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Research Article

Self-Reported Stuttering Severity Is Accurate: Informing Methods for Large-Scale Data Collection in Stuttering

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ABSTRACT

Purpose: To our knowledge, there are no data examining the agreement between self-reported and clinician-rated stuttering severity. In the era of big data, self-reported ratings have great potential utility for large-scale data collection, where cost and time preclude in-depth assessment by a clinician. Equally, there is increasing emphasis on the need to recognize an individual's experience of their own condition. Here, we examined the agreement between self-reported stuttering severity compared to clinician ratings during a speech assessment. As a secondary objective, we determined whether self-reported stuttering severity correlated with an individual's subjective impact of stuttering.
Method: Speech-language pathologists conducted face-to-face speech assessments with 195 participants (137 males) aged 5–84 years, recruited from a cohort of people with self-reported stuttering. Stuttering severity was rated on a 10-point scale by the participant and by two speech-language pathologists. Participants also completed the Overall Assessment of the Subjective Experience of Stuttering (OASES). Clinician and participant ratings were compared. The association between stuttering severity and the OASES scores was examined.
Results: There was a strong positive correlation between speech-language pathologist and participant-reported ratings of stuttering severity. Participant-reported stuttering severity correlated weakly with the four OASES domains and with the OASES overall impact score.
Conclusions: Participants were able to accurately rate their stuttering severity during a speech assessment using a simple one-item question. This finding indicates that self-report stuttering severity is a suitable method for large-scale data collection. Findings also support the collection of self-report subjective experience data using questionnaires, such as the OASES, which add vital information about the participants' experience of stuttering that is not captured by overt speech severity ratings alone.

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Stuttering is a relatively common communication disorder, characterized by involuntary disruptions to speech fluency (World Health Organization [WHO], 2006) and affecting between 5% and 11% of children by 4 years of age (Andrews & Harris, 1964; Dworzynski et al., 2007; Kefalianos et al., 2017; Reilly et al., 2013; Yairi & Ambrose, 2013). Around 70% of children who begin to stutter will recover, either naturally or with intervention (Franken et al., 2018; Yairi et al., 1996), and lifetime population prevalence of stuttering is estimated at 1% worldwide (Craig & Tran, 2005). Stuttering severity appears to decrease with increasing age (Boyce et al., 2022); however, stuttering can still have a profound effect on quality of life, including reduced educational and vocational attainment (Gerlach et al., 2018; McAllister et al., 2012), increased incidence of bullying and social rejection (Langevin et al., 2009), and increased anxiety-related mental health issues in children and adults (Briley et al., 2021; Iverach et al., 2016, 2009).

Historically, the field of stuttering has been represented by small in-depth clinical studies, which have led to the current state of knowledge. Increasingly, stuttering researchers are striving for larger scale, population-based studies for more representative samples to drive further novel insights into these conditions. As a result, we need to employ new methods, which enable large-scale data collection that is cost- and time-effective.

A succinct measure of stuttering severity is one core need for large-scale studies. There is no standardized approach to measuring stuttering severity; however, it is generally agreed that a speech assessment performed by a trained speech-language pathologist should record the presence, type, and frequency of stuttering behaviors, as well as the psychosocial impact of stuttering (American Speech-Language-Hearing Association, n.d.). Traditionally, percent syllables stuttered (%SS) was widely used to assess stuttering severity and is calculated from a speech sample by comparing stuttered moments to otherwise fluent speech (Johnson, 1955). Severity rating scales are increasingly preferred, as they provide an overall impression of frequency, duration, and severity of both primary and secondary stuttering behaviors. As an example, a block that may last for many seconds may be recognized as a severe moment of disfluency for a person who stutters, the gravitas of which is not adequately represented by a single-syllable rating of disfluency with the %SS rating approach. Furthermore, recent studies have demonstrated that %SS ratings performed by experienced speech-language pathologists do not show adequate agreement and that there is no statistical reason to favor %SS over severity rating scales (O'Brian et al., 2020; Onslow et al., 2018). Severity rating scales also demonstrate acceptable relative reliability for use in research contexts (Karimi et al., 2014).

In clinical settings, severity rating scales, for example, 0–7 (Yairi & Ambrose, 1999) or 0–9 (Onslow et al., 2020), are often used to track clients' progress throughout intervention programs. There are no apparent differences between scales using 5, 7, 9, or 15 points (Cullinan et al., 1963; Curran & Hood, 1977). Such scales are advantageous, as they create a common measure that can be employed by the client in the clinic setting and at home without requiring any special training or equipment. Clinician and client ratings are typically considered concordant if they differ by no more than 1 point on the rating scale (Eve et al., 1995; Hoffman et al., 2014; O'Brian et al., 2004), a system that is a fundamental part of the most widely used treatment program for preschool children who stutter, the Lidcombe Program (Onslow et al., 2020).

In addition to a push for "big data," we now also acknowledge the need for patient experience data. It is important to look beyond the impairment level of severity of stuttering behaviors, to measure the subjective impact of the condition, enabling a full appreciation of the stuttering phenotype that, we know, can have a profound impact on quality of life (Craig et al., 2009; Reeves et al., 2023). Previous research has been inconsistent, with some studies demonstrating evidence of associations between more severe stuttering and larger negative impact of stuttering (Blumgart et al., 2012; Caruso et al., 1994; DiLollo et al., 2003) and others demonstrating conflicting findings (Blumgart et al., 2010; Smith & Kelly, 1997). The Overall Assessment of the Speaker's Experience of Stuttering (OASES; Yaruss & Quesal, 2006) contributes rich information about an individual's stuttering phenotype by providing data pertaining to the negative impact of stuttering across different domains (Yaruss & Quesal, 2006).

