ASHA American Speech-Language-Hearing Association

JSLHR

Research Article

Stuttering Behavior in a National Age Cohort of Norwegian First-Graders With Down Syndrome

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ARTICLE INFO

Article History:
Received November 15, 2021
Revision received March 25, 2022
Accepted July 25, 2022

Editor-in-Chief: Bharath Chandrasekaran
Editor: Julie D. Anderson

https://doi.org/10.1044/2022_JSLHR-21-00605

ABSTRACT

Purpose: The aims of this study were to investigate the occurrence of stuttering behavior across time and to evaluate the relationship between stuttering behavior and language ability in children with Down syndrome.
Method: A national age cohort of Norwegian first graders with Down syndrome (*N* = 75) participated in the study. Speech samples from a story-retelling task and a picture book dialogue as well as standardized measures of vocabulary, grammar, and nonverbal mental ability were collected at two time points approximately 5 months apart. Stuttering behavior was evaluated through counting stuttering-like disfluencies and stuttering severity ratings. The relationship between stuttering behavior and language ability was investigated through hierarchical regression analysis.
Results: The participants had stuttering severity ratings ranging from no stuttering behavior to severe and displayed all types of stuttering-like disfluencies. There were significant relationships between stuttering behavior and language ability at the first time point, whereas the relationships were not significant at the second time point. The stuttering severity ratings were significantly predicted by language ability across time, whereas the frequency of stuttering-like disfluencies was not.
Conclusions: The occurrence of stuttering behavior was high across the measures and time points; however, the relationship between stuttering behavior and language ability varied across these variables. Thus, the nature of the relationship does not seem to follow a strict pattern that can be generalized to all children across time.

It has been suggested that stuttering is highly common in individuals with Down syndrome. A key question has been whether the speech behaviors observed in these individuals actually represent stuttering behavior or other types of disfluencies (see review by Kent & Vorperian, 2013). The aims of this study were to investigate the occurrence of stuttering behavior across time and to assess the relationship between stuttering behavior and language ability in first-graders with Down syndrome. Observable characteristics of speech are commonly used to identify stuttering in both clinical practice and research (Eggers & van Eerdenbrugh, 2018; Van Zaalen-Op't Hof et al., 2009). Speech that contains repetitions of sounds, syllables, and monosyllabic words (particularly in young children) as well as prolongations of sounds and blocks is generally considered to reflect stuttering (also called "stuttering-like" disfluencies). In contrast, speech that contains interjections, revisions, and multisyllabic whole-word and phrase repetitions is considered to reflect nonstuttered (or "other") disfluencies (see, e.g., Bloodstein et al., 2021; Yaruss, 1997a).

Stuttering behavior may be challenging to identify in children with Down syndrome. This is due to language

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disorder associated with this syndrome (Næss et al., 2015, 2021), showing impaired sentence production (syntax; Andreou & Chartomatsidou, 2020; Frizelle et al., 2019), as well as the presence of atypical speech characteristics, such as extended silences, a slow or rapid speech rate, and differences related to stress, pitch, loudness, or breathing (Jones et al., 2019; Loveall et al., 2021). As such, children with Down syndrome may speak in an effortful manner. This in turn can influence the validity of an assessment, the distinction of stuttering-like disfluencies (SLDs) from other disfluencies (ODs), and the identification of other characteristics of disordered speech production. Nevertheless, stuttering behavior is important to assess and identify in children with Down syndrome, because it may negatively impact their speech intelligibility and overall communication ability (Evans, 1977; Maessen et al., 2022). These added difficulties associated with stuttering behavior can further hinder this group's ability to express themselves effectively; behavioral, emotional, and social functioning can also be adversely affected (Briley et al., 2019; Jackson et al., 2014).

Previous Research on Stuttering in Individuals With Down Syndrome

In a national health inquiry, Schieve et al. (2009) reported that children and adolescents with Down syndrome were at a greater risk of stuttering than both typically developing individuals and individuals with an intellectual disability not caused by Down syndrome. In a review of the literature, Kent and Vorperian (2013) found that estimates of the co-occurrence of Down syndrome and stuttering in previous studies ranged from 10% to 45%. They noted that precise criteria for identifying stuttering have generally not been provided in the literature, making it difficult to separate SLDs from ODs. Furthermore, the methods used to identify stuttering in previous studies has varied, including medical record reviews (Gottsleben, 1955), parental surveys (Kumin, 1994), and speech sample analyses (Devenny & Silverman, 1990; Preus, 1972). Notably, Preus (1972) found that a greater proportion of the participants in his study of stuttering in individuals with Down syndrome were identified as "individuals who stutter" from caregiver assessments conducted on a rating scale rather than assessments based on the frequency of SLDs. Thus, the variability in these prior findings may reflect differences in how stuttering was defined and operationalized as well as differences in the methods that were used to identify stuttering behavior. Also, many tudies on stuttering in individuals with Down syndrome are older (see, e.g., Evans, 1977; Gottsleben, 1955; Preus, 1972), suggesting that it is necessary to revisit the topic due to theoretical and methodological advances within both the field of stuttering and the field of Down syndrome.

None of the studies included in the review of Kent and Vorperian (2013) focused on children, and the research on stuttering exclusively in children with Down syndrome appears to be limited. In a more recent study however, Eggers and van Eerdenbrugh (2018) presented findings from a study of 26 children (age 3.3–12.6 years) with Down syndrome. They identified eight (31%) participants as "children who stutter" based on their having a frequency of three or more SLDs per 100 syllables in a single speech sample. Although their sample did exclusively include children, the age range was broad, with 9.03 years between the oldest and youngest participant. Age is an important predictor of the frequency of disfluencies in typically developing children (Tumanova et al., 2014). Thus, age-spread samples may cause variability in the study's results. Potential age effects and sampling bias may be controlled for by systematically recruiting narrow age cohorts of children with Down syndrome. Moreover, previous studies of stuttering in individuals with Down syndrome have investigated stuttering behavior at only one time point and in one speaking situation (Eggers & van Eerdenbrugh, 2018; Kent & Vorperian, 2013). As stuttering behavior varies across time and situations (Tichenor & Yaruss, 2021; Yaruss, 1997b), investigating stuttering longitudinally and in different speaking contexts represents a gap in the research literature that needs to be addressed to ensure the reliability of related findings.

The Relationship Between Stuttering Behavior and Language Ability

The language development of children with Down syndrome is generally different compared to that of typically developing children at the same nonverbal mental ability level (Næss, 2016, Næss et al., 2011). These children often exhibit pervasive language disorders that (a) lag behind their cognitive development (Zampini & D'Odorico, 2013) and (b) cannot be explained by their intellectual disability alone (Frizelle et al., 2019). Not all aspects of language appear to be equally affected, with reliable differences between these different aspects (Evans, 1977; Vicari et al., 2000). While receptive vocabulary is often described as a relative strength in children with Down syndrome, expressive vocabulary and grammar are generally described as areas of weaknesses (Loveall et al., 2019; Næss et al., 2011). Their syntactical abilities appear to be particularly affected, evident in difficulties with both syntax comprehension and production (Andreou & Chartomatsidou, 2020). As a group, children with Down syndrome also show substantial variability in terms of language ability, including differences in reaching linguistic milestones, such as the production of first words, the combination of words into utterances, and the use of grammatical markers. By the late preschool/early school–age years (age 4–6 years), most children with Down syndrome can combine words into multiword utterances, but some still mainly communicate via one-word utterances or have very limited spoken language ability (Berglund et al., 2001).

