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Parental perspectives on research for Down syndrome

Amanda Nicole White[1,2] | Marly Chevette[3] | Hampus Hillerstrom[3] | Anna Esbensen[4,5]

[1]Graduate School of Leadership and Change, Antioch University, Yellow Springs, Ohio, USA

[2]Cincinnati Children's Research Foundation, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio, USA

[3]LuMind IDSC Foundation, Burlington, Massachusetts, USA

[4]Department of Pediatrics, University of Cincinnati College of Medicine, Cincinnati, Ohio, USA

[5]Division of Developmental and Behavioral Pediatrics, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio, USA

Correspondence
Amanda Nicole White, Cincinnati Children's Hospital Medical Center, 3333 Burnet Avenue, MLC 3016, Cincinnati, OH 45229, USA.
Email: nwhite2@antioch.edu

Abstract

Background: Down syndrome is the most common genetic disorder associated with intellectual and developmental disabilities. Research to improve health care outcomes in Down syndrome lags significantly behind other disease categories. Among these reasons are funding, recruitment and availability of research studies being conducted.

Methods: We surveyed 228 parents of individuals with Down syndrome to understand their perceptions of research, study design, how they seek out information and topics they would like to see researched.

Results: Parents with children 18 years and younger responded to our survey. Parents indicated their willingness to participate in research (72%), yet few have (36%). Parents identified barriers to participation, research they feel would help their child, and interests in seeing new therapies and drug studies.

Conclusion: These findings identify recommendations and insights from parents on future research agendas, studies and recruitment strategies that may help researchers improve outcomes for individuals with Down syndrome.

KEYWORDS
barriers, Down syndrome, parents, research participation

1 | INTRODUCTION

Down syndrome is one of the most common genetic conditions diagnosed prenatally or postnatally, with an estimated 1 in 5200 children born annually in the United States (de Graaf et al., 2015). Despite being the most common genetic syndrome contributing to intellectual disability, research on Down syndrome is under-funded compared to other disease categories. According to the National Institute of Health's (NIH) Research and Disease Category Database, Down syndrome ranks 178th in the funding of 291 disease categories (NIH, 2020b) and it is under-researched, with only 56 clinical trials registered on the Clinical Trials.gov website (USLoM). The NIH increased its focus and funding for Down syndrome research in 2018 (NIH, 2020a). In anticipation of increased research on Down syndrome, this study aimed to understand parent attitudes and perspectives towards research to inform subsequent research recruitment efforts (Becker et al., 2004; Brown et al., 2001; Heller et al., 2006; Hodapp, 2007; Long et al., 2001). Understanding parental perspectives towards research is crucial given reported challenges with recruiting individuals with Down syndrome for clinical trials (Brown et. al, 2010; NIH, 2020a; Williams et al., 2018). This study aims to identify parental outreach and engagement needs, to inform researchers of potential recruitment barriers. Research topics that interested parents of individuals with Down syndrome most were also collected to direct research efforts to those most likely to engage families and research participants.

Parents of children with intellectual and developmental disabilities are critical stakeholders in the decision-making process for enrolling their child in any research study (Bye & Aston, 2016; Hodapp, 2007; Johnson & DeLeon, 2016; Kripke, 2018). To support the critical role of these parents in research recruitment, prior research has identified that all parents could benefit from increased support and education about research, the consent process and clinical trials (Clausen et al., 1954; Heller et al., 2006; Lidz & Appelbaum, 2002). Providing education and support to parents in general enables more parental participation in healthcare and

healthcare research (Comer, 2005; Harris & Roberts, 2003; Magrab & Bronheim, 2018; Rao et al., 2011). Further, parents of typically developing children and individuals with intellectual and developmental disabilities are reported to be willing to allow their child to participate in research when there are perceived benefits for their child (Brody et al., 2005; Clausen et al., 1954; Kassam-Adams & Newman, 2005; Nock & Kazdin, 2001; Ouellette-Kuntz et al., 2013). Engaging parents as stakeholders in research agendas increases engagement and research participation, and researchers in turn benefit from understanding parental perspectives to improve their study design (Lister et al., 2003; Scotti et al., 2012; Witting et al., 2012).

