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Piloting the use of global health measures in a Down syndrome clinic

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Abstract

Purpose: People with Down syndrome (DS) have a unique medical profile which may impact views of health. We aimed to explore the use of global health measures in DS.

Methods: Prospective survey in the Mass General Hospital Down Syndrome Program (MGH DSP) from December 2018 to July 2019 with Patient Reported Outcomes Measurement Information System (PROMIS)® instruments of global health. Analyses included use of scoring manuals, descriptive statistics and dependent samples t test.

Results: Seventeen adolescents, 48 adults with DS and 88 caregivers returned surveys; 137 were complete. Incomplete responses and notes showed limitations of the instruments in this population. Global health T-scores did not differ from the available comparative standardized scores to these measures from PROMIS® reference population ($p > 0.05$).

Conclusions: In the MGH DSP, pilot global health instruments were completed by some adults with DS and caregivers, with some limitations and scores similar to the PROMIS® reference population.

KEYWORDS
Down syndrome, global health, health, health-related quality of life, trisomy 21

1 | INTRODUCTION

Down syndrome (DS) is caused by extra genetic material from chromosome 21 and is diagnosed in infants around the world (de Graaf et al., 2017; Hughes-McCormack et al., 2020; Korenberg et al., 1994; Presson et al., 2013; Wu & Morris, 2013). Due to advances in medical care and access, life expectancy for people with DS has greatly improved over time (de Graaf et al., 2017; Yang et al., 2002), with a median lifespan of 58 years as of 2010 (de Graaf et al., 2017). The health profile of individuals with DS is unique due to differences in rates of co-occurring medical conditions in children and adults (Bull & Committee on Genetics, 2011; Jensen & Bulova, 2014; Kinnear et al., 2018). Compared to the neurotypical population, individuals with DS have an increased risk for dementia but a decreased risk for cardiovascular disease (Esbensen, 2010). Further, obesity is increased in people with DS and associated with risk for sleep apnoea, behavioural effects, impacts on mobility and independence (Basil et al., 2016; Bertapelli et al., 2016; Chen & Ringenbach, 2018; Smith & Ulrich, 2008). To address these unique medical needs, DS health care guidelines have been published (Bull & Committee on Genetics, 2011; Jensen & Bulova, 2014; Tsou et al., 2020). These additional health needs may lead to additional medical surveillance, medical visits and associated costs (Bull & Committee on Genetics, 2011; Kageleiry et al., 2017) and could impact how families' view the health of their son/daughter with Down syndrome.

While previous work has measured adhere to health care guidelines (Santoro et al., 2018), health care outcomes (Graves et al., 2016; Jacola et al., 2014; Rafii, 2016; Santoro et al., 2018). (Schieve et al., 2009), and the effects of comorbidities (LonDownS Consortium et al., 2020), very little research has focused on patient-reported outcomes. In one study, adults with Down syndrome completed self-report of online health survey about health indicators, such as physical activity, diet / nutrition and BMI (Havercamp et al., 2017), but more research measuring patient-reported outcomes in DS is needed. Patient-reported health measures are an area of emphasis with NIH initiatives, such as Patient Reported Outcomes Measurement Information System (PROMIS)® that provides item banks of generic measures applicable across populations and chronic conditions, measuring 6 chronic paediatric conditions, but not DS (Cella et al., 2007; DeWalt et al., 2015). Three instruments in PROMIS®, the Global Health Scale v1.0, the Pediatric Scale v1.0 and the Parent Proxy Scale v1.0 (Cella et al., 2007), assess global health. Global health is an overall measure of an individual's perceived physical, mental and social health. The measures are generic, rather than disease-specific. Global health is one's overall self-perceived health status—that is, *how healthy do you feel?* In contrast, other instruments measure health-related quality of life (HRQOL), which is defined as a multi-dimensional concept that includes domains related to physical, mental, emotional and social functioning. HRQOL goes beyond direct measures of population health, life expectancy and causes of death and focuses on the impact that one's health status has on quality of life – in other words, *how does your health impact your QOL?* (Health-Related Quality of Life & Well-Being, 2019). Global health and HRQOL are both patient- (or parent-) reported perceptions of health. However, *global health* is a metric of health status while *HRQOL* measures the impact of health status on QOL. As related but separate concepts, a given medical diagnosis, such as DS, can therefore be understood to impact a person's quality of life and perceived health status in distinct ways.