In typically developing children, the onset of stuttering usually occurs at approximately age 3 years (Reilly et al., 2009; Yairi, 2004). Because this is a time of rapid lexical and syntactical development (Owens, 2012; Smith & Weber, 2017), research has examined potential associations between stuttering and language. The majority of this research—involving otherwise typically developing children who stutter—has confirmed a disadvantage in terms of language development and abilities in children who stutter versus children who do not stutter (see review by Brundage & Bernstein Ratner, 2022). Studies have found significant differences between these two aforementioned groups in terms of general language level (Ntourou et al., 2011; Zaretsky et al., 2017) as well as vocabulary (expressive and receptive; Luckman et al., 2020; Ntourou et al., 2011) and, more specifically, grammar (Ntourou et al., 2011; Zaretsky et al., 2017). Others have suggested that stuttering is associated with a dissociation between different areas of language ability (Anderson & Conture, 2000).

Stuttering behavior generally first occurs when children are in the early stages of grammatical development (Bloodstein, 2006). For children with Down syndrome, the transition from one-word to multiword utterances generally happens much later than it does in typically developing children (Berglund et al., 2001). This is not only due to a general delay in language development but also a difference in the synchrony of developmental milestones and an asymmetry in the growth of different aspects of language. For example, it is hypothesized that children with Down syndrome need a larger vocabulary size than typically developing children before they start combining words (Lewis, 2003). Studies have also shown that these children, after beginning to use multiword utterances, still produce more one-word utterances and fewer multiword utterances (both simple and complex sentences) than typically developing children when matched for developmental age (Zampini & D'Odorico, 2011). Thus, it is not surprising that children with Down syndrome display the onset of stuttering or stuttering-like behavior at a later point in their development than typically developing children. Considering the specific language profile of children with Down syndrome, investigating the relationship between stuttering behavior and language ability—and, more specifically, vocabulary and grammar ability—may provide valuable information about stuttering behavior in this group. Thus far, only one study has investigated the stuttering–language association in adults with Down syndrome, finding that stuttering was associated with better vocabulary skills (Devenny & Silverman, 1990); however, such associations have not yet been investigated in children with Down syndrome. Næss et al. (2021) recently published findings from parental reports on difficulties with fluency (not isolated to stuttering behavior) in their children with Down syndrome. They found that the children with the weakest language abilities were also those reported to have greater difficulties with fluency. Due to the possible mix in the reporting of language functioning and fluency, the authors noted that these parent reports of elevated disfluency should be confirmed through further studies directly investigating speech fluency.

This study seeks to address gaps in the existing literature by examining stuttering behavior and the relationship between stuttering behavior and language ability in a systematically recruited sample of children with Down syndrome in the same school year (first grade) in two speaking situations and at two different points in time (Time Point 1 = T1; Time Point 2 = T2). The following three research questions are addressed.

1. What is the occurrence of stuttering behavior among the participants based on the percentage of SLDs and stuttering severity ratings (SSRs)?
2. What specific types of disfluencies are identified?
3. Is there a significant relationship between language ability and stuttering behavior as measured by (a) the percentage of SLDs and (b) SSRs?

Research has indicated that typically developing children who stutter may have a disadvantage in language ability (Luckman et al., 2020; Ntourou et al., 2011; Zaretsky et al., 2017). Some have also suggested a dissociation between different aspects of language in this group of children (Anderson & Conture, 2000). In individuals with Down syndrome, research has indicated a high occurrence of stuttering behavior (see, e.g., Eggers & van Eerdenbrugh, 2018; Preus, 1972), a language profile of lower levels of language ability compared to typically developing children on the same nonverbal mental age level (see, e.g., Næss et al., 2011), and reliable intraindividual differences between language domains (see, e.g., Vicari et al., 2000). We therefore hypothesize that there is a concurrent relationship between stuttering behavior and language ability in children with Down syndrome at T1 and T2, respectively.

Unlike the typical population showing a low occurrence of stuttering in school-aged children and adults compared with preschool-aged children (Yairi & Ambrose, 2013), stuttering behavior in individuals with Down syndrome seems to be consistently high across the lifespan (see, e.g., Eggers & van Eerdenbrugh, 2018; Preus, 1972). This forms the basis for an exploratory hypothesis that language ability at T1 predicts stuttering behavior at T2 in children with Down syndrome when controlling for stuttering behavior at T1.

The concurrent analysis provides information about the strength of the association between language ability and stuttering behavior at T1 and T2, respectively. The longitudinal analysis predicts the importance of language ability to the residual change in stuttering behavior between T1 and T2. This provides added information about the relationship between language ability and stuttering behavior as it puts a direction on the relationship between the variables (VanderWeele et al., 2020). Also, by controlling for prior stuttering status, any variability in the outcome (stuttering behavior at T2) caused by stuttering at T1 will be set aside, leaving only variability that is unexplained by stuttering at T1. Thus the results provide the unique explanatory value of language abilities to stuttering behavior in the longitudinal model (see, e.g., Tabachnick & Fidell, 2018). The results of this study provide novel information about the potential relationships between stuttering and Down syndrome to support a greater understanding of whether and how stuttering should be addressed in the assessment and treatment of this population.

Method

Participants

The study participants were an age cohort of 75 Norwegian first-graders with Down syndrome drawn from the original sample (N = 104) of the digital vocabulary intervention, the Down Syndrome LanguagePlus project (DSL+; Næss et al., 2022). The original sample was systematically recruited through the national habilitation services. Thus, this study's sample included participants from all parts of Norway—encompassing both urban and rural areas. All participants spoke Norwegian as their first language. No predefined inclusion criteria were set with regard to language ability level; however, because the co-occurrence of Down syndrome and autism spectrum disorders (ASD) is high compared to that that seen in the general population and this dual diagnosis has implications for communication abilities (Versaci et al., 2021), children with a known diagnosis of ASD were not eligible for participation.

Inclusion in this specific substudy was based on the availability of speech samples from the DSL+ intervention (see the section on data collection below); thus, all participants in this study received the DSL+ vocabulary intervention. Of the original participant pool (N = 104), individuals were excluded due to having no spoken language at the time of assessment (n = 8), attrition (n = 6), or missing data (n = 15). The final sample was comprised of 35 girls and 40 boys, and the mean age of the participants at the start of the study was 80.02 months (SD = 5.84, min–max = 68.38–94.68 months). Ethical approval was granted by the Norwegian Centre for Research Data; informed parental consent was obtained for all children, and consent was also obtained from the participating schools and practitioners (teachers, special education teachers, and teacher assistants) involved in the project.

Data Collection and Measures

We collected audio speech samples from all study participants to assess stuttering behavior; from these samples, we identified and categorized disfluencies and rated the stuttering severity. Standardized measures of vocabulary and grammar were used to assess language ability. Additionally, we used a standardized measure of nonverbal mental ability. All measurements were done at two time points (T1 and T2) approximately five months apart. Each measure is presented in detail below.

