the examined attitudes in the final multiple regressions.

TABLE II. Shared Perspectives and Attitudes Among Members of the Same Family

Family attitude	N	% [95%CI]	Family attitude	N	%	*P*
Of the families that had at least one parent, one sibling, and one person with DS responding, what % *all* felt **proud**?	143/172	83.1 [76.7, 88.4]				
Of the families that had at least one parent, one sibling, and one person with DS responding, what % *all* felt **love**?	150/172	87.2 [81.3, 91.8]				
If one or more parents/guardians responded, and all *did* feel **embarrassed** by child with DS, what . . .	33/1,331	2.5 [1.7, 3.5]	If one or more parents/guardians responded, and all did *not* feel **embarrassed** by child with DS, what . . .	1,298/1,331		
% of at least one younger siblings also feels embarrassed?	0/1	0.0	% of at least one younger siblings also feels embarrassed?	16/186	8.6	ns
% of at least one older siblings also feels embarrassed?	1/10	10.0	% of at least one older siblings also feels embarrassed?	36/422	8.5	ns
% of people with DS report being sad about life?	0/6	0.0	% of people with DS report being sad about life?	9/256	3.5	ns
If one or more parents/guardians responded, and all did *not* feel **embarrassed** by child with DS, what . . .	1,203	90.4 [88.7, 91.9]	If one or more parents/guardians responded, and all *did* feel **embarrassed** by child with DS, what . . .	128		
% of at least one younger sibling also feels embarrassed?	12	7.1	% of at least one younger sibling also feels embarrassed?	4	23.5	0.04
% of at least one older siblings also feels embarrassed?	29	7.7	% of at least one older siblings also feels embarrassed?	8	15.1	ns
% of people with DS report being sad about life?	9	3.9	% of people with DS report being sad about life?	0	0.0	ns
If one or more parents/guardians responded, and all expressed **regret** having child with DS, what . . .	28	2.1 [1.4, 3.0]	If one or more parents/guardians responded, and all *did not* express **regret** having child with DS, what . . .	1,303		
% of at least one younger siblings also wished they could trade in their sibling with DS for one that did not have DS?	0	0.0	% of at least one younger siblings also wished they could trade in their sibling with DS for one that did not have DS?	8	4.3	ns
% of at least one older siblings often wished they could trade in their sibling with DS for one that did not have DS?	2	22.2	% of at least one older siblings often wished they could trade in their sibling with DS for one that did not have DS?	17	4.0	ns
% of people with DS reported that they love their family?	0	0.0	% of people with DS reported that they love their family?	2	0.8	ns
If one or more parents/guardians responded, and all expressed that they did *not* have **regret** having child with DS, what . . .	1,179	88.6 [86.7, 90.2]	If one or more parents/guardians responded, and all expressed that they *did* have **regret** having child with DS, what	152	11.4	
% of at least one younger siblings said they would *not* trade in their sibling with DS for one that did not have DS?	162	96.4	% of at least one younger siblings said they would *not* trade in their sibling with DS for one that did not have DS?	17	89.5	ns
% of at least one older siblings *did not* wish they could trade in their sibling with DS for one that did not have DS?	363	96.5	% of at least one older siblings *did not* wish they could trade in their sibling with DS for one that did not have DS?	50	89.3	0.03
% of people with DS reported that they love their family?	224	99.1	% of people with DS reported that they love their family?	36	100.0	ns

(*Continued*)

TABLE II. (*Continued*)