Despite the empirical evidence for increasing stakeholder engagement in research, barriers to engaging parents of children in research persist. Socioeconomic status and low health literacy correlate to reduced engagement levels, limited means of transportation to participate and parental hesitancy towards participating in studies that involve more than 'minimal risk' to their child (Brody et al., 2005; Clausen et al., 1954; Kassam-Adams & Newman, 2005; Nix et al., 2009; Nock & Kazdin, 2001; Yin et al., 2012). Relationship trust is another barrier. Parents report distrust in the use of data is another. Further, there is inferred power differentiation between the participant and the researcher, contributing to scepticism from parents towards research, access to information and trusting the importance of the research (Banas et al., 2019; Barnes, 2006). These barriers are shared by families of typically developing children and of families of children with an intellectual disability (Becker et al., 2004; Freedman, 2001; Ouellette-Kuntz et al., 2013). Researchers also face additional systemic barriers in research recruitment. Limited funding or resources to aid in recruitment process led to low recruitment rates (Banas et al., 2019; Green et al., 2006). Access to individuals with or families of individuals with intellectual and developmental disabilities is an additional challenge to recruitment. Utilising other resources such as partnerships with other organisations is warranted. However, these partnerships are not always reliable, and some organisations act as 'gatekeepers' to sharing information with populations needed for research (Lennox et al., 2005; Nary et al., 2011; Williams, 2020). There is a need to understand these research barriers specific to target study populations in order to facilitate research on specific groups of individuals. Barriers for research participation may be averted through better alignment of education and information distribution channels specifically related to research purposes on a target population, such as Down syndrome (Clausen et al., 1954; Cleaver et al., 2010; Faragher, 2019; Magrab & Bronheim, 2018; Wooten et al., 2006).

Few studies have inquired about parent perspectives on research related specifically to Down syndrome. A study focused on evaluating parents of dependents with Down syndrome found their perspectives on finding a 'cure' for Down syndrome (43%) would be a desirable research study as would studies that can reverse cognitive impairment (61%; Inglis et al., 2014). Overall, parents do have a favourable outlook towards raising a child with Down syndrome and desire the best possible outcomes in life, health and development of independence for their child (Skotko, Capone, & Kishnani, 2009; Skotko, Kishnani, et al., 2009). Research done by Skotko, Capone, & Kishnani, 2009, Skotko, Kishnani, et al., 2009), which collected parental perspectives on how they would prefer to receive a diagnosis of Down syndrome was instrumental in changing healthcare perspectives, highlighting the importance and impact of parental perspectives and need for engagement to solve problems. Identifying parental perspectives specific to research in Down syndrome is anticipated to have a similar impact on engaging the community to help advancing research in Down syndrome and how to engage with participants better.

The advancement of healthcare is and should be a collective effort that includes the voice and exchange of ideas between many stakeholders, including the parent, provider(s) and social and basic science researchers. These stakeholders should be engaged intrasectionally in the design and development of research agendas, engagement practices and discuss the barriers and challenges from each perspective (Lister et al., 2003; Northway, 2014; Scotti et al., 2012; Witting et al., 2012). As specialisations pull away from the generalisation of practice, there is a greater need to manage this gap of understanding between researchers and participants in order to create the engagement necessary to overcome these barriers in research (Rao et al., 2011). This need is especially significant to the field of Down syndrome, where maintaining the information related to multiple medical co-morbidities and addressing changes in practice is challenging (Chicoine et al., 2021).

Given the recent increased research focus and funding on Down syndrome in the United States (INCLUDE Project), there is a need to understand stakeholder perspectives related to research to enhance research recruitment efforts specific to Down syndrome. Our study evaluates parent perspectives to inform researchers in clinical and basic science. Additionally, this study aims to support the development of engagement practices for recruitment strategies that address parental concerns and needs. We distributed surveys to parents of individuals with Down syndrome to understand their research interests, research design preferences and barriers to participation. The research goals were to understand (1) how parents obtain and understand information about research in Down syndrome; (2) parental willingness to participate in research specific to Down syndrome; (3) barriers and parental concerns with participating in research; and (4) what areas of research parents would like to see for Down syndrome.

2 | METHODS

2.1 | Study design

Surveys were distributed online in early 2018 through a broad contacts listserv and specific social media platforms maintained by the LuMind Research Down Syndrome Foundation (LuMind RDS) and International Down Syndrome Coalition (IDSC) non-profit organisations, which subsequently merged in early 2019. These distribution lists are comprised of individuals who had previously engaged with or donated to the organisations, had an interest in community engagement or research, or were raising a dependent with Down syndrome.