Here, we sought to understand whether participants' self-reported stuttering severity ratings correlated with direct clinical speech pathology assessment. We also examined the association between stuttering severity rating and the subjective impact of stuttering as measured with the OASES domains and overall impact score. We hypothesized that stuttering severity would correlate positively with the subjective impact of stuttering.

Method

Participants

This study was nested within a larger international genome-wide association study of stuttering (http://www.geneticsofstutteringstudy.org.au). We invited 1,071 participants from the Australian arm of this cohort with a self-reported history of stuttering described in the work of Boyce et al. (2022) for deep phenotypic characterization.

This study is based on the first 195 participants who consented. Participants were included if they were aged 5 years or older and stuttered currently or had stuttered in the past. The following description of stuttering was provided: "People who stutter have trouble getting their words out. Stuttering is where people repeat sounds over and over (e.g., 'c-c-can I go'), repeat words or syllables over and over (e.g., 'can-can-can I go?'), make prolonged sounds (e.g., 'caaaaaaan I go?'), and have speech 'stoppages' or 'blocks' where no sound comes out." Participants were excluded if they had experienced any acquired neurological disorders, such as traumatic brain injury, before the onset of their stuttering.

The larger international genome-wide association study recruited participants via media campaigns and promotion through support organizations, university departments, and stuttering clinics. Participants self-enrolled through the website (http://www.geneticsofstutteringstudy.org.au), where they provided their consent and completed survey questions (Boyce et al., 2022).

For the present study, participants were sent an e-mail with a link to an online information statement and consent form, inviting them to take part in further surveys and direct speech assessment via videoconference. The human research ethics committee at the Royal Children's Hospital, Melbourne, approved the study (No. 37353).

Data Collection

Health and Medical Survey

As part of their enrollment in the larger study, participants (aged ≥ 18 years) or parents (participants aged < 18 years) answered survey questions about the nature and impact of their or their child's stuttering, including whether they had previously accessed speech pathology intervention.

OASES Survey

The OASES reflects the WHO's International Classification of Functioning, Disability and Health (WHO, 2006). Questions are divided into four domains: (a) general information, (b) reactions to stuttering, (c) communication in daily situations, and (d) quality of life. Questions are rated on a 5-point scale, with higher scores reflecting a more negative impact of stuttering. Participants receive scores for each subscale and for the survey overall. All OASES response forms have been developed and validated within their respective age groups (Yaruss & Quesal, 2006). The OASES has a high degree of test–retest reliability, and normative data have been collected in North America (Yaruss & Quesal, 2006), the Netherlands (Koedoot et al., 2011), and Australia (Blumgart et al., 2012).

In the present study, participants took around 15–20 min to complete the OASES online. Adult participants completed 100 questions from the OASES-A independently. Teenagers (13–17 years) and school-age children (7–12 years) were advised to complete the OASES-T and OASES-S surveys, respectively, in conjunction with a parent or trusted adult. The OASES-T contains 80 questions, and the OASES-S contains 60 questions. Children younger than 7 years did not complete this survey. The OASES response forms were scored in accordance with the test manual guidelines (Yaruss & Quesal, 2006).

Stuttering Severity Ratings

Participants then took part in a telehealth speech assessment with one of six qualified speech-language pathologists over the videoconferencing platform Zoom. The videoconference testing procedure was modified slightly to take account of participant age. Adult and teenage participants (individuals aged ≥ 13 years) were asked about their experience of stuttering and of stuttering interventions that they had received, if applicable. School-age participants (5–12 years) were also asked about access to stuttering intervention if this was indicated and were asked questions about their hobbies and schooling to obtain a conversation sample. Stuttering severity ratings were based on 5 min of conversational speech. Throughout the conversation, the speech-language pathologist took note of participants' stuttering behaviors, including repetitions, prolongations, and blocks, as well as secondary stuttering behaviors, such as movements of the head, trunk, or limbs, yet this information was not used further in the current study. After 5 min of conversation, the speech-language pathologist recorded stuttering severity using the 10-point severity rating scale, where 1 = *no stuttering at all*, 2 = *extremely mild stuttering*, and 10 = *extremely severe stuttering* (Reilly et al., 2009). Without knowing the speech-language pathologist's rating, the participant and/or parent was then asked "On a scale of 1–10, how would you rate your/your child's speech during our conversation today? Where 1 is no stuttering at all, 2 is extremely mild stuttering, and 10 is extremely severe stuttering." A score of 1 on this scale reflects that the participant or parent did not believe that they or their child demonstrated any stuttering during the 5-min conversation. The speech-language pathologist then recorded the participant or parent's response.

Another qualified speech-language pathologist, hereafter referred to as Rater 2, was not involved in the speech assessments and was blinded to the participant and the assessing speech-language pathologist's rating. Rater 2 watched the 5-min conversation samples using video recordings from the videoconference assessments. Rater 2 then provided a stuttering severity rating using the same 10-point scale. Interrater reliability was calculated based on the pairs of ratings made by Rater 2 and the assessing speech-language pathologists.