Family attitude	N	% [95%CI]	Family attitude	N	%	*P*
If one or more parents/guardians all responded that they felt their **outlook on life** was more positive because of child with DS, what . . .	959	72.1 [69.6, 74.4]	If one or more parents/guardians *did not* respond that they felt their **outlook on life** was more positive because of child with DS, what . . .	372	27.9	
% of at least one older siblings felt they were better people because of their sibling with DS?	281	88.1	% of at least one older siblings felt they were better people because of their sibling with DS?	97	85.8	ns
% of people with DS said that they feel that they help other people?	153	88.1	% of people with DS said that they feel that they help other people?	64	87.7	ns
If one or more parents/guardians all responded that they felt their **outlook on life** was *not* more positive because of child with DS, what . . .	45	3.4 [2.5, 4.5]	If one or more parents/guardians responded that they felt their **outlook on life** *was* more positive because of child with DS, what . . .	1,286		
% of at least one older siblings felt they were better people because of their sibling with DS?	12	92.3	% of at least one older siblings felt they were better people because of their sibling with DS?	366	87.4	ns
% of people with DS said that they feel that they help other people?	10	100.0	% of people with DS said that they feel that they help other people?	207	82.1	ns
If one or more parents/guardians all responded that they felt their children without DS had a **good relationship** with their child with DS, then what . . .	1,068	80.2 [78.0, 82.3]	If one or more parents/guardians responded that they felt their children without DS *did not* have a **good relationship** with their child with DS, then what . . .	263		
% of at all siblings responded that they love their sibling with DS?	489	96.6	% of at all siblings responded that they love their sibling with DS?	31	77.5	<0.001
% of people with DS said that they like their siblings with DS?	211	96.3	% of people with DS said that they like their siblings with DS?	19	44.2	<0.001
% of people with DS said that they feel their sibling with a good friend?	193	88.1	% of people with DS said that they feel their sibling with a good friend?	17	39.5	<0.001
% of all of the above were true?	130	82.8	% of all of the above were true?	9	60.0	0.04
If one or more parents/guardians all responded that they felt their children without DS did *not* have a **good relationship** with their child with DS, then what . . .	22	1.7 [1.0, 2.5]	If one or more parents/guardians all responded that they felt their children without DS did have a **good relationship** with their child with DS, then what . . .	1,309		
% of at all siblings responded that they love their sibling with DS?	7	63.6	% of at all siblings responded that they love their sibling with DS?	513	95.9	0.001
% of people with DS said that they like their siblings with DS?	3	50.0	% of people with DS said that they like their siblings with DS?	227	88.7	0.03
% of people with DS said that they feel their sibling with a good friend?	3	50.0	% of people with DS said that they feel their sibling with a good friend?	207	80.9	ns
% of all of the above were true?	1	33.3	% of all of the above were true?	138	81.7	ns
If one or more parents/guardians all responded that they felt **comfortable answering** their children's questions about DS, what . . .	1,090	81.9 [79.7, 83.9]	If one or more parents/guardians responded that they *did not* feel **comfortable answering** their children's questions about DS, what . . .	241		
% of at all younger siblings said that they would ask their parents if they had a question about DS?	155	84.7	% of at all younger siblings said that they would ask their parents if they had a question about DS?	3	75.0	ns
% of all older siblings said that they do feel comfortable asking their parents questions about DS?	350	86.2	% of all older siblings said that they do feel comfortable asking their parents questions about DS?	21	80.8	ns

(*Continued*)

TABLE II. *(Continued)*

Family attitude	N	% [95%CI]	Family attitude	N	%	*P*
If one or more parents/guardians all responded that they did *not* feel **comfortable answering** their children's questions about DS, what . . .	16	1.2 [0.7, 1.9]	If one or more parents/guardians all responded that they *did* feel **comfortable answering** their children's questions about DS, what . . .	1,315	98.8	
% of all younger siblings said that they would ask their parents if they had a question about DS?	0/0	ne	% of all younger siblings said that they would ask their parents if they had a question about DS?	158/187	84.5	ne
% of all older siblings said that they do feel comfortable asking their parents questions about DS?	6	85.7	% of all older siblings said that they do feel comfortable asking their parents questions about DS?	365	85.9	ns
If at least one younger and one older sibling felt that their parents *did* pay **too much attention** to their sibling with DS, what	566	91.0 [88.5, 93.1]	If at least one younger and one older sibling felt that their parents did *not* pay **too much attention** to their sibling with DS, what	622		
% of people with DS felt the opposite—that is, that more attention was being given to their sibling without DS?	24	13.9	% of people with DS felt the opposite—that is, that more attention was being given to their sibling without DS?	27	14.8	ns

ns, not significant; ne, not estimable.

Associations relative to sibling relationships were also noted. When parents/guardians felt that their children without DS *did not* have a good relationship with their child with DS, siblings were less likely to express love ($P=0.001$) and affection ($P=0.03$) toward their brother/sister with DS. Conversely, if one or more parents/guardians felt that their children without DS *did* have a good relationship with their child with DS, siblings were more likely to report that they loved ($P<0.001$) and liked ($P<0.001$) their brother/sister with DS (Table II). This was more likely the case when the person with DS was biologically related ($P=0.01$) and the parents were older at the time of giving birth to the family member with DS ($P=0.006$) (Table III). Similarly, their sibling with DS was more likely to view their brothers/sisters as good friends ($P<0.001$) (Table II). This feeling of friendship was associated more often if the person with DS was older ($P=0.005$) and less often when the parents had a higher education ($P=0.019$) (Table III). No statistical differences existed for our other measures of family attitudes (Table II), and the functional skills of the person with DS, as reported by their parents, were not associated with any of the examined attitudes in the final multiple regressions.