Roughly, 12,000 constituents across the two organisations were sent an email inviting them to complete the survey if they were parents of individuals with Down syndrome. The number of email recipients who are parents of individuals with Down syndrome is unknown. The investigational survey remained open for 30 days, and two additional follow up emails were sent through the LuMind Down syndrome listserv. There was no compensation for participation in this survey and participants self-selected themselves to complete the study. The Institutional Review Board approved the study at Antioch University under the protocol title: Identifying Research Disparity between Community Interest and Research Practice.

2.2 | Participants

Participants were required to be a parent of an individual with Down syndrome. A total of 228 parents completed the Down Syndrome Parental Perspectives Survey (see Table 1). Respondents ranged in age from 18 to 79 years and were predominantly female (89.3%). Ethnically, most of the respondents self-identified as Caucasian (86.9%). The majority of parents (73.0%) who responded had a child under 18 years, and most responses were from parents who had a child between the ages of 6 and 11 years (28.5%). Respondents self-reported an average household income of $100,000–149,000, and 39.3% indicated they completed graduate school. Respondents indicated they were from 43 different states in the United States and nine other countries.

TABLE 1 Demographics of surveyed respondents ($n = 215$)

	Percentage
Gender (female)	89.3%
Age of parent	
Under 30	2.4%
31–39	17.7%
40–49	36.7%
50–59	29.3%
Over 60	14.0%
Ethnicity	
American Indian or Alaskan Native	0.5%
Asian/Pacific Islander	2.8%
Black or African American	1.4%
White/Caucasian	86.9%
Multiple ethnicities	8.2%
Household income	
$0–49,999	8.8%
$50,000–99,999	21.9%
$100,000–149,999	24.2%
$150,000–199,999	16.3%
$200,000 and over	15.3%
Prefer not to answer	13.5%
Highest level of education	
High school	6.1%
Some college	13.5%
College	31.3%
Some graduate school	9.8%
Graduate school	39.3%
Gender of the child (female)	49.7%
Age of child	
0–5	21.0%
6–11	28.5%
12–18	23.5%
19–30	21.0%
31 and older	6.0%

2.3 | Measures

The survey included 42 questions addressing four research goals. The first goal was to understand how parents obtain and interpret information on Down syndrome research. The questions were designed to gauge parental knowledge about research and confidence in deciphering the research information. Survey questions specifically addressed how parents obtain research information, their level of confidence in understanding and evaluating the research literature, and whether parents had adequate access to research literature (journals, articles and peer-reviewed references). The second goal was to understand parental willingness to participate in research specific to Down syndrome. Questions were designed to understand current research perceptions, their willingness to participate in different research types, and their past participation research experiences. Parents who had previously participated in research studies were asked additional questions on the purpose and level of involvement. We defined invasive research as procedures that include drug delivery (oral or injection), blood draws, CAT scan, ultrasound, MRI's, X-rays and implantation of a device. Non-invasive procedures included participation in a survey or observational study. The third aim addressed parental attitudes towards research and barriers with respect to parental concerns with research studies. The fourth aim addressed parental interests on types of research and research subjects they feel would benefit their child's health outcomes. Questions were posed using Down syndrome relevant topics with the option for open-ended responses. Parents were also asked about their perceptions of the current state of research on Down syndrome, since their health extends across several co-morbid conditions. Parents also responded on specific topics they wanted to see more focused research studies on.

Questions varied from ordinal response options (1–5; Not likely, Somewhat Likely, Likely, Most Likely and Extremely Likely) to closed-(multiple choice) and open-ended (comment) questions. Response types were dependent on the type of question.

2.4 | Data analysis

Descriptive statistics were used to summarise demographics and closed response options. Missing data were excluded and numerators of participants completing a survey question are reported where appropriate. Chi-square statistics were used to determine whether responses were related to demographic variables.

Open-ended responses were analysed using thematic analysis by two study staff. Groupings were determined independently, and thematic analysis for consistency in interpretation was compared against each other's results.

3 | RESULTS

3.1 | Aim 1: How parents obtain and understand research

Gauging how comfortable parents are with understanding results of research and seeking information about a co-morbidity associated with Down syndrome was part of this aim. Parents reported obtaining information on Down syndrome research from non-profit foundations (84.2%), Google (75.4%), handouts from medical providers (43.4%), social media (41.7%), scientific journals (41.7%) and PubMed (29.4%). Less than half of parents indicated they used scientific journals or peer-reviewed references to obtain information on research on Down syndrome.