Statistical Analysis

Paired t tests were used to investigate differences between participant stuttering severity ratings (self-report for adult participants; parent report for participants < 18 years) and those made by Rater 2. We repeated these analyses stratified for (a) whether ratings were self- or parent reported and (b) ratings for participants with and without a history of speech pathology intervention.

Pearson correlations were used to investigate the strength of the association between Rater 2 and participant stuttering severity ratings. Rater 2 was chosen because they rated all samples and were at less risk of bias, as they did not engage with the participant directly.

Pearson correlations were also used to investigate the association between participant stuttering severity ratings and subjective impact of stuttering, as measured by the four OASES domains and the OASES overall impact score. Stuttering severity and OASES scores were both rescaled to values between 0 and 1.

Results

Participant Demographics

One hundred ninety-five participants took part (137 males), aged 5–84 years (M_{age} = 48, SD = 22). There were 23 school-age children (5–12 years), eight teenage children (13–17 years), and 164 adults (≥ 18 years). A demographic summary is provided in Table 1.

Stuttering Severity Ratings

A paired t test indicated no significant difference in ratings of stuttering severity between Rater 2 (M = 2.89, SD = 1.68) and participant or parent self-report (M = 2.80, SD = 1.83) during direct telehealth speech-language pathology assessment, p = .43, Cohen's d = −0.06. There was a strong positive correlation between Rater 2's rating and participant or parent self-report, r = .68, p < .001, and 76% of participant ratings were within 1 point of Rater 2's rating on the 10-point scale, indicating clinical concordance (O'Brian et al., 2004). Paired severity ratings are represented in Figure 1 by age group.

We conducted paired t tests with analysis stratified by whether the participant (aged ≥ 18 years, n = 164) or the parent (participants aged < 18 years, n = 31) performed the rating (see Figure 2a). There was no evidence of a difference between self-rating (M = 2.71, SD = 1.74) and Rater 2's rating (M = 2.81, SD = 1.58), p = .45, Cohen's d = −0.06. There was a strong positive correlation between Rater 2's rating and adult participants' self-ratings, r = .62, p < .001. Similarly, there was no evidence of a difference between parent rating (M = 3.31, SD = 2.23) and Rater 2 rating (M = 3.36, SD = 2.07), p = .82, Cohen's d = −0.04. There was also a strong positive correlation between Rater 2 ratings and parent-performed ratings of child and adolescent participants, r = .85, p < .001.

Finally, we examined whether there was a difference between participant ratings compared to Rater 2's ratings, stratified for those with and without a history of speech-language pathology intervention (see Figure 2b). Here, we were examining whether there was a difference in ratings between those who had experienced therapy and, hence, who may have arguably been better at self-rating. There was no evidence for a difference between ratings made by participants with a history of speech-language pathology intervention (n = 153; M = 2.85, SD = 1.88) and Rater 2 (M = 2.97, SD = 1.72), p = .32, Cohen's d = −0.08. There was a strong positive correlation between this group's ratings and Rater 2's ratings, r = .69, p < .001.

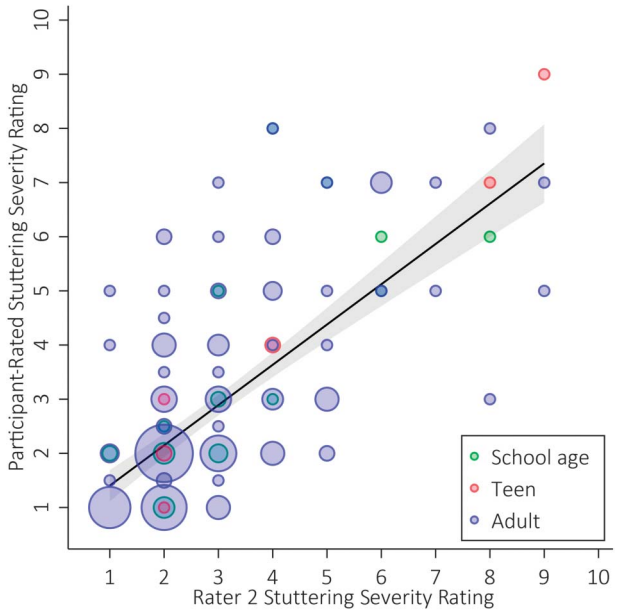
Table 1. Demographic summary of people who stutter, by age, person reporting, and speech-language pathology (SLP) intervention subgroups.

Age group	*n*	Sex		Age in years *M* (*SD*)	SLP intervention *n* (%)	Self- or parent-rated SR *M* (*SD*)[a,b]	SLP-rated SR *M* (*SD*)[a]	Self- or parent-rated SR median (IQR)[a,b]	SLP-rated SR median (IQR)[a]
		Male	Female						
School-age (5–12 years)	23	17	6	9.68 (2.22)	18 (78%)	3.07 (2.05)	3.09 (1.73)	3 (2–5)	3 (2–4)
Teenage (13–17 years)	8	7	1	15.52 (1.70)	8 (100%)	4.00 (2.73)	4.25 (2.87)	3.5 (2–6.25)	3 (2–7.25)
Adult (≥ 18 years)	164	113	51	55.20 (16.89)	127 (77%)	2.71 (1.74)	2.81 (1.58)	2 (1.5–4)	2 (2–3)
Total	195	137	58	48.24 (22.25)	153 (79%)	2.80 (1.83)	2.89 (1.68)	2 (2–4)	2 (2–3)

Note. SR = severity rating; IQR = interquartile range.