Audio Speech Samples

As stuttering shows notable situational variability (Tichenor & Yaruss, 2021; Yaruss, 1997b), the speech samples were collected from two different speaking situations—a picture book dialogue with a teacher and a story-retelling task with an assessor—from the DSL+ project.

The picture book dialogue was drawn from a teaching situation in which a child interacts with their teacher or teaching assistant (TA; Næss et al., 2022). The DSL+ intervention aimed to increase the number of words the children know and how well they know the words. In this intervention, children participated in daily (five days a week) digital picture book activities over the course of 15 weeks. Each picture book was repeated every day for 1 week, with increasing cognitive difficulty levels of dialogue as the week progressed (Days 1–3 were conducted one-on-one with the teacher/TA; Days 4–5 were conducted with the teacher/TA and peers). In this study, we selected the first and last available one-on-one picture book dialogues for each child.

The story-retelling test (Bus Story Test; Renfrew, 2010), originally designed to measure narrative skills in 3- to 8-year-old children, is part of the clinical measures collected before and after the DSL+ intervention. In this task, the assessor first tells the story about "a bus that ran away"; then, the child is asked to retell the story. The retelling is supported by a textless story book with 12 color pictures. This test has been used in several studies of speech disfluency in typically developing children (see, e.g., Van Zaalen-op't Hof et al., 2009), and it has been recommended as an appropriate addition to a conversational context in the assessment of stuttering (Byrd et al., 2012).

Identification and categorization of speech disfluencies. First, the speech samples were transcribed by research assistants and checked by the first author. Instances of disagreement were discussed and resolved jointly after relistening to the speech sample. Next, the transcriptions were coded for (a) SLDs, including repetition of sounds, syllables, or monosyllabic words, prolongations of sounds, and blocks; and (b) other disfluencies (ODs), including repetition of multisyllabic words and phrases, interjections, and revisions (see, e.g., Bloodstein et al., 2021; Yaruss, 1997a; see Appendix A for a thorough description of the coding system with examples). The number of these different types of disfluencies as well as the number of total syllables produced were tallied per study participant to calculate the percentage of SLDs (100 × [total number of SLDs/total number of syllables] = %SLD) and ODs (100 × [total number of ODs/total number of syllables] = %OD). In situations where two or more separate disfluencies appeared together (e.g., a repetition followed by a prolongation) within the same phrase (e.g., "th-th-th-the dog aaaate it") or word (e.g., "th-th-th-theeeee dog ate it"), each disfluency type was counted (Ingham & Ingham, 2011). Clusters of two SLDs in one single word were relatively rare, occurring in 5.59% of words at T1 and 3.45% of words at T2. Syllables were counted based on the child's pronunciation of the word (e.g., "bana" for "banana" equals two syllables). Interjections and yes/no words were included, whereas unintelligible words (i.e., instances where the meaning of the word could not be understood), sounds, vocalizations, and onomatopoeias were excluded.

Speech disfluencies were coded by the first author. A randomly selected 20% of the speech samples were recoded by the fourth author. The interrater reliability was 93.29% (SLD = 89.07%, OD = 97.52%) at T1 and 94.25% (SLD = 92.86%, OD = 95.64%) at T2. Any disagreements were resolved through consensus, with the coders listening to the samples again and discussing their judgments.

SSR. We used the Stuttering Severity Rating Scale (Onslow et al., 2020) to evaluate stuttering severity. It is a 10-point scale with scores ranging from 0 to 9, where 0 represents *no stuttering*, 1 reflects *extremely mild stuttering*, and 9 reflects *extremely severe stuttering* (no other points on the scale are labeled). This evaluation method is perceptually based and conveys how the *listener* perceives the severity of the stuttering, including both the frequency of the stuttering as well as any accompanying struggle behavior, such as audible tension.

The speech samples were divided between the fourth and fifth authors—both speech-language pathologists (SLPs) specialized in speech fluency disorders—who independently rated the stuttering severity. A randomly selected 20% (n = 32) of the speech samples were double-coded. In accordance with O'Brian et al. (2004, 2011), scores that were within one scale point of each other were judged to indicate agreement between the raters' judgments. At T1, 93.75% of the scores (30 pairs of a total of 32 pairs) were within one scale point of each other. Of the remaining scores, one pair differed by 2 points and the other differed by 4 points. At T2, 90.63% of the scores (29 of a total of 32 pairs) were within one scale point of each other. The remaining three pairs differed by 2 points. Any rating disagreement between the two listeners was solved by reaching consensus through discussion after relistening to the recordings.

Standardized Measures of Language and Nonverbal Mental Ability

In order to investigate the relationship between stuttering and language ability, we included measures of expressive vocabulary (Wechsler Preschool and Primary Scale of Intelligence–Third Edition [WIPPSI-III]; Picture Naming; Wechsler, 2002), receptive vocabulary (British Picture Vocabulary Scale-II; Dunn et al., 1997, Norwegian translation by Lyster et al., 2010), expressive grammar (Illinois Test of Psycholinguistic abilities [ITPA]; Grammatic Closure, Kirk et al., 1967, Norwegian translation by Gjessing & Nygaard, 1975), and receptive grammar (Test of Reception of Grammar–Revised; Bishop, 2003, Norwegian translation by Lyster & Horn, 2009). In order to control for nonverbal mental ability in our regression models, we also administered the Block Design items of the WIPPSI-III (Wechsler, 2002). See Appendix B for supplementary information on all the standardized measures.

All standardized measures were part of a larger assessment battery carried out in two or more sessions at each time point. In order to accommodate children's possible challenges related to sustained attention and task perseverance, the examiners allowed for breaks and playtime between tests. The assessment was conducted by members of the DSL+ team and research assistants who had received training and certification prior to conducting the assessment.

Data Analysis

Statistical analysis was conducted using IBM SPSS Statistics Version 27, and the significance level was set to .05 (two-tailed). As participants were preselected based on the existence of speech samples (at both time points), there was no missing data for the stuttering behavior scores. However, for the Bus Story Test, four children at T1 and one child at T2 did not produce any intelligible utterances. In these instances, only their respective scores from the picture book dialogue were used. There were missing data at both time points for the language and the nonverbal mental ability variables. Overall, there were missing values for 24% of the cases. At T1, there were five cases with one missing value; in total, 1.33% of the data involved missing values. At T2, there were 13 cases with missing values; in total, 4.27% of the data involved missing values. The result from Little's missing completely at random test (chi-square (60) = 78.30, sig. = .056) indicated that the missing data were distributed randomly. Multiple imputation is generally considered a good approach to handling missing data, as it avoids excluding any data. However, SPSS does not produce all the necessary pooled results from the hierarchical regression analysis based on the multiply imputed data. The missing data were therefore handled by listwise deletion (Anani et al., 2017), and the results were confirmed by running the analysis on a multiply imputed data set (five imputations), which yielded comparable results. The data were winsorized to handle extreme values; we modified the data points using the mean ± 3 *SD*s of the variable as cutoff points. All statistical analyses were based on these winsorized values.