We began this study to explore the use of *global health* measures with people with DS and caregivers to augment our existing clinical intake (Chung et al., 2020). Specifically, we surveyed adolescents and adults with DS in our clinic collecting information about views of their overall health. As such, we aimed (1) to test if these measures from the general population can utilized for people with DS in our clinic and benchmarked against the PROMIS® reference population, (2) to gather feedback on any difficulty answering the questions and (3) to inform further research using these measures in individuals with DS.

2 | METHODS

2.1 | Participants

The Massachusetts General Hospital Down Syndrome Program (MGH DSP) provides medical care for more than 500 individuals with DS each year, with the majority of individuals coming from the New England area. The MGH DSP is a comprehensive, outpatient, subspecialty clinic focused on medical care related to an individual's diagnosis of DS. The MGH DSP consists of multiple interprofessional team members (e.g., nutritionist, social worker, physician). During a clinic day, individuals are in the MGH DSP for a few hours with multiple appointments during an encounter.

All individuals and their accompanying caregiver who arrived for visits with one of three physicians in the MGH DSP from December 2018 to July 2019 were eligible. Inclusion criteria for caregivers included a clinic visit to the MGH DSP, individual with DS age 5 years or greater and English speaking. Additional inclusion criteria for individual with DS self-report included paediatric age 7–17 years, adult age 18 years or greater. Exclusions included those families who did not have English proficiency. Individuals were not excluded based on cognitive status, and proxy response was accepted if an individual could not complete on their own. Demographic information including age and gender were collected from the electronic health record; race and ethnicity were asked directly.

2.2 | Instruments

We sought out instrument(s) to assess global health or an overall measure of an individual's perceived physical, mental and social health. Our primary goals were to identify instrument(s) which could be also be administered to both individuals and accompanying caregivers (i.e., both parent proxy and adult versions were available), were freely available, could be scored by a physician (i.e., did not require additional training to complete scoring) and were feasible to be completed in clinic. Due to lack of consistent access to a tablet or laptop, paper versions were also required.

To search for candidate instruments, we conducted a literature review with terms such as 'health status', 'global health', and others used in the United States general population or in specific conditions. In searching for instruments, we identified some that measure health-related quality of life or specific components of health (e.g., cognition, sleep, behaviour). These were excluded as they did not capture *overall* self-perceived health. Literature review did not identify any instruments which met these criteria and were previously validated in individuals with intellectual disability or DS; therefore, this was not a cause for exclusion. Candidate instruments were compared, and study instruments were selected by the physician team based on meeting the criteria described above (Currie & World Health Organization, 2004; Watrowski & Rohde, 2014).

In this study, we used three instruments which measure global health: the Global Health Scale v1.0, the Pediatric Scale v1.0 and the Parent Proxy Scale v1.0 (Cella et al., 2007). These three related instruments have versions for both parent- and self-report, are freely available on the PROMIS® website with available scoring guides and could be completed in a clinic visit. All are fixed-length scales. These three instruments are generic, rather than disease-specific and often use an 'In General' item context, as it is intended to globally reflect individual health. The three PROMIS®

global health measures include questions such as 'In general, would you say your health is:', with 5-point Likert responses from 'Excellent' to 'Poor'. These PROMIS® global health measures have been validated in the general population (Forrest et al., 2014, 2016). PROMIS® measures have been used in arthrogryposis, Turner syndrome and neurologic conditions including epilepsy and Parkinson's disease (Shulman et al., 2019; Thompson et al., 2020; Wall et al., 2020).