[a]Stuttering SR rated on 1–10 scale, where 1 = *no stuttering at all*, 2 = *extremely mild stuttering*, and 10 = *extremely severe stuttering*. [b]Self-rated for adult participants; parent rated for school-age and teenage participants.

Figure 1. Weighted scatter plot between participant and Rater 2's stuttering severity ratings by participant age group, with adult participants (n = 164) in blue, teenage participants (n = 8) in red, and school-age participants (n = 23) in green. Line of best fit represents the correlation between ratings, and gray bands around the line represent the confidence interval.



Similarly, there was no evidence for a difference between ratings made by participants without a history of speech-language pathology intervention (n = 42; M = 2.64, SD = 1.66) and Rater 2 (M = 2.60, SD = 1.48), p = .82, Cohen's d = 0.04. There was also a strong positive correlation between this group's ratings and Rater 2's ratings, r = .63, p < .001.

OASES Survey

Of the 195 participants in the recruited sample, 142 completed the OASES (73%). Twenty-six of those who completed the OASES had never received speech-language pathology intervention (mean overall impact score = 2.26, SD = 0.66), and the remaining 116 had received intervention (mean overall impact score = 2.19, SD = 0.66). OASES data are summarized in Table 2 by domain and age group.

Stuttering Severity Ratings and Subjective Impact of Stuttering

Pearson correlations were used to examine the association between self-rated stuttering severity scores and subjective impact of stuttering scores, as measured by the four OASES domains and the OASES overall impact score. Correlations between stuttering severity rating and scores on all domains of the OASES are summarized in Table 3. There was a weak positive correlation between self-reported stuttering severity rating and overall impact score on the OASES survey, r = .30, p < .001 (see Figure 3).

Interrater Reliability

We compared ratings made by Rater 2 to those made by the six speech-language pathologists who completed

Figure 2. Weighted scatter plots between participant and Rater 2's stuttering severity ratings. (a) Scatter plot stratified by rater, with adult self-reported rating (n = 164) in blue and parent-reported rating (n = 31) in red. (b) Scatter plot stratified by participant history of speech-language pathology intervention, with positive history (n = 153) in red and negative history (n = 42) in green. Line of best fit represents the correlation between ratings, and gray bands around the line represent the confidence intervals. SLP = speech-language pathology.

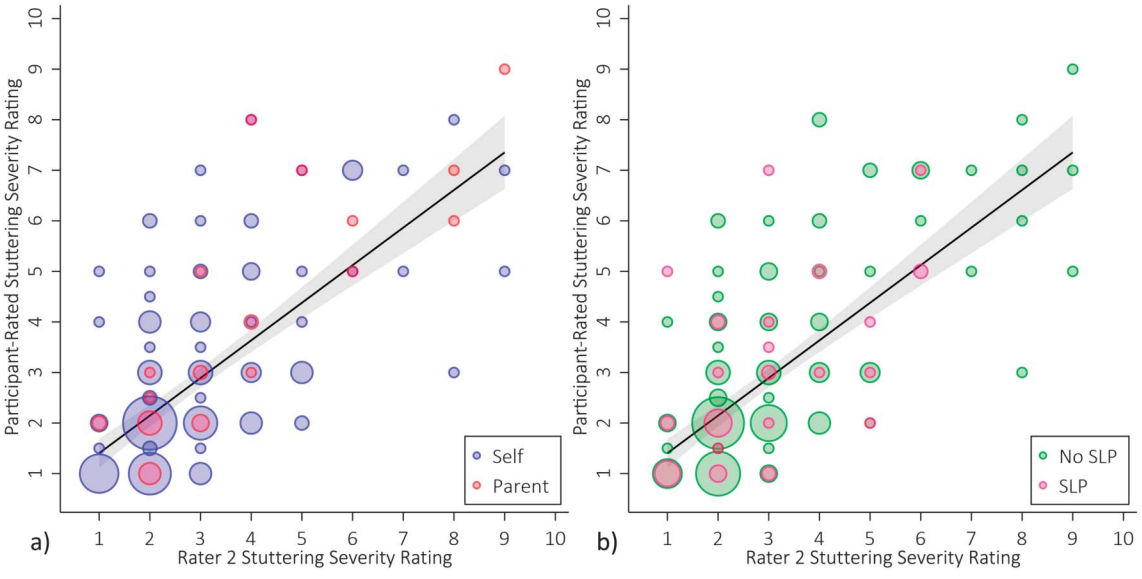


Table 2. Overall Assessment of the Subjective Experience of Stuttering (OASES) data by domain and age group.