All findings were averaged across the two speech samples at each time point. First, we calculated the relative frequency (or proportions) of the disfluency types (based on the raw scores), the mean percentage of each disfluency type (based on the winsorized scores), and the percentage of participants displaying each disfluency type at each time point out of the entire sample. Next, we calculated the percentage of SLDs and ODs for each participant (100 × [total number of disfluent syllables/total number of syllables]). We created composite scores of the two speech samples at each time point for the %SLD, SSR, and speech sample size variables. As is common for measures of stuttering (Jones et al., 2006; O'Brian et al., 2004), the dependent variables were not normally distributed for either the %SLD variable (T1, skewness = 1.21 [*SE* = 0.28], kurtosis = 1.20 [*SE* = 0.55]; T2, (skewness = 1.69 [*SE* = 0.28], kurtosis of 2.90 [*SE* = 0.55]) or the SSR variable (T1, skewness = 1.45 [*SE* = 0.28], kurtosis = 1.38 [*SE* = 0.55]; T2 skewness = 1.11 [*SE* = 0.28], kurtosis = 0.41 [*SE* = 0.55]). Due to indications of collinearity between the language measures, we created two composite scores: one for vocabulary based on the BPVS (receptive vocabulary) and Picture Naming (expressive vocabulary) tests and one for grammar based on the TROG (receptive grammar) and ITPA (expressive grammar) tests.

A %SLD at or above 3% (see, e.g., Ambrose & Yairi, 1999; Boey et al., 2007; Natke et al., 2006) as well as an SSR above 0 (Onslow et al., 2020) are often considered indicative of a diagnosis of stuttering. According to the International Classification of Diseases 11th revision (ICD-11) (World Health Organization, 2019), stuttering is a speech fluency disorder that, in addition to being characterized by interruptions of the normal flow of speech, also results in significant impairment in functioning across areas such as social communication and academic achievement. Because this study examined observable characteristics only, we did not attempt to diagnose stuttering in our participants. Also, all statistical analyses were based on data from the entire sample (N = 75). However, for descriptive and comparison purposes, we report findings from the grouping of participants into subsamples based on whether or not they had (a) a frequency of SLDs above 0%, (b) a frequency of SLDs at or above 3%, or (c) a severity rating above 0 points.

Paired-samples t tests were conducted to determine if there were significant differences between the time points in terms of (a) the mean percentage of each disfluency type, (b) the mean %SLD and mean %OD, (c) the mean SSR, (d) language ability, (e) nonverbal mental ability, and (f) the speech sample size. A correlation analysis was performed among all the included variables. Hierarchical regression analysis was performed to determine whether there was a relationship between language ability and stuttering behavior at each time point (concurrent analysis) and whether language ability at T1 predicted stuttering behavior at T2 (longitudinal analysis) beyond the stuttering behavior at T1. Due to differences between the two stuttering behavior measures, %SLD and SSR were investigated in separate models. Because children with Down syndrome show dissociations between vocabulary and grammar abilities (see, e.g., Vicari et al., 2000), we treated vocabulary and grammar separately in the hierarchical regression models. Due to the sequence of vocabulary and grammar development in young children (Kristoffersen et al., 2012), vocabulary was included before grammar in the hierarchical regression models. Overall, we constructed six models: four with concurrent data and two with longitudinal data. As expected, based on the considerable variability in the language abilities of children with Down syndrome (Karmiloff-Smith et al., 2016), the amount of intelligible speech produced by the participants varied. Additionally, nonverbal mental ability is also expected to vary within a narrow age cohort of children with Down syndrome (Karmiloff-Smith et al., 2016). Therefore, we controlled for speech sample size (number of syllables) and nonverbal mental ability in our regression models. Based on visual inspection, the assumptions of homoscedasticity and normally distributed residuals (linearity) were fulfilled for all models. There were no indications of collinearity.

Results

Table 1 shows the descriptive disfluency data at each time point as well as the effect sizes for the differences in the mean percentage of the disfluency types between T1 and T2. According to the relative frequencies, prolongation was the most frequent type of SLD, whereas interjection was the most frequent type of OD. There were no

Table 1. Relative frequency of disfluency types (percentages), mean percentage of disfluency types, and the percentage of participants displaying the different disfluency types at Time Point 1 (T1) and Time Point 2 (T2).

	T1			T2			Effect size[d]	
Category	**Relative freq.[a]**	***M* (*SD*)[b]**	**Participants[c]**	**Relative freq.[a]**	***M* (*SD*)[b]**	**Participants[c]**	***d***	***p***
Sound rep.	4.75	0.28 (0.52)	30.67	4.07	0.28 (0.53)	31.58	0.733	.964
Syllable rep.	5.76	0.43 (0.65)	42.67	5.58	0.32 (0.60)	28.95	0.837	.266
Monosyllabic word rep.	6.22	0.52 (0.94)	40.00	5.81	0.30 (0.60)	31.58	1.114	.093
Prolongation	32.43	3.40 (4.09)	77.33	34.38	2.37 (3.25)	61.84	4.463	.051
Block	9.27	0.65 (1.11)	41.33	7.78	0.50 (0.89)	38.16	0.950	.191
Total SLD	58.43	5.65 (5.37)		57.61	4.42 (5.14)		6.310	.096
Multisyllabic word rep.	1.24	0.05 (0.13)	14.67	1.74	0.10 (0.27)	17.11	0.277	.102
Phrase rep.	1.58	0.04 (0.16)	8.00	1.28	0.07 (0.23)	9.21	0.275	.454
Interjection	35.59	3.56 (3.92)	85.33	35.77	3.13 (3.45)	81.58	4.644	.426
Revision	3.16	0.18 (0.37)	26.67	3.60	0.18 (0.42)	25.00	0.493	.916
Total OD	41.57	3.91 (3.90)		42.39	3.68 (3.73)		4.802	.687

Note. freq. = frequency; *d* = Cohen's *d*; rep. = repetition; SLD = stuttering-like disfluency; OD = other disfluency.

[a]Relative frequencies (or proportions) are based on average raw scores from the picture book dialogue and bus story retelling task. [b]Mean percentage of disfluency types are based on adjusted scores (winsorized). [c]Percentage of participants who display each disfluency type is based on dichotomization of whether or not the child displays the disfluency type. [d]Effect size (Cohens's *d*) calculated based on means and standard deviations adjusted for outliers by winsorization.

significant differences between time points in either the mean percentage of the different disfluency types or the total SLD and total OD. Thus, the distribution of disfluency types was highly similar across the time points.

Table 2 shows group data regarding the frequency of stuttering behavior at T1 and T2, including the means and standard deviations for the full sample and subsamples (%SLD above 0%, %SLD at or above 3%, and SSR above 0 points). There was no significant difference in terms of either mean %SLD or mean SSR for the full sample between the time points. The majority of the participants displayed one or more SLD (i.e., %SLD above 0%). Also, a high number of participants exhibited a %SLD of 3 or above at both time points. Half of the participants received an SSR rating above 0 (i.e., they were judged to display at least extremely mild stuttering behavior).