The PROMIS® Global Health Scale v1.2 measures an individual's physical, mental and social health (Hays et al., 2009, 2017). The 10-item adult PROMIS® Global Health Scale v1.2 produces two scores: Physical Health and Mental Health. This is available for self-report in adults, age 18 years and older.

The PROMIS® Pediatric Scale v1.0 assesses a child's overall evaluations of his or her physical, mental and social health and is conceptually equivalent to its PROMIS® adult counterpart (Forrest et al., 2014, 2016). This is available for self-report in paediatrics, age 7–17 years of age. The 7-item paediatric and global health measure include a single factor and consequently one global health score. The '7 + 2' scales includes the same global health score plus one fatigue and one pain interference item which are scored independently. We used the '7 + 2' version.

The PROMIS® Parent Proxy Scale v1.0 assesses a parent's overall evaluations of his or her physical, mental and social health and is conceptually equivalent to its PROMIS® adult counterpart (Forrest et al., 2014, 2016). This is available for parents serving as proxy reporters for their child (youth ages 5–17). The 7-item parent proxy global health measures include a single factor and consequently one global health score. The '7 + 2' scale include the same global health score plus one fatigue and one pain interference item which are scored independently. We used the '7 + 2' version.

2.3 | Procedure

2.3.1 | Recruitment and survey administration

Individuals and caregivers at the MGH DSP were given paper versions of the three Global Health instruments during their clinic visit. They were provided with a packet that included an information sheet cover page that described the study, emphasized voluntary participation and specified consent/assent through completion of the survey and the relevant instruments for age. All persons gave their informed consent prior to their inclusion in the study. The physician explained the study; individuals and accompanying caregivers were invited to participate by the clinician during the visit, the relevant instruments and who should complete were explained, and the clinician answered questions about the study. We instructed respondents to answer all of the items presented. If respondents had questions about a specific question or components, they were instructed to list questions or comments in the margin. Individuals and accompanying caregivers were often able to complete the instrument while in the exam room waiting between providers. The individual with DS and accompanying caregiver then returned the completed survey to a team member.

Sources of data: Data from two sources: (1) collected survey responses in clinic to benchmark against, (2) available comparative standardized scores to measures from PROMIS®. The studies of the psychometric properties of PROMIS® measures use populations which reflect the general population and national norms (Forrest et al., 2014, 2016; Hays et al., 2017). The reference population for the PROMIS global health measures is the United States general population (*PROMIS® Reference Populations*, n.d.). We describe scoring in the Scoring section below.

Scoring: Responses were collected, recorded and translated from categorical responses to numerical values using the PROMIS® Measure-Specific Scoring Guides available online. Response options correlate to a value from 1 to 5 for each question. The total raw score is calculated through summing the values for the questions for a given respondent. Raw sum scores were converted to T-scores using the conversion tables in appendix 1 of the PROMIS® Scoring Guide (page 13). T-Score distributions are standardized such that a 50 represents the average (mean) for the United States general population and the standard deviation around that mean is 10 points (Cella et al., 2007). A higher PROMIS® T-score represents more of the concept being measured. Thus, as described in the Scoring Guides, a person who has T-scores of 60 for the Global Physical Health or Global Mental Health scales is one standard deviation better (i.e., more healthy) than the United States general population.

Scoring details for each instrument:

1. PROMIS Global Health v1.2: This instrument includes Physical Health and Mental Health subscales; the raw score for these is calculated from two questions each. The raw scores for the Physical Health subscale can range from 2 to 10 and raw scores for the Mental Health subscale can range from 2 to 10. The T-scores listed in Conversion Table for Physical Health range from 23.4 to 63.3 with Standard Errors of 4.8–7.1. The T-scores listed in the Conversion Table for Mental Health range from 25.8 to 64.6 with Standard Errors of 4.1–5.7. In subsequent text, referred to as 'Global Health'.
2. PROMIS Parent Proxy Global Health 7 + 2: This instrument includes seven questions. The raw score can range from 7 to 35. The T-scores listed in the Conversion Table range from 14.7 to 66.1 with Standard Errors of 2.9–6.5. In further text, referred to as 'Parent Proxy Global Health 7 + 2'.
3. PROMIS Pediatric Global Health 7 + 2: This instrument includes seven questions. The raw score can range from 7 to 35. The T-scores listed in the Conversion Table range from 16.0 to 67.5 with Standard Errors of 3.4–6.1. In subsequent text, referred to as 'Pediatric Global Health 7 + 2'.