	(a) General information			(b) Reactions to stuttering			(c) Communication in daily situations			(d) Quality of life			Overall impact score		
Age	*M* (*SD*)	CI	Impact rating	*M* (*SD*)	CI	Impact rating	*M* (*SD*)	CI	Impact rating	*M* (*SD*)	CI	Impact rating	*M* (*SD*)	CI	Impact rating
School-age (*n* = 17)	2.83 (0.34)	[2.66, 3.01]	Moderate	2.08 (0.77)	[1.69, 2.48]	Mild-to-moderate	1.91 (0.68)	[1.56, 2.26]	Mild-to-moderate	1.53 (0.65)	[1.20, 1.87]	Mild-to-moderate	2.13 (0.55)	[1.85, 2.41]	Mild-to-moderate
Teenage (*n* = 5)	3.07 (0.53)	[2.41, 3.86]	Moderate-to-severe	2.49 (1.17)	[1.03, 3.94]	Moderate	2.39 (1.05)	[1.09, 3.69]	Moderate	2.19 (1.17)	[0.74, 3.64]	Mild-to-moderate	2.50 (0.95)	[1.32, 3.68]	Moderate
Adult (*n* = 120)	2.67 (0.64)	[2.55, 2.78]	Moderate	2.42 (0.75)	[2.28, 2.55]	Moderate	1.98 (0.74)	[1.84, 2.11]	Mild-to-moderate	1.78 (0.81)	[1.63, 1.93]	Mild-to-moderate	2.20 (0.67)	[2.08, 2.32]	Mild-to-moderate
Total (*n* = 142)	2.70 (0.61)	[2.60, 2.80]	Moderate	2.38 (0.77)	[2.25, 2.51]	Moderate	1.98 (0.74)	[1.86, 2.11]	Mild-to-moderate	1.76 (0.81)	[1.63, 1.90]	Mild-to-moderate	2.20 (0.66)	[2.09, 2.31]	Mild-to-moderate

Note. CI = 95% confidence interval of the mean.

Table 3. Pearson correlations between Overall Assessment of the Subjective Experience of Stuttering (OASES) domains and participant stuttering severity rating.

OASES domain	Self- or parent-rated SR (n = 142)		speech-language pathology–rated SR (n = 142)	
	r [CI]	p	r [CI]	p
(a) General information	.19 [.02, .34]	.025*	.35 [.19, .49]	< .001**
(b) Reactions to stuttering	.28 [.12, .43]	.001**	.39 [.24, .52]	< .001**
(c) Communication in daily situations	.29 [.13, .43]	< .001**	.47 [.33, .59]	< .001**
(d) Quality of life	.28 [.12, .42]	.001**	.40 [.26, .54]	< .001**
Overall impact score	.30 [.15, .45]	< .001**	.46 [.32, .58]	< .001**

Note. SR = severity rating; CI = confidence interval.

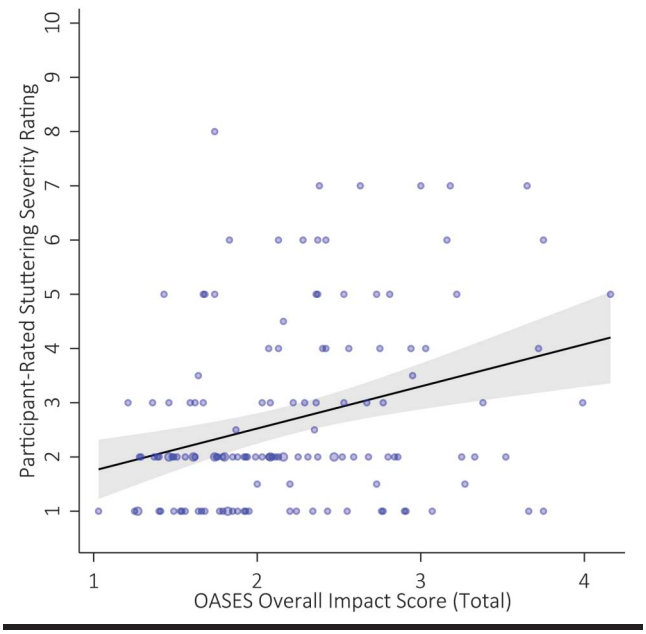
*Correlation is significant at the .05 level (two-tailed). **Correlation is significant at the .01 level (two-tailed).

speech assessments with the participants. There was a strong positive correlation between the two raters, r = .89, p < .001, and 93% of ratings were within 1 point of the paired rating.

Discussion

Through in-depth phenotypic characterization, this study examined the agreement between self-reported stuttering severity (participant or parent report) and clinician ratings. We found no significant difference in ratings performed by qualified speech-language pathologists when compared to participant self-report, and ratings were significantly positively correlated. There was a positive correlation between self-reported stuttering severity and scores on the OASES survey (Yaruss & Quesal, 2006); however, this correlation was weak.

Figure 3. Scatter plot between participant self-rated stuttering severity and OASES overall impact score. Line of best fit represents the correlation between ratings, and gray bands around the line represent the confidence interval. OASES = Overall Assessment of the Subjective Experience of Stuttering



Previous research suggests that speaking about ratings and identifying stuttering events with a client at the beginning of a stuttering treatment program will increase clinical agreement (Ingham & Cordes, 1997a, 1997b; O'Brian et al., 2004). During the speech-language pathology assessment, participants were presented with the 10-point stuttering severity scale (Reilly et al., 2009) but were not engaged in discussion about stuttering types or behaviors. Even so, parents' rating of their child's stuttering and adults' rating of their own stuttering were in agreement with speech-language pathologist ratings. Overall, 76% of participant-reported ratings were within 1 point of the speech-language pathologist's rating, which is indicative of concordant ratings in clinical intervention programs (O'Brian et al., 2004; Onslow et al., 2020) and also close to 93% concordance seen between speech-language pathologists' ratings. Participant ratings agreed with speech-language pathologist ratings, regardless of presence or absence of past speech-language pathology intervention, indicating that participants and parents were able to apply the rating scale appropriately, even if they had not rated stuttering severity previously. These findings support the accuracy of stuttering self-report for large-scale data collection in that participants do not need training before being able to apply the rating in practice.