Table 3 shows the descriptive statistics for vocabulary and grammar ability, nonverbal mental ability, and speech sample size at T1 and T2. The internal consistency assessment revealed good reliability (based on Cronbach's alpha) for all standardized measures, with an alpha value ranging from .82 to .93. Table 4 show the results from the paired-samples *t* test regarding the differences in the measures between the time points. Even though there was an increase in the mean score for all measures, the difference between the time points was significant for receptive vocabulary and expressive grammar only.

Table 5 shows the correlations among the study variables. The correlations among the stuttering behavior measures were moderate to weak, indicating variability in the identification of stuttering behavior across time and measures. As expected, the correlations among the language measures were strong. Furthermore, the correlations

Table 2. Stuttering behavior at Time Point 1 (T1) and Time Point 2 (T2) based on percentage of stuttering-like disfluencies and stuttering severity ratings.

	T1				T2				Effect size	
Variable	***n* (%)**	***M* (adj. *M*)**	***SD* (adj. *SD*)**	**Min–max**	***n* (%)**	***M* (adj. *M*)**	***SD* (adj. *SD*)**	**Min–max**	***d***	***p***
Full sample										
%SLD	75 (100)	5.86 (5.65)	6.04 (5.37)	0–29.37	75 (100)	4.62 (4.42)	5.83 (5.14)	0–27.89	6.310	.096
SSR	75 (100)	1.17 (1.15)	1.65 (1.63)	0–6.50	75 (100)	1.31 (1.29)	1.67 (1.63)	0–7.50	1.50	.423
Subsamples										
%SLD > 0	64 (85.33)	6.86 (6.66)	5.98 (5.38)	0.58–29.37	57 (76.00)	6.08 (5.76)	5.99 (5.21)	0.35–27.89		
%SLD ≥ 3	42 (56)	9.50 (9.20)	5.83 (5.00)	3.22–29.37	35 (46.67)	8.89 (8.37)	6.14 (5.12)	3.27–27.89		
SSR > 0	38 (50.67)	2.30 (2.28)	1.67 (1.60)	0.5–6.50	38 (50.67)	2.58 (2.55)	1.49 (1.41)	0.5–7.50		

Note. Means and standard deviations are presented based on raw scores with adjusted (winsorized) values in brackets. adj. = adjusted; %SLD = percentage stuttering-like disfluencies; SSR = stuttering severity rating. %SLD > 0 = results for participants with a %SLD above 0%–100%, %SLD ≥ 3 = results for participants with a %SLD above 3%–100%, SSR > 0 = results for participants with an SSR score above 0–9.

Table 3. Descriptive statistics on language ability, nonverbal mental ability, and speech sample size at Time Point 1 (T1) and Time Point 2 (T2).

Variable (max. score)	T1					T2				
	n	α	*M*	*SD*	Min–max	*n*	α	*M*	*SD*	Min–max
Vocabulary expressive (38)[a]	75	.92	9.29	6.59	0–24	74	.93	9.73	6.92	0–25
Vocabulary receptive (144)[a]	75	.93	24.85	11.47	3–55	73	.92	30.22	11.55	4–64
Grammar expressive (33)[a]	75	.85	1.73	3.00	0–15	73	.85	2.15	3.27	0–13
Grammar receptive (80)[a]	72	.83	8.76	5.27	0–28	71	.84	9.48	5.86	2–29
Nonverbal ability mental (40)[a]	73	.82	13.10	5.70	0–26	68	.84	13.98	5.99	0–28
Speech sample size[b]	75		78.16	78.46	4–561	75		77.72	52.80	6.5–213.50

Note. α = Cronbach's alpha.

[a]Raw scores. [b]Speech sample size variables are based on the average number of syllables produced across the two speech samples.

among the language measures and the speech sample size variables were moderate to strong—that is, children with higher scores on the standardized language measures also produced more speech in the two speaking situations. There were no significant correlations between %SLD and speech sample size, vocabulary, grammar, or nonverbal mental ability. There were significant, moderate to weak correlations between SSR and speech sample size and vocabulary.

To address our third research question, which examines whether there was a significant relationship between stuttering behavior (as measured by %SLD or SSR) and language ability (based on vocabulary and grammar), we estimated six hierarchical regression models. First, we tested the concurrent relationship between language ability and %SLD. Table 6 contains the standardized regression coefficients β along with the confidence intervals (95%), change R^2 (ΔR^2), and *p* values of the regression models for the relationship between language ability and %SLD at T1 and T2, controlling for speech sample size and nonverbal mental ability. At T1, in accordance with our hypothesis, the independent variables significantly predicted %SLD, $F(4, 65) = 4.361$, $p = .003$: The total variance explained (R^2) was 21.2%. Vocabulary accounted for 7.5% of the variance in the %SLD variable ($p = .021$), whereas grammar accounted for 10.7% of the variance in the %SLD variable ($p = .004$). At T2, contrary to our hypothesis, the independent variables did not significantly predict %SLD, $F(4, 57) = 0.778$, $p = .544$: The total variance explained (R^2) was 5.2%.

Second, we tested the concurrent relationship between language ability and SSR. Table 7 contains the standardized regression coefficients β along with the confidence intervals (95%), change R^2 (ΔR^2), and *p* values of the regression models of the relationship between language ability and SSR at T1 and T2, controlling for speech sample size and nonverbal mental ability. At T1, in accordance with our hypothesis, the independent variables significantly predicted SSR, $F(4, 65) = 8.459$, $p < .001$: The total variance explained (R^2) was 34.2%. Vocabulary accounted for 6.3% of the variance in the SSR variable ($p = .019$), whereas grammar accounted for 5.7% of the variance in the SSR variable ($p = .020$). At T2, the independent variables significantly predicted SSR, $F(4, 57) = 4.088$, $p = .006$: The total variance explained (R^2) was 22.3%. However, contrary to our hypothesis, language ability did not significantly contribute to the explained variance in SSR at T2.

Third, we tested whether language ability at T1 could predict stuttering behavior at T2 when controlling for stuttering behavior at T1. Table 8 contains the

Table 4. Results from paired-samples *t* test of the difference in language ability, nonverbal mental ability, and speech sample size between Time Point 1 (T1) and Time Point (T2).

Variable	T1		T2		Effect size		
	Adj. *M*	Adj. *SD*	Adj. *M*	Adj. *SD*	*N*	*d*	*p*
Vocabulary expressive	9.29	6.59	9.73	6.92	74	2.942	.225
Vocabulary receptive	24.85	11.47	30.22	11.55	73	9.865	**< .001**
Grammar expressive	1.67	2.74	2.12	3.19	73	1.787	**.046**
Grammar receptive	8.70	5.09	9.45	5.78	68	3.952	.154
Nonverbal mental ability	13.10	5.70	13.98	5.99	66	4.826	.119
Speech sample size	74.24	58.77	77.72	52.80	75	47.125	.231

Note. Significant values ($p < .05$) are shown in bold. Effect sizes (Cohen's *d*) are based on means and standard deviations adjusted for outliers by winsorization. Adj. = adjusted.

Table 5. Pearson correlations among the study variables.