Parental confidence in understanding scientific language was high, with 51.8% reported being comfortable, and 43.6% being somewhat comfortable. Only 4.5% stated discomfort with understanding scientific language in literature searches. There was no relationship between comfort with scientific language and race ($\chi^2[10] = 11.22$, $p = .34$) or level of income ($\chi^2[10] = 10.75$, $p = .38$). Confidence in understanding scientific language was the greatest among respondents with graduate degrees (64.3%) and lower than expected among college graduates (40.3%; $\chi^2[14] = 26.86$, $p = .02$).

3.2 | Aim 2: Parental willingness to participate in research specific to Down syndrome

Knowing parental willingness to participate in research, different research types and collect rates of prior participation in research was the focus of Aim 2. A single respondent indicated resistance to including their child in a research study, 72.4% of parents indicating they would enrol their child in a research study, and 27.2% indicated that they would consider enrolment in a study.

Thematic analysis of open-ended responses indicated several factors that would impact parental willingness to enrol their child with Down syndrome in research. These factors include associated risks/benefits to their child, the purpose and level of transparency in the study. Benefits to the child was a sentiment shared by most parents, with parents indicating that they would be more likely to participate in a research study if it benefitted their child (93.4%) or the larger Down syndrome population (80.7%). A parent in the survey stated, 'I am interested in research that will improve the quality of life for individuals with Down syndrome, not the research of those with Down syndrome to impact individuals with 46 chromosomes'. Parents generally expressed concern regarding study risks and protection for their child's well-being. Another parent shared, 'I do not want her to be poked and prodded and looked at like some weird science experiment. She has been treated this way by a few geneticists and specialists, and I quickly found others to replace them'.

Parents expressed that a research study must have a clear research aims and goals and direct purpose for people with Down syndrome. One parent commented: 'Some studies have collected data with unclear use which invalidates the study [for me]. Nobody wants to be a guinea pig'. A consistent theme related to the parents' willingness to sign up for a research study was the need for transparency and assurance people with Down syndrome would benefit. Parents expressed it was essential to have access to the study results, particularly medical study-related results.

Other factors impacting parental willingness to participate in the research included: perceived level of child cooperation, the capacity of the child to participate actively in the study, the capacity of the parent to actively participate in the study (time commitment, competing responsibilities), perceived ethics and participant privacy, whether the study included invasive procedures and inclusion/exclusion study criteria.

Parents were asked to rank their likelihood of participation on a 1–5 scale based on different research study reinforcers. Parents were most likely to participate if they received a copy of medical results ($M = 4.1$, $SD = 1.0$), without any reimbursement ($M = 3.4$, $SD = 1.2$) or with mileage reimbursement ($M = 3.1$, $SD = 1.3$), and somewhat likely to participate if the parents received financial reimbursement ($M = 2.8$, $SD = 1.4$) or their child received a reward ($M = 2.8$, $SD = 1.2$).

3.2.1 | Previous participation in research

Parents involved in previous research studies indicated their experience. Over one-third, (36%) of parents had previously enrolled their child in a research study. Of these, 55.6% participated in only one study, 20.4% in two, 15.9% in three and 7.9% in four or more research studies. Families most often participated in survey research, studies involving biospecimens, and clinic-based research (see Table 2). Parents reported the focus of the research was on sleep, cognition, language and genotyping.

3.3 | Aim 3: Parental barriers and concerns about participating in future research

Parents had an even distribution regarding concerns (32.9%), having no concerns (35.5%), and being uncertain (31.6%) regarding enrolling their child in a research study. Barriers to enrolling in research studies reported by parents included risk/benefits to the child, access to

TABLE 2 Research types of research participation ($n = 228$)

	Percentage
Ever enrolled in research study	36.5%
Type of research study enrolled in	
Survey	29.8%
Observational study with daily journaling	7.0%
Observational study	10.1%
Biospecimen donation (blood, DNA, urine, tissue)	18.9%
Clinic-based research	16.2%
Clinical trial	5.3%
Use of device (watch, oxygen sensor, monitoring device)	5.3%

TABLE 3 Parental perceived research needs and areas of interest ($n = 228$)