In the present study, we expected to find a positive correlation between self-reported stuttering severity and overall impact score on the OASES (Yaruss & Quesal, 2006). Previous research has been inconsistent, with some studies demonstrating evidence of associations between more severe stuttering and larger negative impact of stuttering (Blumgart et al., 2012; Caruso et al., 1994; DiLollo et al., 2003) and others demonstrating conflicting findings (Blumgart et al., 2010; Smith & Kelly, 1997). We hypothesized that the more overtly disrupted somebody's speech

is, the more barriers they face across their personal and professional lives, and the bigger the impact on their quality of life overall. We did find a relationship in the expected direction; however, the correlation was weak for each of the four OASES domains and for the OASES overall impact score. This finding is in line with Ward et al. (2021), who identified a gap between behavioral and social measures of stuttering in commonly used stuttering assessment tools. This also suggests that the subjective impact of stuttering is influenced by other factors beyond stuttering severity that were not assessed in this study and could be further explored in future research.

Compared to Australian adult OASES normative data (n = 200) presented by Blumgart et al. (2012), our group of 120 adults scored higher on general information and lower on reaction to stuttering, communication in daily situations, and quality of life, resulting in a lower overall impact score (M = 2.20, SD = 0.66). Comparatively, Blumgart et al. (2012) demonstrated a mean of 2.66 (SD = 0.65) in their sample. This could be partly explained by demographic differences, with 43% of our adult participants being greater than 60 years, compared to their 21.5% greater than 60 years. Blumgart et al. (2012) also found a weak positive association between more severe stuttering (> 4%SS) and scores on general information. Interestingly, in our group, general information was the only domain not significantly correlated to stuttering severity rating at the 0.01 level. This could be due to methodological differences of using a severity rating scale, which takes into account secondary stuttering behaviors, rather than %SS, which only takes into account proportion of stuttered speech.

As in other areas of the world, stuttering treatment in Australia is shifting toward a more disfluency-affirming approach (Reeves et al., 2023). A recent investigation by Lowe et al. (2021) raised some important points around how traditional speech restructuring interventions may induce or increase speech-related anxiety, with potential for harm. Our data showed evidence of a correlation between overt stuttering severity and subjective impact of stuttering; however, the correlation was weak. As part of a comprehensive description of the stuttering phenotype, it is important to capture the subjective experience of stuttering, as well as measuring overt stuttering severity. Furthermore, it is important to capture the direct views and experiences of individuals with the condition of interest. A limitation of the current study is that our younger age groups are small (n = 23 and n = 8 for school-age and teenage groups, respectively). In the future, it would be useful to consider parent-reported and/or self-reported stuttering severity ratings in a larger sample of school-age and teenage children. It would also be useful to investigate the correlation between these ratings and subjective impact of stuttering as a factor of age group and person rating. Here, we focused on severity ratings without any additional training on stuttering behaviors. Further work could examine whether agreement is even higher for severity self-report when factoring in types of stuttering behaviors.

Conclusions

Overall, our findings support the use of self-report in large cohort studies of people who stutter, where a speech assessment conducted by a speech-language pathologist is not typically feasible. Our findings also support the use of self-report measures, such as the OASES, which can provide insight into the subjective impact of stuttering. The subjective impact of stuttering is not captured by symptom-based speech severity ratings; however, it contributes importantly to holistic phenotypic characterization in a complex human condition such as stuttering.

Data Availability Statement

Data are available on request due to privacy and ethical restrictions.

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References

American Speech-Language-Hearing Association. (n.d.). *Fluency disorders* [Practice portal]. Retrieved January 4, 2023, from http://www.asha.org/practice-portal/clinical-topics/fluency-disorders/

Andrews, G., & Harris, M. (1964). *The syndrome of stuttering*. Heinemann.

Blumgart, E., Tran, Y., & Craig, A. (2010). Social anxiety disorder in adults who stutter. *Depression and Anxiety, 27*(7), 687–692. https://doi.org/10.1002/da.20657

Blumgart, E., Tran, Y., Yaruss, J. S., & Craig, A. (2012). Australian normative data for the Overall Assessment of the Speaker's Experience of Stuttering. *Journal of Fluency Disorders, 37*(2), 83–90. https://doi.org/10.1016/j.jfludis.2011.12.002

Boyce, J. O., Jackson, V. E., Van Reyk, O., Parker, R., Vogel, A. P., Eising, E., Horton, S. E., Gillespie, N. A., Scheffer, I. E., Amor, D. J., Hildebrand, M. S., Fisher, S. E., Martin, N. G., Reilly, S., Bahlo, M., & Morgan, A. T. (2022). Self-reported impact of developmental stuttering across the lifespan. *Developmental Medicine & Child Neurology, 64*(10), 1297–1306. https://doi.org/10.1111/dmcn.15211

Briley, P. M., Gerlach, H., & Jacobs, M. M. (2021). Relationships between stuttering, depression, and suicidal ideation in young adults: Accounting for gender differences. *Journal of Fluency Disorders, 67,* Article 105820. https://doi.org/10.1016/j.jfludis.2020.105820