Variable	1	2	3	4	5	6	7	8	9	10	11
1. % SLD T1											
2. % SLD T2	.266*										
3. SSR T1	.238*	.095									
4. SSR T2	.217	.310**	.561**								
5. Speech sample size T1	−.046	−.069	.433**	.330**							
6. Speech sample size T2	.096	.091	.337**	.510**	.627**						
7. Vocabulary T1	.129	.070	.353**	.457**	.511**	.620**					
8. Vocabulary T2	.049	.008	.301*	.385**	.475**	.640**	.850**				
9. Grammar T1	−.170	−.046	.088	.180	.428**	.455**	.748**	.744**			
10. Grammar T2	−.132	−.053	.053	.269*	.531**	.490**	.656**	.739**	.824**		
11. Nonverbal mental age T1	−.146	−.129	−.086	.148	.289*	.335**	.443**	.455**	.502**	.379**	
12. Nonverbal mental age T2	−.153	−.091	−.108	.188	.301**	.397**	.433**	.476**	.397**	.470**	.681**

Note. Correlation coefficients are based on values adjusted for outliers by winsorization. %SLD = percentage of stuttering-like disfluencies; T1 = Time Point 1; T2 = Time Point 2; SSR = stuttering severity rating.
*$p < .05$ **$p < .01$.

standardized regression coefficients β along with the confidence intervals (95%), change in R^2 (ΔR^2), and p values reflecting the longitudinal relationship between language ability at T1 and %SLD at T2, controlling for %SLD at T1, speech sample size at T2, and nonverbal mental ability at T1. Contrary to our hypothesis, the independent variables did not significantly predict %SLD at T2, $F(5, 64) = 1.885$, $p = .109$: The total variance explained (R^2) was 12.8%. Table 9 contains the standardized regression coefficients β along with the confidence intervals (95%), change in R^2 (ΔR^2), and p values reflecting the longitudinal relationship between language ability at T1 and SSR at T2, controlling for SSR at T1, speech sample size at T2, and nonverbal mental ability at T1. In accordance with our hypothesis, the independent variables significantly predicted the change between T1 and T2 in SSR, $F(5, 64) = 11.541$, $p < .001$: The total variance explained (R^2) was 47.4%. Vocabulary accounted for 1.1% of the variance in the SSR variable ($p = .270$), whereas grammar accounted for 4.1% of the variance in the SSR variable ($p = .030$).

Discussion

Overall, we found a high occurrence of stuttering behavior in our study group of children with Down syndrome as measured by %SLD and SSR. The participants displayed a variety of disfluency types, including SLDs and ODs. SLDs were identified in more than 75% of the children's speech samples, and half of the children were judged to have some degree of stuttering behavior based on their respective SSR score. There was a significant concurrent relationship between language ability and stuttering behavior at T1, but not at T2. Language

Table 6. Summary of hierarchical regression models of the relationship between language ability and percentage of stuttering-like disfluencies at Time Point 1 (T1) and Time Point 2 (T2), controlling for speech sample size and nonverbal mental ability.

	T1					T2				
Variable	β	95% CI of *b*	R^2	ΔR^2	*p*	β	95% CI of *b*	R^2	ΔR^2	*p*
Step 1			.005	.005	.554			.005	.005	.595
Speech sample size	−.072	[−1.715, 0.928]			.554	.069	[−0.824, 1.425]			.595
Step 2			.029	.024	.202			.008	.003	.670
Speech sample size	−.028	[−1.519, 1.216]			.826	.095	[−0.837, 1.666]			.510
Nonverbal mental ability	−.161	[−0.386, 0.083]			.202	−.061	[−0.246, 0.160]			.670
Step 3			**.104**	**.075**	**.021**			.023	.016	.340
Speech sample size	−.167	[−2.390, 0.558]			.219	−.005	[−1.568, 1.525]			.978
Nonverbal mental ability	−.270	[−0.497, −0.011]			.041	−.094	[−0.276, 0.142]			.525
Vocabulary	.339	[0.278, 3.374]			.021	.172	[−0.856, 2.441]			.340
Step 4			**.212**	**.107**	**.004**			.052	.028	.197
Speech sample size	−.148	[−2.204, 0.588]			.252	.007	[−1.510, 1.569]			.969
Nonverbal mental ability	−.159	[−0.390, 0.091]			.220	−.046	[−0.247, 0.181]			.761
Vocabulary	.668	[1.711, 5.485]			< .001	.329	[−0.463, 3.505]			.130
Grammar	−.518	[−4.699, −0.922]			.004	−.255	[−2.821, 0.594]			.197

Note. Values adjusted for outliers by winsorization. Significant values ($p < .05$) are shown in bold. CI = confidence interval.

Table 7. Summary of hierarchical regression models of the relationship between language ability and stuttering severity rating at Time Point 1 (T1) and Time Point 2 (T2), controlling for speech sample size and nonverbal mental ability.

Variable	T1					T2				
	β	95% CI of *b*	R^2	ΔR^2	*p*	β	95% CI of *b*	R^2	ΔR^2	*p*
Step 1			**.175**	**.175**	**< .001**			**.189**	**.189**	**< .001**
Speech sample size	.419	[0.199, 0.639]			< .001	.435	[0.206, 0.680]			< .001
Step 2			**.223**	**.047**	**.048**			.189	.000	.929
Speech sample size	.481	[0.258, 0.705]			< .001	.430	[0.174, 0.702]			.002
Nonverbal mental ability	−.226	[−0.077, 0.000]			.048	.012	[−0.041, 0.045]			.929
Step 3			**.285**	**.063**	**.019**			.223	.033	.120
Speech sample size	.354	[0.113, 0.595]			.005	.284	[−0.033, 0.611]			.077
Nonverbal mental ability	−.326	[−0.096, −0.016]			.007	−.037	[−0.050, 0.037]			.782
Vocabulary	.309	[0.051, 0.557]			.019	.251	[−0.072, 0.614]			.120
Step 4			**.302**	**.057**	**.020**			.223	.000	.954
Speech sample size	.368	[0.135, 0.602]			.002	.284	[−0.036, 0.615]			.080
Nonverbal mental ability	−.244	[−0.082, −0.002]			.041	−.035	[−0.051, 0.040]			.801
Vocabulary	.550	[0.226, 0.857]			.001	.258	[−0.141, 0.697]			.190
Grammar	−.378	[−0.691, −0.060]			.020	−.010	[−0.371, 0.350]			.954

Note. Values adjusted for outliers by winsorization. Significant values ($p < .05$) are shown in bold. CI = confidence interval.

ability significantly predicted SSR, but not %SLD, at T2 when controlling for the same stuttering behavior at T1.

The Occurrence of Stuttering Behavior in Children With Down Syndrome

This study's findings regarding higher occurrences of stuttering behavior in this population than usually found in typically developing children (see, e.g., Reilly et al., 2009) are consistent with existing research on stuttering in Down syndrome (Eggers & van Eerdenbrugh, 2018; Kent & Vorperian, 2013). In the general population, the prevalence of stuttering is suggested to be higher in younger children than in older children, teenagers, and adults. This is due to the fact that many young children who stutter ultimately stop stuttering—either on their own or following treatment (Yairi & Ambrose, 2013). In this study, we examined children aged 6–7 years. Although subject to variability, as a group, children with Down syndrome in this age range generally exhibit nonverbal mental abilities

Table 8. Summary of hierarchical regression model of the relationship between language ability at Time Point 1 (T1) and percentage of syllables stuttered at Time Point 2 (T2), controlling for percentage of syllables stuttered at T1, speech sample size at T2, and nonverbal mental ability at T1.