Area of research	Needing more investigation (%)	Area of parental interest (%)
Alzheimer's	68.4	61.0
Cognition	62.7	73.2
Gene therapy	46.9	35.1
Speech	43.9	65.4
Meaningful inclusion	41.7	47.4
Independence	40.8	61.4
Immune conditions	37.7	32.0
Digital medicine (online games to improve learning, memory, etc.)	35.5	40.4
Nutrition	34.6	41.2
Sleep conditions	33.3	32.9
Social development	28.9	36.8
Thyroid conditions	26.3	26.3
Breathing conditions	18.4	13.2
Heart conditions	16.7	11.8
Early intervention	16.7	11.8
Motor development (sitting, crawling, walking, etc.)	13.2	14.0

study results, adequate study compensation, and the logistics of participating in the study (time commitment). The factors most often reported by parents for considering participation in a research study were the risks ($M = 4.2$, $SD = 1.4$) and benefits ($M = 4.2$, $SD = 0.9$). Parents were likely to consider access to study results ($M = 3.9$, $SD = 1.0$) and time commitment ($M = 3.9$, $SD = 1.0$), and somewhat likely to consider reimbursement for time ($M = 2.6$, $SD = 1.2$).

Parents shared and identified logistical barriers of participating in research. Concerning travel to the study location, many parents were willing to travel to research within 50 miles of their home (57.5%). An additional 23.7% of parents were willing to travel 51–100 miles, 7.5% would travel 100–200 miles and 11.4% would travel over 200 miles to participate in a research study. Parents were generally willing to participate in longitudinal studies lasting 1 year or more (80.9%). Few parents were willing to participate in studies with a shorter duration of 6 months (10.2%), 3 months (5.6%), 1 month (1.4%) or 1 day (1.9%).

Parents rated their likelihood of enrolling in non-invasive research procedures higher than invasive procedures. Parents were willing, in descending order, to include their child in a research study that involved surveys ($M = 4.3$, $SD = 0.9$), focus groups ($M = 3.9$, $SD = 1.1$), journaling ($M = 3.6$, $SD = 1.1$), blood draws ($M = 3.5$, $SD = 1.2$), ultrasounds ($M = 3.3$, $SD = 1.2$), wearing devices ($M = 3.2$, $SD = 1.2$), X-ray ($M = 3.2$, $SD = 1.2$), MRI ($M = 2.8$, $SD = 1.3$), CAT scan ($M = 2.7$, $SD = 1.3$), PET scan ($M = 2.7$, $SD = 1.3$), taking a medication ($M = 2.1$, $SD = 2.1$), or receiving an injection ($M = 1.8$, $SD = 1.0$). Parents were not likely to enrol their children in a study where their child would receive an injection (49.1%), take a medication (34.2%), receive a PET scan (21.9%), an MRI (21.5%) or CAT scan (21.5%). Other study designs were endorsed at less than 10% of parental willingness to enrol their child in that type of study.

3.4 | Aim 4: What kind of research parents would like emphasised in Down syndrome

Parents shared research areas, specific to Down syndrome, they would like to see more investigation and which they perceived as being most interested in (see Table 3). Parents perceived the greatest need to investigate Alzheimer's Disease, cognition, gene therapy and speech/language. Similarly, parents were most interested in research on cognition, speech/language, independence and Alzheimer's Disease. Parents expressed extremely high rates of wanting to see more research to improve health and independence for individuals with Down syndrome (99.5%) and want more treatment options related to drugs, therapies and interventions available for individuals with Down syndrome (91.8%).

The majority of parents (85.9%) indicated not enough research was being done on Down syndrome. Comments exemplary of this position include: 'I think that the Down syndrome population is underserved and undervalued. I think we have a great deal to learn about the 21st chromosome and especially how it relates to Alzheimer's disease'. Parents also expressed concerns on current gaps for research in Down syndrome. With the number of overlapping health co-morbidities impacting individuals with Down syndrome, one parent expressed, 'All aspects of Down syndrome that do not involve intellectual disability have been severely understudied'. An additional theme surrounded parental concerns with the availability of technology and interventions to support their child's needs. Parent's expressed concerns with the limited choice in therapies and treatments available for their child with Down syndrome.