Caruso, A. J., Chodzko-Zajko, W. J., Bidinger, D. A., & Sommers, R. K. (1994). Adults who stutter: Responses to cognitive stress. *Journal of Speech and Hearing Research, 37*(4), 746–754. https://doi.org/10.1044/jshr.3704.746

Craig, A., Blumgart, E., & Tran, Y. (2009). The impact of stuttering on the quality of life in adults who stutter. *Journal of Fluency Disorders, 34*(2), 61–71. https://doi.org/10.1016/j.jfludis.2009.05.002

Craig, A., & Tran, Y. (2005). The epidemiology of stuttering: The need for reliable estimates of prevalence and anxiety levels over the lifespan. *Advances in Speech Language Pathology, 7*(1), 41–46. https://doi.org/10.1080/14417040500055060

Cullinan, W. L., Prather, E. M., & Williams, D. E. (1963). Comparison of procedures for scaling severity of stuttering. *Journal of Speech and Hearing Research, 6*(2), 187–194. https://doi.org/10.1044/jshr.0602.187

Curran, M. F., & Hood, S. B. (1977). Listener ratings of severity for specific disfluency types in children. *Journal of Fluency Disorders, 2*(2), 87–97. https://doi.org/10.1016/0094-730X(77)90012-2

DiLollo, A., Manning, W. H., & Neimeyer, R. A. (2003). Cognitive anxiety as a function of speaker role for fluent speakers and persons who stutter. *Journal of Fluency Disorders, 28*(3), 167–186. https://doi.org/10.1016/S0094-730X(03)00043-3

Dworzynski, K., Remington, A., Rijsdijk, F., Howell, P., & Plomin, R. (2007). Genetic etiology in cases of recovered and persistent stuttering in an unselected, longitudinal sample of young twins. *American Journal of Speech-Language Pathology, 16*(2), 169–178. https://doi.org/10.1044/1058-0360(2007/021)

Eve, C. L., Onslow, M., Andrews, C., & Adams, R. (1995). Clinical measurement of early stuttering severity: The reliability of a 10-point scale. *Australian Journal of Human Communication Disorders, 23*(2), 26–39. https://doi.org/10.3109/asl2.1995.23.issue-2.03

Franken, M.-C. J. P., Koenraads, S. P. C., Holtmaat, C. E. M., & van der Schroeff, M. P. (2018). Recovery from stuttering in preschool-age children: 9 year outcomes in a clinical population. *Journal of Fluency Disorders, 58,* 35–46. https://doi.org/10.1016/j.jfludis.2018.09.003

Gerlach, H., Totty, E., Subramanian, A., & Zebrowski, P. (2018). Stuttering and labor market outcomes in the United States. *Journal of Speech, Language, and Hearing Research, 61*(7), 1649–1663. https://doi.org/10.1044/2018_JSLHR-S-17-0353

Hoffman, L., Wilson, L., Copley, A., Hewat, S., & Lim, V. (2014). The reliability of a severity rating scale to measure stuttering in an unfamiliar language. *International Journal of Speech-Language Pathology, 16*(3), 317–326. https://doi.org/10.3109/17549507.2014.898097

Ingham, R. J., & Cordes, A. K. (1997a). Identifying the authoritative judgments of stuttering: Comparisons of self-judgments and observer judgments. *Journal of Speech, Language, and Hearing Research, 40*(3), 581–594. https://search.ebscohost.com/login.aspx?direct=true&AuthType=sso&db=eric&AN=EJ558194&site=eds-live&scope=site&custid=s2775460

Ingham, R. J., & Cordes, A. K. (1997b). Self-measurement and evaluating stuttering treatment efficacy. In R. F. Curlee & G. M. Siegel (Eds.), *Nature and treatment of stuttering: New directions* (pp. 413–437). Allyn & Bacon.

Iverach, L., Jones, M., McLellan, L. F., Lyneham, H. J., Menzies, R. G., Onslow, M., & Rapee, R. M. (2016). Prevalence of anxiety disorders among children who stutter. *Journal of Fluency Disorders, 49,* 13–28. https://doi.org/10.1016/j.jfludis.2016.07.002

Iverach, L., Jones, M., O'Brian, S., Block, S., Lincoln, M., Harrison, E., Hewat, S., Cream, A., Menzies, R. G., Packman, A., & Onslow, M. (2009). The relationship between mental health disorders and treatment outcomes among adults who stutter. *Journal of Fluency Disorders, 34*(1), 29–43. https://doi.org/10.1016/j.jfludis.2009.02.002

Johnson, W. (1955). The time, the place, and the problem. In *Stuttering in children and adults: Thirty years of research at the University of Iowa* (pp. 3–24). University of Minnesota Press.