Variable	%SLD				
	β	95% CI of *b*	R^2	ΔR^2	*p*
Step 1			**.088**	**.088**	**.012**
Stuttering T1	.297	[0.062, 0.491]			.012
Step 2			.104	.016	.277
Stuttering T1	.288	[0.052, 0.482]			.016
Speech sample size T2	.127	[−0.529, 1.815]			.277
Step 3			.117	.013	.330
Stuttering T1	.264	[0.025, 0.465]			.029
Speech sample size T2	.168	[−0.396, 2.096]			.178
Nonverbal mental ability T1	−.122	[−0.324, 0.110]			.330
Step 4			.122	.005	.565
Stuttering T1	.250	[0.007, 0.458]			.043
Speech sample size T2	.121	[−0.885, 2.110]			.417
Nonverbal mental ability T1	−.150	[−0.364, 0.103]			.268
Vocabulary T1	.092	[−1.127, 2.047]			.565
Step 5			.128	.007	.492
Stuttering T1	.219	[−0.038, 0.445]			.098
Speech sample size T2	.115	[−0.922, 2.090]			.441
Nonverbal mental ability T1	−.128	[−0.352, 0.128]			.355
Vocabulary T1	.192	[−1.194, 3.119]			.376
Grammar T1	−.136	[−2.672, 1.299]			.492

Note. Values adjusted for outliers by winsorization. Significant values ($p < .05$) are shown in bold. %SLD = percentage of stuttering-like disfluencies. Values adjusted for outliers by winsorization. CI = confidence interval.

Table 9. Summary of hierarchical regression model of the relationship between language ability at Time Point 1 (T1) and stuttering severity rating at Time Point 2 (T2), controlling for stuttering severity rating at T1, speech sample size at T2, and nonverbal mental ability at T1.

	SSR				
Variable	β	95% CI of *b*	R^2	ΔR^2	*p*
Step 1			**.283**	**.283**	**< .001**
SSR T1	.532	[0.323, 0.727]			< .001
Step 2			**.417**	**.133**	**< .001**
Stuttering T1	.409	[0.210, 0.597]			< .001
Speech sample size T2	.385	[0.186, 0.572]			< .001
Step 3			.423	.006	.409
SSR T1	.428	[0.223, 0.621]			< .001
Speech sample size T2	.353	[0.138, 0.555]			.001
Nonverbal mental ability T1	.084	[−0.020, 0.048]			.409
Step 4			.433	.011	.270
SSR T1	.394	[0.181, 0.596]			< .001
Speech sample size T2	.289	[0.047, 0.521]			.020
Nonverbal mental ability T1	.038	[−0.030, 0.043]			.728
Vocabulary T1	.145	[−0.112, 0.395]			.270
Step 5			**.474**	**.041**	**.030**
SSR T1	.337	[0.124, 0.540]			.002
Speech sample size T2	.285	[0.050, 0.511]			.018
Nonverbal mental ability T1	.093	[−0.021, 0.053]			.396
Vocabulary T1	.389	[0.053, 0.703]			.024
Grammar T1	−.329	[−0.611, −0.033]			.030

Note. Values adjusted for outliers by winsorization. Significant values ($p < .05$) are shown in bold. SSR = stuttering severity rating. CI = confidence interval.

and receptive language abilities that are approximately on par with that of typically developing 3-year-old children (see the work of Næss, 2012). As the onset of stuttering in typically developing children generally happens around the age of 3 years, coinciding with a time of rapid semantic and syntactic growth (Yairi, 1983, 2004), it may be that these first graders are at an age where they are especially likely to exhibit SLDs and ODs.

The distributions of the different disfluency types were highly similar across the time points. A clear majority of the participants presented with prolongations and interjections. We also saw that, based on the relative frequencies, prolongation was the most frequently observed SLD, whereas interjection was the most frequently observed OD. The reporting of prolongations and interjections as common in children with Down syndrome is consistent with previous studies of speech characteristics in this group (see, e.g., Eggers & van Eerdenbrugh, 2018; Jones et al., 2019; Kumin, 2006; Willcox, 1988). In contrast, for young, otherwise typically developing children who stutter, repetitions are described as the most frequent SLD (Ambrose & Yairi, 1999). Some have questioned whether SLDs in individuals with Down syndrome represent genuine stuttering behavior or "something else" (see a review by Van Borsel & Tetnowski, 2007). Willcox (1988) did, for example, argue that there were qualitative differences in the prolongations and part-word repetitions in children with Down syndrome, separating them from SLDs. Although she did not specify what these qualitative differences were, we recognize some ambiguity related to the identification of SLDs in this group of children, as audible struggle may not always be present in prolongations and repetitions. As such, there may be differences in children with Down syndrome versus otherwise typically developing children who stutter—both in regard to the frequency of disfluency types as well as the qualitative characteristics (e.g., rate and tension) of the disfluencies. According to Ratner (2015), disfluencies may reflect difficulties with language formulation rather than stuttering. Given that children with Down syndrome have pronounced difficulties with verbal processing (see a review by Grieco et al., 2015), some disfluencies may simply reflect a need for more time to plan an utterance, retrieve vocabulary, or repair articulation errors. Separating "typical" SLDs from more "atypical" (or ambiguous) SLDs in future research could provide more insight into these qualitative differences that can occur, especially for prolongations and repetitions.

Even though the occurrence of stuttering behavior in general was high in the current study, there was individual variability across the time points and measures. Variability across time is a common characteristic of stuttering (Reilly et al., 2009; Tichenor & Yaruss, 2021). Thus, children may have a varying frequency and/or severity of stuttering behavior, or they may have started (or stopped) exhibiting stuttering behavior between the time points. Alternatively, they might not have displayed stuttering behavior at the specific time when the speech sample was collected (Tichenor & Yaruss, 2021; Yairi & Ambrose, 2013). Furthermore, the frequencies revealed from the SLD measure did not consistently correspond with those of the SSR measure and vice versa. The stuttering behavior measures in this study were chosen because they have been commonly used in previous research on stuttering in typically developing children (O'Brian et al., 2004) and children with Down syndrome (Eggers & van Eerdenbrugh, 2018). While the %SLD measure is generally considered to be relatively objective, as it is based on counting the number of observable SLDs, the SSR measure is designed to account for more subjective judgments, such as the degree of struggle as perceived by the listener. Potential reasons for the differing results between the %SLD and SSR measures may be related to (a) quantitative evaluation versus qualitative evaluation, (b) intelligible speech only versus all available speech (intelligible and unintelligible speech included), and (c) the categorical evaluation of single words and utterances versus the global evaluation of stuttering behavior. It was not within the scope of this study to compare the applicability of these measures. However, it is important to be aware of the differences in the results they produce as well as their respective benefits and limitations.