DISCUSSION

When a person with DS becomes part of a family, parents and siblings often describe significant and lifelong changes. While positive experiences more frequently triumph, the modern-day American family that has a member with DS is not immune to challenges and tough moments. For this investigation, we analyzed the relationship of attitudes and emotions among different members of the same family.

The overwhelming majority of families reported unanimous feelings of love for their member with DS, particularly when the person with DS was biologically related to the other family members. Such love, of course, could take on different meanings for different family members: loyalty, sympathy, lack of regret, compassion, and agape, among others. These emotions have also been well chronicled in popular books written by family members [Berube, 1996; Beck, 1999; Skotko and Kidder, 2001; Soper, 2007; Groneberg, 2008; Soper, 2009]. Conversely, love did not resonate uniformly within all families, and parental feelings were not always similar to those of the siblings without DS. The overwhelming majority of families also expressed pride for the person with DS. Inconsistencies were more often noted when the sibling was younger. In our survey of siblings, ages 9–11, approximately 13% did not feel proud of their brother or sister with DS [Skotko et al., 2011a]. Perhaps, younger children are more likely to be jealous of the attention afforded to their sibling with DS or unable to separate the challenges of their sibling from their own, consistent with Erik Erikson's theory of personality development when school-aged children compare their self-worth to others [Erikson, 1959]. Their responses also reflect national data of neurotypically developing children that has found approximately 16% of neurotypically developing children between the ages of 6–11 felt "worthless or inferior" [National Center of Health Statistics at the Centers for Disease Control, 2007].

We note, however, that feelings of pride seem to evolve as a sibling matures. Only 6% of siblings older than 12 did not report

TABLE III. Characteristics Associated With Family Attitudes

			Odds ratio 95%CI		
Attitude	**Variable**	**Odds ratio**	**Lower**	**Upper**	***P***
Families that had at least one parent, one sibling, and one person with DS responding ALL feeling **proud**.	Sibling education (mean among siblings $\geq$12 years on a scale where 0 = less than 8th grade to 5 = doctorate)	2.00	1.32	3.03	0.001
Of the families that had at least one parent, one sibling, and one person with DS responding ALL feeling **love**.	Biological status of person with DS (biological vs. adopted or discordant responses)	4.81	1.27	18.13	0.020
When one or more parents/guardians responded, and all did *not* feel **embarrassed** by child with DS and younger siblings did feel embarrassed.	Age of person with DS, parental report (per year)	1.23	1.08	1.40	0.002
	Parental race (White vs. non-White)	0.16	0.03	0.79	0.024
When one or more parents/guardians responded, and all expressed that they did *not* have **regret** having child with DS and older siblings wished they could trade their sibling with DS for another sibling.	Sibling education (mean among siblings $\geq$12 years on a scale where 0 = less than 8th grade to 5 = doctorate)	0.60	0.34	1.08	0.088
	DS birth order, per parents (count of live births)	0.36	0.17	0.77	0.008
When one or more parents/guardians all responded that they felt their children without DS had a **good relationship** with their child with DS and all siblings reported that they love their sibling with DS.	Biological status of person with DS (biological vs. adopted)	5.52	1.52	20.07	0.010
	Parental age at birth of child with DS (per year)	1.14	1.04	1.26	0.006
When one or more parents/guardians all responded that they felt their children without DS had a **good relationship** with their child with DS and all siblings reported that they like their sibling with DS.	(No variables significant)				
When one or more parents/guardians all responded that they felt their children without DS had a **good relationship** with their child with DS and all siblings reported that they feel their sibling with DS is a friend.	Age of person with DS, per parents (per year)	1.14	1.04	1.25	0.005
	Parental education (mean among parents on scale where 0 = less than 8th grade to 5 = doctorate)	0.49	0.27	0.89	0.019
When one or more parents/guardians all responded that they felt their children without DS did *not* have a **good relationship** with their child with DS and all siblings reported that they love their sibling with DS.	(No variables significant)				
When one or more parents/guardians all responded that they felt their children without DS did *not* have a **good relationship** with their child with DS and all siblings reported that they like their sibling with DS.	(No variables significant)				

being proud in our surveys [Skotko et al., 2011a]. Relatedly, younger siblings were also more likely to feel embarrassed of their brother and sister with DS if their parents felt the same. Oftentimes, younger siblings and parents spend more time with their family member with DS than older siblings, who may live elsewhere or be involved in different social environments, removed from the occasional moments of social awkwardness. Older siblings might also be able to appreciate all of the benefits of their sibling with DS without the responsibilities that their parents have. Additionally or alternatively, the younger siblings' feelings of embarrassment might be developmentally typical, since school-aged children recognize, for the first time, disparities in personal development relative to other children at this age [Erikson, 1959]. Adolescent and adult siblings, with more securely established identities, may be more likely to value the intimacy of family relationships, ultimately settling into lives where they know what is intrinsically important to them.