Karimi, H., O'Brian, S., Onslow, M., & Jones, M. (2014). Absolute and relative reliability of percentage of syllables stuttered and severity rating scales. *Journal of Speech, Language, and Hearing Research, 57*(4), 1284–1295. https://doi.org/10.1044/2014_JSLHR-S-13-0106

Kefalianos, E., Onslow, M., Packman, A., Vogel, A., Pezic, A., Mensah, F., Conway, L., Bavin, E., Block, S., & Reilly, S. (2017). The history of stuttering by 7 years of age: Follow-up of a prospective community cohort. *Journal of Speech, Language, and Hearing Research, 60*(10), 2828–2839. https://doi.org/10.1044/2017_JSLHR-S-16-0205

Koedoot, C., Versteegh, M., & Yaruss, J. S. (2011). Psychometric evaluation of the Dutch translation of the Overall Assessment of the Speaker's Experience of Stuttering for Adults (OASES-A-D). *Journal of Fluency Disorders, 36*(3), 222–230. https://doi.org/10.1016/j.jfludis.2011.03.002

Langevin, M., Packman, A., & Onslow, M. (2009). Peer responses to stuttering in the preschool setting. *American Journal of Speech-Language Pathology, 18*(3), 264–276. https://doi.org/10.1044/1058-0360(2009/07-0087)

Lowe, R., Menzies, R., Onslow, M., Packman, A., & O'Brian, S. (2021). Speech and anxiety management with persistent stuttering: Current status and essential research. *Journal of Speech, Language, and Hearing Research, 64*(1), 59–74. https://doi.org/10.1044/2020_JSLHR-20-00144

McAllister, J., Collier, J., & Shepstone, L. (2012). The impact of adolescent stuttering on educational and employment outcomes: Evidence from a birth cohort study. *Journal of Fluency Disorders, 37*(2), 106–121. https://doi.org/10.1016/j.jfludis.2012.01.002

O'Brian, S., Heard, R., Onslow, M., Packman, A., Lowe, R., & Menzies, R. G. (2020). Clinical trials of adult stuttering treatment: Comparison of percentage syllables stuttered with self-reported stuttering severity as primary outcomes. *Journal of*

Speech, Language, and Hearing Research, 63(5), 1387–1394. https://doi.org/10.1044/2020_JSLHR-19-00142

O'Brian, S., Packman, A., & Onslow, M. (2004). Self-rating of stuttering severity as a clinical tool. *American Journal of Speech-Language Pathology, 13*(3), 219–226. https://doi.org/10.1044/1058-0360(2004/023)

Onslow, M., Jones, M., O'Brian, S., Packman, A., Menzies, R., Lowe, R., Arnott, S., Bridgman, K., de Sonneville, C., & Franken, M.-C. (2018). Comparison of percentage of syllables stuttered with parent-reported severity ratings as a primary outcome measure in clinical trials of early stuttering treatment. *Journal of Speech, Language, and Hearing Research, 61*(4), 811–819. https://doi.org/10.1044/2017_JSLHR-S-16-0448

Onslow, M., Webber, M., Harrison, E., Arnott, S., Bridgman, K., Carey, B., Sheedy, S., O'Brian, S., MacMillan, V., & Lloyd, W. (2020). The Lidcombe Program treatment guide. *Lidcombe Program Trainers Consortium*.

Reeves, N. A., Flynn, T. W., & Schuff, R. Z. (2023). Ableism to empowerment: Navigating school structures when working with students who stutter. *Language, Speech, and Hearing Services in Schools, 54*(1), 8–26. https://doi.org/10.1044/2022_LSHSS-22-00026

Reilly, S., Onslow, M., Packman, A., Cini, E., Conway, L., Ukoumunne, O. C., Bavin, E. L., Prior, M., Eadie, P., Block, S., & Wake, M. (2013). Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics, 132*(3), 460–467. https://doi.org/10.1542/peds.2012-3067

Reilly, S., Onslow, M., Packman, A., Wake, M., Bavin, E. L., Prior, M., Eadie, P., Cini, E., Bolzonello, C., & Ukoumunne, O. C. (2009). Predicting stuttering onset by the age of 3 years: A prospective, community cohort study. *Pediatrics, 123*(1), 270–277. https://doi.org/10.1542/peds.2007-3219

Smith, A., & Kelly, E. (1997). Stuttering: A dynamic, multifactorial model. *Nature and Treatment of Stuttering: New directions, 2,* 204–217.

Ward, D., Miller, R., & Nikolaev, A. (2021). Evaluating three stuttering assessments through network analysis, random forests and cluster analysis. *Journal of Fluency Disorders, 67,* Article 105823. https://doi.org/10.1016/j.jfludis.2020.105823

World Health Organization. (2006). *The world health report 2006: Working together for health*. https://doi.org/10.30875/5b142cc2-en

Yairi, E., & Ambrose, N. (2013). Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders, 38*(2), 66–87. https://doi.org/10.1016/j.jfludis.2012.11.002

Yairi, E., & Ambrose, N. G. (1999). Early childhood stuttering I: Persistency and recovery rates. *Journal of Speech, Language, and Hearing Research, 42*(5), 1097–1112. https://doi.org/10.1044/jslhr.4205.1097

Yairi, E., Ambrose, N. G., Paden, E. P., & Throneburg, R. N. (1996). Predictive factors of persistence and recovery: Pathways of childhood stuttering. *Journal of Communication Disorders, 29*(1), 51–77. https://doi.org/10.1016/0021-9924(95)00051-8

Yaruss, J. S., & Quesal, R. W. (2006). Overall Assessment of the Speaker's Experience of Stuttering (OASES): Documenting multiple outcomes in stuttering treatment. *Journal of Fluency Disorders, 31*(2), 90–115. https://doi.org/10.1016/j.jfludis.2006.02.002