The Institutional Review Board at Massachusetts General Hospital, the Partners Human Subjects Committee, approved this study.

2.4 | Data analysis

Descriptive statistics such as mean and standard deviation were calculated. T-scores on each measure were compared to the PROMIS® reference population mean using a one-sample *t* test. Among the adolescents and adults with DS who completed the instrument and had a parent complete the parent proxy form, comparisons were made within the individual between self-report score and parent report score using a paired sample, 2-tailed *t* test.

In the interest of completeness and transparency, we have reported on two small subgroups parents of children with DS age 5–6 years (*N* = 12) and children and adolescents with DS age 7–17 (*N* = 12).

The data that support the findings of this study are available from the corresponding author upon reasonable request.

3 | RESULTS

3.1 | Respondent characteristics

In the MGH DSP, 192 individuals and their caregivers were approached, of which 89 individuals returned at least one survey (Table 1). The 89 individuals included 14 individuals aged 5–6 years, 21 individuals aged 7–17 years, and 54 individuals aged 18 years and older (overall mean age: 19.0 years). The caregivers were all parents of the individuals and adults. A subset of our survey respondents was enrolled in a registry providing demographic information; this group of 50 individuals showed male predominance, mostly White or Caucasian race, and more were follow-up visits than new patient visits. The other 49 surveys were returned anonymously without demographic information available. Among this total group, 84 individuals had at least one complete survey, including the 50 individuals with demographic information available.

TABLE 1 Demographic Information of survey respondents

	All surveys received	Complete surveys
	N	N
Individual with DS age (years)		
5–6	14	12
7–17	21	20
18+	54	52
Mean (SD)	19.0 (12.0)	19.0 (12.0)
Additional demographic information, when available		
Sex available (*N* = 50)		
M	27	27
F	23	23
Race available (*N* = 35)		
White	31	31
Black or African American	0	0
Other	4	4
Visit type available (*N* = 52)		
New	20	20
Follow-up	30	30
Instruments		
Parent Proxy Scale v1.0	88	80
Respondent: parents of individuals with Down syndrome of age ≥5 years		
Global Health Scale v1.2 (Mental)	48	45
Respondent: adults with Down syndrome of age ≥18 years		
Global Health Scale v1.2 (Physical)	48	43
Respondent: adults with Down syndrome of age ≥18 years		
Peds Scale v1.0	17	12
Respondent: individual with Down syndrome age 7–17 years		

3.1.1 | Responses

In total, 153 surveys were returned: 88 Parent Proxy Global Health 7 + 2 surveys, 48 Global Health surveys and 17 Pediatric Global Health 7 + 2 surveys. In reviewing the responses, 16 surveys contained blank, n/a or'?' to questions that comprised the raw score calculation and were excluded from final analyses; additional notations in the margin were recorded (Table 2). Of the eleven Parent Proxy Global Health 7 + 2 surveys with missing items or annotations, the question 'Pf2pain5r' and 'PedGlobal6_PXR1' were the items most often not answered with 5 and 6 parents not answering these, respectively. Of the twelve Global Health scales with missing items or annotations, some were completed but with parental notations, while missing items were distributed among the questions. Of the five Pediatric Global Health 7 + 2 surveys, the question 'PedGlobal6R1' was blank in four, while the fifth was completed by the parent. Overall comments focused on language ability, factors on mood and uncertainty about the individual with DS's ability to complete the survey.