While all families have their challenges, the attitudes of those having the greatest difficulties are worth focusing on. For example, some older siblings more likely wished they could trade in their sibling with DS for another brother or sister without DS. This was felt when their parents were more likely to have expressed regret in their decision to have their son/daughter with DS. This seemed to occur in families if the person with DS was born among the first few children or if the sibling had a lower educational level. However, the functional skills of the people with DS, as rated by their parents, were not associated with this attitude, suggesting that level of cognitive disability does not have as much of an impact as other family variables. Perhaps, when the person with DS is the oldest (or among the older), the parents/caregivers and siblings are investing more time in that person's care and are all more sensitive to challenges. If a sibling advances in his or her education, however, he or she might begin to live outside of the household, becoming more removed from the frustrations being experienced by these parents. Healthcare professionals should be aware that some parents do struggle with their emotions, even beyond the initial year after their child with DS is born. Our previous research demonstrated that parents who expressed regret were more likely to have children with DS who had greater perceived health problems, learning difficulties, or were of Jewish background [Skotko et al., 2011b].

In general, parents had a keen understanding of the relationship between their son/daughter with DS and their siblings. As they grew older, people with DS were more likely to consider their sibling a friend, suggesting, perhaps, that tensions are healed and bonds strengthened with maturity. At the same time, this can be part of normal development, where family and relationships take on a different level of meaning as a person enters early adulthood. Parents who had advanced degrees, however, were less likely to feel that their children with and without DS had a genuine friendship. This might be because their expectations were higher, their conceptualization of friendship different, or both.

Regardless of whether parents felt that their outlook on life was more positive because of their child with DS, older siblings reported, similarly, that they were better people because of the brother/sister with DS. This could suggest that siblings are shaped differently by the experience of having a family member with DS. They might have slightly different values, expectations, and understandings of strength that can occur growing up beside a person with DS. Previous studies have demonstrated that important life lessons occur between siblings at formidable times during development [Skotko and Levine, 2006; Skotko et al., 2011a]. Additionally, whether or not parents felt comfortable answering their children's questions about DS, siblings felt comfortable asking them, regardless of age. With that said, not all parents were comfortable explaining, and not all siblings were comfortable asking, suggesting that supports and resources might be helpful to open communication channels for some families.

This study is subject to the same limitations as previously described for this dataset, including selection bias and non-response bias [Skotko et al., 2011a,b,c]. Our data are also not drawn from a population-based sample, so generalizability to all families who have members with DS in the United States is limited beyond those family characteristics described in this paper. All of our respondents were from families whose parents received a postnatal diagnosis or chose to continue their pregnancies after a receiving a prenatal diagnosis. To the extent that our data are used in prenatal counseling sessions, genetic professionals should advise expectant couples of the composition of our informants. There are no population-based registries for DS in the United States, although efforts are underway to build a contact registry (https://dsconnect.nih.gov/), patient database [Lavigne et al., 2015], and biobank. Also, as people with DS have differing opportunities and supports in other countries, future research should explore cross-cultural comparisons to our U.S. families. We did not survey a sample of "typical" families, so conclusions cannot be drawn on how families who have a member with DS are similar to or different than "typical" families. Further research might explore the degree to which these same issues are evident in a comparison sample with neurotypical children or children with other diagnoses. Such questions are not currently included in national household surveys such as the National Survey of Children's Health [National Center of Health Statistics at the Centers for Disease Control, 2007].

Families who have children with DS are not alone, either in number or experience. In this largest study to date, mothers, fathers, brothers, sisters, and people with DS have shared their nuanced feelings and varying perspectives. As future expectant couples seek more information about DS, our data demonstrate that positive themes tend to dominate for modern day families who have members with DS, although challenges were not insignificant for some. Healthcare professionals now have a current portrait of these families, which can inform their counseling to new and expectant couples. Oftentimes, expectant couples also seek out parents who already have a child with DS [Massachusetts Down Syndrome Congress, 2014]. Our data suggest, however, that people with DS are not all alike, and neither are their families. Expectant couples—and the health care professionals and advocacy organizations that support them—should seek to involve a range of parents so that a fuller conceptualization of DS will emerge. The same extra chromosome does not script a common playbook for family relationships.

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