4 | DISCUSSION

This survey aimed to understand parental perceptions about research in Down syndrome. Questions specifically elicited responses from parents of children with Down syndrome regarding how they obtain and

understand research, their willingness to participate in research, barriers and concerns regarding participation in research, and their research priorities and interests. Overall, parents surveyed reported that they obtain information online, and were comfortable understanding the content and would be more willing to enrol in research studies that directly benefited individuals with Down syndrome. Barriers to participation noted in the survey responses included invasive procedures and some study logistics or demands. Parents also identified several research areas of critical interest that are not current targets within the Down syndrome research community.

Survey responses help understand how some parents of individuals with Down syndrome access research and understand research findings. Parental responses suggested a strong level of comfort with accessing and digesting information about research studies and findings that are obtained online or from non-profit foundations. Unfortunately, the information offered through Google, non-profit websites and printed materials is often over-generalised and damaging the impact of how research is communicated to the community (Burki, 2019; Grimshaw et al., 2012). Further, the average person does not evaluate a website with rigour, caring more about the superficial appearance than its content and privacy policy (Fogg et al., 2003). Generally, scholarly articles are not publicly accessible nor written in language a lay person can interpret. Together with reports on how families of children with Down syndrome obtain information, this finding highlights the research community's need to share research findings in lay terms online with the support of non-profit foundations. Resources to better understand the status and promise of research need to be developed in partnerships centred on collective leadership with non-profit foundations, the research community, medical providers, families and self-advocates to disseminate more explicit information regarding research to families. These strategies are essential considerations for strengthening research collaborations and participation.

Our investigation into understanding a parents' willingness to allow their child to participate in a research study demonstrated that surveyed parents were sceptical about participation in research. Despite over 70% of parents reporting willingness to participate, only about one-third of parents had enrolled their children in a research study. These results are consistent with other research investigating parents' willingness to allow their dependent to participate in research (McDonald et al., 2018; Reines et al., 2017). Parents reported concerns for who benefits from the results of a research study and wanted to ensure that research would benefit the Down syndrome population directly and not utilised them as stepping stone for other research initiatives outside of the Down syndrome population (Faragher, 2019).

Barriers to research participation were consistent with those reported for the general population, including weighing risks and benefits to the child, access to study results, study compensation and time commitment (Brody et al., 2005; Clausen et al., 1954; Kassam-Adams & Newman, 2005; Nock & Kazdin, 2001; Ouellette-Kuntz et al., 2013). Additional barriers to research participation included the study design or use of invasive procedures. Parents were less willing to enrol their child with Down syndrome in a research study involving injections, medication or PET, MRI or CAT scans. Open-ended responses from parents alluded to having insufficient scientific knowledge to make informed decisions about participation in a research study. These responses indicate a potential need to increase educational information regarding study purposes and using recruitment strategies that support parents in making decisions regarding study involvement (Lister et al., 2003; Scotti et al., 2012; Witting et al., 2012).

Surveyed parents expressed a desire to know more about research efforts related to cognition, speech/language, independence, Alzheimer's disease, new therapies, technologies and healthcare advancements. Parental research interest areas are in line with ongoing research studies as listed on clinicaltrials.gov (USLoM, 2020). However, parents expressed an interest in more research on the multitude of medical co-morbidities present among individuals with Down syndrome (INCLUDE Project Research Plan, 2020). While clinicaltrials.gov listings of current research studies being done for Down syndrome is not comprehensive of all current research in this population, it does demonstrate that studies currently recruiting are shy of meeting the vast parent research interests, and topics for future study on medical comorbidities.

Survey responses elucidated a disparity between the proportion of parents wanting more therapy and treatment options for their children (91.8%), the proportion of parents willing to have their child with Down syndrome participate in research (72.4%), and the proportion of parents who have enrolled their children in research studies (36%). This disparity highlights the need to understand barriers to research participation and the type of engagement practices needed to encourage participation. This disparity could be accounted for by power distance and patient protection between researchers and parents caring for dependents with Down syndrome (Hofstede, 2011; McDonald & Keys, 2008; Stringer et al., 2018). Power distance refers to the relationship between a person of position, like a researcher, and other roles or, in this case, participants (Hofstede, 2011). Creating relational engagement and dialogue between parents and researchers as well as the research enterprise could benefit recruitment, consenting procedures, participation rates and ultimately the success of research outcomes in benefitting individuals with Down syndrome. We recommend that parents and self-advocates be engaged in the design and planning of research studies with researchers (McDonald & Kidney, 2012). This involvement and engagement would not only benefit the recruitment and participation in research, it would also aid in saving valuable investigator time and resources in the recruitment process. Additional resources such as non-profit organisations that support research efforts in Down syndrome, provider conversations, websites, social media and other interfaces can also be engaged to help facilitate this power distance.