3.2 | Complete Surveys

In total, 137 complete instruments that did not have missing items could be scored. Eighty surveys were received from parents of individuals with DS on the Parent Proxy Global Health 7 + 2. Mean T-score was 46.3 for all parent responses; mean T-scores for parents

TABLE 2 Responses to Instruments including those which were left blank, and margin notations

Parent Proxy Scale v1.0—Global Health 7+2										
	Global01_PXR1	Global02_PXR1	Global03_PXR1	Global04_PXR1	PedGlobal2_PXR1	PedGlobal5_PXR1	PedGlobal6_PXR1	Pf4fatigue3r	Pf2pain5r	Overall comments
A1							Cannot express self		Cannot express pain	
A2	B						?		B	
A3							?	B	B	
A4										A4 is very close to his family incl. his sister & nephew. He occasionally texts friends, which he enjoys. A4 Is searching for office work after a layoff 1.5 years ago now. It's a challenge and this affects is mood & anxiety understandably.
A5	B	B								
A6							n/a			
T1						has fun with family often				
T2									?	
C1									n/a	
C2							n/a		n/a	
C3									n/a	

Scale v1.2—Global Health											
	Global01	Global02	Global03	Global04	Global05	Global09r	Global06	Global10r	Global08r	Global07r	Overall comments
A7									B	B	'Guessing' 'Can't really answer these'
A8										B	Sometimes my mood is not good. I get anxious because I worry about a job. Sometimes I feel pain when I'm upset
A9	B	B	B	B	B	B	B				
A10						B	B	B	B	B	
A11								B	B	B	
A12											A12 was unable to understand questions
A13											*completed by Dad
A14											I'm not sure if he understands the questions or word meanings even with definitions
A15											*mom answered
A16											*As per A16 (patient)

(Continues)

TABLE 2 (Continued)

Scale v1.2—Global Health											
	Global01	**Global02**	**Global03**	**Global04**	**Global05**	**Global09r**	**Global06**	**Global10r**	**Global08r**	**Global07r**	**Overall comments**
A17											*completed by parents because patient could not complete
A18	B	B	B	B	B	B	B	B	B	B	Not possible

Pediatric Scale v1.0—Global Health 7 + 2										
	Global01R1	**Global02R1**	**Global03R1**	**Global04R1**	**Ped Global2R1**	**Ped Global5R1**	**Ped Global6R1**	**2876R1r**	**3793R1r**	**Overall comments**
T3										*Completed primarily by parent
T4	B	B	B	B	B	B	B	B	B	Non-verbal
T5						B	B	B	B	
T6		B					B			
T7							B			

Abbreviations: A, patient age ≥18 years; B, blank; C, patient age 5–6 years; T, patient age 7–17 years.

TABLE 3 T-scores of Global Health in Down syndrome (DS) on 4 measures in comparison with standard score in general population.

	N	Mean	Min	Max	SD	*p*-value[a]
Parent Proxy Scale v1.0 (all ages)	80 parents	46.4	31.2	66.1	8.30	.056
Child with DS age 5–6	12 parents	47.4	31.2	60.2	8.47	.198
Child with DS age 7–17	20 parents	48.5	37.9	66.1	8.23	.262
Child with DS age 18+	48 parents	45.2	34.6	63.2	8.07	.054
Global Health Scale v1.2 (Mental)	45 adults with DS	52.3	38.8	67.6	7.20	.108
Global Health Scale v1.2 (Physical)	43 adults with DS	51.5	34.9	67.7	8.32	.183
Peds Scale v1.0	12 children and adolescents (age 7–17) with DS	49.5	37.2	58.3	6.32	.622

[a]Two-tailed t test comparing to general population mean = 50, standard deviation = 10.

TABLE 4 Matched T-score measures compared for teens and adults with Down syndrome and parent proxy (*N* = 46).

						Compared to PP	
						N	53
						Mean (SD)	46.2 (8.01)
						2-tailed, paired *t* test	
Instrument	***N***	**Mean**	**Min**	**Max**	**SD**	***t*-value**	***p*-value**
Adult Phys	40 adults with DS	51.3	34.9	67.7	8.56	5.601	<.00001
Adult Mental	41 adults with DS	52.6	41.1	67.6	7.07	6.448	<.00001
Peds	12 children and adolescents with DS	49.5	37.2	58.3	6.32	0.220	.83

Abbreviations: Adult Mental, Global Health Scale v1.2 (Mental); Adult Phys, Global Health Scale v1.2 (Physical); Peds, Peds Scale v1.0; PP, Parent Proxy Scale v1.0 (all ages).

of children ages 5–6, parents of children and adolescents ages 7–17 and parents of adults with DS were 47.4, 48.5 and 45.2, respectively (Table 3). Twelve surveys were received from children and adolescents with DS aged 7–17 years on the Pediatric Global Health 7 + 2; mean T-score was 49.51 (range 37.2–58.3). Forty-five surveys were received from adults with DS on Global Health; mean T-scores on the global mental health scale and the global physical health scales were 52.3 and 51.5, respectively. Scores on the three global health instruments, as reported by parents, children and adolescents and adults with DS, did not differ from the PROMIS® reference population mean ($p > .05$).

3.3 | Paired responses

In some instances, responses from the individual with DS and the accompanying caregiver were returned and completed; paired responses were available in 53 total cases, of which 12 were paired between a Pediatric Global Health 7 + 2 from individuals with DS aged 7–17 years and their parent, and 41 were paired between the adult with DS response on Global Health and their parent (Table 4). Responses from adults with DS, on both Physical and Mental scales, differed from parent responses (t-value = 5.601 and 6.448, respectively, with $p < .001$), such that adults self-report better global health for themselves than their parents report for them. Children and adolescents self-report global health scores that did not differ from those from the parent ($t = 0.220$, $p = .83$).

4 | DISCUSSION

We collected perspectives about overall health from individuals with DS and their parents by using a survey instrument (PROMIS®) that was already validated in the U.S. general population. Our study is unique because we prospectively collected views from individuals with DS and their caregivers using a standardized instrument.

We learned that the PROMIS® global health instruments can be completed by some individuals with DS in clinic. We benchmarked responses from individuals with DS and their caregivers against the PROMIS® reference population. Global health scores in individuals with DS, on all three instruments studied, did not significantly differ from the PROMIS® reference population mean ($p > .05$). Mean T-scores fell within −0.5 to +0.25 SD, which is to say that survey respondents report health that ranges from feeling 0.5 SD less healthy

than the United States general population to feeling 0.25 SD more healthy than the United States general population. DS has a unique profile of medical and developmental concerns. Due to these differences in comorbidities, one might have anticipated that health would be negatively impacted. However, there are also some diseases in those without DS, which are less common in individuals with DS, such as solid tumours (Hasle et al., 2016). Our pilot results suggest that scores from people with DS on these measures might not differ from those of the PROMIS® reference population.

In our sample, global health responses, however, differed by who completed the survey. Comparing paired scores for each individual (i.e., self-report versus parent report) showed differences in global health score, with adults with DS reporting better health than that of their parent proxy report ($p < .001$). This may be explained by differences in perception (self-report versus parent report) of one's current health status, in an individual's definition of health and/or in perception of health in general (e.g., how to interpret the response options of 'Excellent' to 'Poor'). Previous studies show a difference in HRQOL between parents and adolescents with obesity (Bianchini et al., 2013), while the HRQOL among individuals with attention deficit hyperactivity disorder did not differ from parents (Lee et al., 2016). Future study could involve focus groups of individuals with DS and their parents to discuss the definition of health. The difference in score by respondent type is an important consideration in future survey work involving individuals with DS and their parents.