These survey results offer researchers insights into study design that can create greater engagement and increase parents' participation levels. Results from this study help to inform researchers about the parental concerns on research and research participation to guide recruitment efforts and reduce participation barriers. Research will

benefit from engaging with key stakeholders (caregivers, non-profit foundation) early and often to inform the study design and recruitment plan. It is important to stress how messaging and education is delivered to parents by ensuring the intent, purpose and message of the study are delivered and received by parents effectively. Surveyed parents heavily weighed the benefit of research to individuals with Down syndrome and expressed a strong interest in increased research on more co-morbid conditions. Researchers are encouraged to design and communicate their research studies to benefit the individual with Down syndrome while mentioning an additional potential benefit that will advance the understanding of the same co-morbid conditions in the general population (INCLUDE Project Research Plan, 2020). This messaging will likely mitigate some parental concerns in participating in research.

Non-profit foundations that promote education, awareness and advocacy related to Down syndrome can also benefit from these survey results. Survey findings have implications for topic areas for foundations to target when providing education regarding community research efforts and research findings. Ensuring that the information is easy to understand and that parents can differentiate research rigour in what they read online is essential. Collaborations between researchers, clinicians and non-profit foundations is essential for creating greater transparency and purpose in healthcare advancements by educating parents on evaluating research and interpreting research findings.

Despite the strengths of obtaining perspectives from surveyed parents on research in Down syndrome, there are some limitations present in the current study. LuMind RDS and IDSC are two US-based non-profit organisations. LuMind RDS's mission primarily focuses on research. IDSC's primary mission focus was not on research but advocacy for people with Down syndrome and their families. The results from both non-profit organisations were similar, and the data were merged. The listserv used for this survey was non-targeted. It included duplicate contact information and emails for philanthropic donors who may not have a child with Down syndrome, limiting the ability to determine the response rate to the survey. Further, a majority of the individuals on the listserv were likely interested in research. The survey was leveraged as a sampling of questions to help target further research into our understanding of creating better engagement with families and research on Down syndrome. When conducting web-based survey research to a non-targeted audience, there can be many associating limitations in using this method. Research participation is generally greater when participants are engaged with the researcher and understand the study's intent (Fan, 2010; Nardi, 2018; Ruel et al., 2015). As the survey was distributed blindly via a web-based distribution to a non-targeted population, the response rate appears low. However, the correct response rate is indeterminate as the actual number of eligible participants contacted is unknown. Despite the unknown response rate, participants' information contributes vital information and recommendations on creating stronger relationships and engagement with study participants. Experts in survey methodology argue that findings from low response rates contribute to research and that low response rates are not always a critical limitation to research (Hendra, 2019). A further limitation of the study is that the socioeconomic status and education levels skewed to affluent and educated participants. While these sample demographics suggest concerns for survey responses' generalisability, they also demonstrate responses from an invested sample and where they would like to see more research conducted. Further research is needed to understand socioeconomic barriers and access to receiving information about research studies.

Surveyed parents of individuals with Down syndrome offered insight on how research studies could be designed, how research education for parents could be developed and how research agendas for Down syndrome can be shaped. The broad need for increased research on co-morbidities highlights the importance of increased funding for research in Down syndrome. The need for support and extended time when working with vulnerable populations in research is also acknowledged as an important factor for designing studies to benefit people with Down syndrome. The information provided through the parental survey responses emphasises the need for designing better research studies and engaging parents in the research processes. Perspectives from parents, researchers, advocacy groups and even medical personnel offer an inclusive opportunity for developing robust research agendas for Down syndrome. Funding for research in Down syndrome has been historically limited, finding alignment between research and parental interests will lead to greater research discoveries and improving health outcomes for individuals with Down syndrome.

CONFLICT OF INTEREST

The authors declare no conflicts of interest to disclose. This research would not have been possible without the contributions of the participating families.

DATA AVAILABILITY STATEMENT

Data are available on request from the authors.

ORCID

Amanda Nicole White https://orcid.org/0000-0002-1099-1325

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