We gathered feedback on any difficulty answering the questions, and limitations of using these measures in individuals with DS were identified. Sixteen of the returned surveys (10%) were excluded because they were incomplete or marked with'?' or 'N/A'. Two items in the Parent Proxy Global Health 7 + 2 survey which had highest non-completion rate were (1) 'How often does your child feel that you listen to his or her ideas?' and (2) 'My child had trouble sleeping when he/she had pain…' Margin notes on these two questions comment on the person with DS's ability to express self or express pain. These two questions may be difficult to answer due to requirement of expression and varying language abilities in people with DS. Similarly on the Pediatric Global Health 7 + 2 survey, individuals with DS most often did not answer, 'How often do your parents listen to your ideas?' On Global Health intended for adults with DS to complete, many notations highlight overall difficulty in understanding questions. In the future, the specific questions with relation to language abilities might be modified to create an adapted Global Health instrument.

Lessons learned in this pilot study can inform further research using these measures in individuals with DS. Limitations of our study include sampling bias as our study was limited to a single clinic population, which limits generalizability to all individuals with DS. We did not have access to standardized cognitive test scores to assess severity of intellectual disability, which may also limit generalizability. Number of responses may also be a limitation. We acknowledge that our small pilot study may not be powered for the analysis of measures in small subgroups; further studies in larger populations of patients with DS are needed. We were also limited by individuals' responses on the paper-based Global Health instruments, which enabled respondents to easily leave a question blank. In future study, an electronic version could require that a response be given to each question. For this study, we intentionally wanted to trial the PROMIS® instrument, as is, so that we could identify possible barriers to completion. We did not directly observe survey completion; it is possible that some instruments intended for individuals with DS were completed by their accompanying caregiver, although their differing responses within dyads would suggest that such possibilities were likely rare, and none of the scores on parent proxy were identical to the self-report score. Another limitation is that our demographic data was collected from a separate, but linked, research registry with its own consent and study protocols. In instances in which our survey participants were not enrolled in that registry, we could not report their demographic information. Future study would be useful to distinguish the role of demographic factors, such as race and gender, on global health scores. In the future, it will be important to build upon the preliminary work in this pilot study with external measures of health, such as completion of health surveillance and comparison to existing measure of health behaviours, to determine if instruments to measure global health are valid, reliable and adequate to capture perceptions of health in Down syndrome.

5 | CONCLUSION

Perceived global health is an overall measure of an individual's reported physical, mental and social health. Validated global health measures were successfully used in individuals with DS and their parents. Limitations were identified for future survey adaptations. Using measures of Global Health, from the PROMIS® reference population, in people with DS continues the trend of medical, academic and social inclusion for DS, as we begin to consider how to define health in DS.

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CONFLICT OF INTEREST

Dr. Skotko occasionally consults on the topic of DS through Gerson Lehrman Group. He receives remuneration from DS non-profit organizations for speaking engagements and associated travel expenses. Dr. Skotko receives annual royalties from Woodbine House, Inc., for the publication of his book, *Fasten Your Seatbelt: A Crash Course on DS for Brothers and Sisters*. Within the past two years, he has received research funding from F. Hoffmann-La Roche, Inc. and LuMind IDSC Down Syndrome Foundation to conduct clinical trials for people with DS. Dr. Skotko is occasionally asked to serve as an expert witness for legal cases where DS is discussed. Dr. Skotko serves in a non-paid capacity on the Honorary Board of Directors for the Massachusetts Down Syndrome Congress, the Board of Directors for the Band of Angels Foundation, and

the Professional Advisory Committee for the National Center for Prenatal and Postnatal Down Syndrome Resources. Dr. Skotko has a sister with Down syndrome. Dr. Santoro receives research funding from the National Institutes of Health. Dr. Santoro receives research funding from the LuMind IDSC Down Syndrome Foundation to conduct clinical trials for people with DS and serves on the Professional Advisory Board for the Massachusetts Down Syndrome Congress. The other authors have no conflicts of interest relevant to this article to disclose.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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