The Relationship Between Stuttering Behavior and Language Ability

The results from the hierarchical regression analysis are somewhat ambigous with regard to the relationship between language ability and stuttering behavior. First, the hypothesis that there is a significant concurrent relationship between language ability and stuttering behavior in children with Down syndrome at both time points was partly supported, as this relationship was only significant at T1. The significant concurrent relationship between language ability and stuttering behavior at T1 is in line with previous research on typically developing children who stutter (Brundage & Bernstein Ratner, 2022). Thus, we find that the relationship between language ability and stuttering behavior is not stable across time points.

Second, the hypothesis that language ability at the initial time point predicts stuttering behavior at the later time point was supported for the SSR measure but not for the %SLD measure. In this study, we found that there were inconsistencies between children's SSR scores and %SLD; the rank order of the children with regard to degree of stuttering behavior depends on the measure used. Studies of stuttering in otherwise typically developing children who stutter have also found such inconsistencies between SSR scores and %SLD (Onslow et al., 2018). The inconsistencies between the SSR measure and the %SLD measure may be related to distinct differences in how stuttering is operationalized in these two measures (O'Brian et al., 2004; Onslow et al., 2018). Whereas the %SLD measure only includes one quantitative aspect of stuttering, the SSR measure reflects both quantitative and qualitative aspects of stuttering behavior. Thus, the SSR considers the more general influence of stuttering behavior on communication. Such differences have also been pointed to in earlier research by O'Brian et al. (2004), who identified two distinct types of stuttering that could lead to inconsistencies between SSR and %SLD. First, high percentages of prolongations and blocks relative to repetitions were seen in participants with low %SLD scores and high SSR scores. Second, high percentages of repetitions relative to prolongations and blocks were seen in participants with high %SLD scores and low SSR scores. It may be that one of these profiles are more common in children with Down syndrome, contributing to the inconsistencies between the two stuttering measures. This is not investigated in the current study, and should therefore be investigated in future research. Interestingly, we note that, even though not all the models yielded significant findings, the pattern across all the models showed positive relationships between stuttering behavior and vocabulary ability and a negative relationship between stuttering behavior and grammar ability. Thus, stuttering behavior seemingly increases with increased vocabulary. This finding is consistent with results reported by Devenny and Silverman (1990), who used the Peabody Picture Vocabulary Test, which is a test similar to the BPVS used in this study, to show that stuttering was associated with better vocabulary skills in adults with Down syndrome. This finding contradicts findings from several previous studies of typically developing children who stutter who show significant negative associations (Luckman et al., 2020; Ntourou et al., 2011) or no significant association (Zaretsky et al., 2017) between vocabulary ability and stuttering behavior. As early stuttering in young children is more likely to occur in long and complex utterances (Yaruss, 1999; Zackheim & Conture, 2003) and rarely happens in one-word utterances (Melnick et al., 2003), it may be that children in this study with better vocabulary skills produced more speech in both the assessment and learning situations, thereby increasing the likelihood of stuttering behavior.

Unlike vocabulary ability, stuttering behavior seemingly decreased with increasing grammar ability in the current sample. No prior study has investigated the relationship between grammar ability and stuttering behavior in individuals with Down syndrome. Ntourou et al. (2011) noted lower grammar abilities in otherwise typically developing children who stutter; however, they concluded that the number of studies comparing grammar ability in children who do and do not stutter was too low for making confident claims. In a more recent study of typically developing German preschoolers, Zaretsky et al. (2017) found that stuttering was significantly associated with lower grammar ability. As grammar ability is a specific weakness in children with Down syndrome (Næss et al., 2011), these children may be especially vulnerable to stuttering behavior. Also, Næss et al. (2011) suggested that the specific weakness seen in verbal short-term memory in children with Down syndrome may influence their performance on measures of grammatical ability. Limited verbal short-term memory can introduce a third variable problem. Therefore, we cannot disregard the possibility that the association between stuttering behavior and grammar ability can be explained from this group's limited verbal short-term memory, which was not accounted for in this study.

Practical Implications

As stuttering is found to have negative consequences in terms of communicational, emotional, behavioral, and social functioning (see, e.g., Briley et al., 2019; Jackson et al., 2014; Maessen et al., 2022), the high occurrence of stuttering behavior found in children with Down syndrome in this study and previous studies calls for support by SLPs. However, previous research has reported that individuals with Down syndrome rarely receive systematic treatment by an SLP (Næss, 2018) or a diagnosis of speech fluency disorder (Sommer et al., 2021). According to ICD-11 (World Health Organization, 2019), a diagnosis of stuttering (developmental fluency disorder) should only be used when speech disfluency is not better accounted for by a disorder of intellectual development. Since the identification of stuttering behavior in individuals with Down syndrome may be challenging due to the complex nature of their speech and language difficulties (see, e.g., Preus, 1972), diagnostic guidelines for the assessment of stuttering in this group are needed. Diagnosing stuttering in this population is important in order to understand the nature of the disorder and provide treatment for those who stutter. The assessment approaches in this study focused on audible characteristics only and produced differing results; thus, it seems necessary to combine information from different assessment approaches and aspects of stuttering. As an impact on daily living is considered a feature of stuttering in ICD-11 (World Health Organization, 2019), impact on daily living should also be considered in the diagnostic assessment of children with Down syndrome.

Limitations

There are some limitations that need to be taken into consideration when interpreting the results of this study. Although the present sample is representative of the variability in speech and language ability seen in children of this age who have Down syndrome (see, e.g., Berglund et al., 2001), the sizes of the speech samples were inconsistent and, for some participants, limited. Also, the evaluation of stuttering was based on audio samples only. Although comparisons of audio versus audio/video samples for evaluating the frequency and severity of stuttering have shown a high observer agreement across modalities (Rousseau et al., 2008), video would have made the assessment of physical tension, such as body movements and facial grimaces, possible.

Conclusions

We found a high occurrence of stuttering behavior in this group of Norwegian first-graders with Down syndrome. The relationship between stuttering behavior and language ability varied across time and the measures. As such, the relationship between language ability and stuttering behavior does not seem to follow a strict pattern that can be generalized to all children across time. Although not significant in all the models, the directions of the relationships between stuttering behavior and vocabulary ability and that between stuttering behavior and grammar ability were similar. Overall, the results indicate that it is important to consider the ways in which stuttering behavior might contribute to the serious difficulties with communication commonly experienced by children with Down syndrome. Further knowledge about this group's speech disfluency patterns is an important stepping stone toward the development of diagnostic guidelines for identifying stuttering in these individuals.

Author Contributions

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Institutional Review Board Statement

This study was approved by the Norwegian Centre for Research Data.

Informed Consent Statement

Informed consent was obtained from all families and practitioners (teachers, special needs education teachers, and teacher assistants) involved in the study.

Data Availability Statement

The data can be made available by contacting Kari-Anne Bottegård Næss (k.a.b.nass@isp.uio.no).

Acknowledgments

This research was funded by the Research Council of Norway, Grant 238030 awarded to Kari-Anne B. Næss, University of Oslo. The authors thank the participating children, parents, schools, and research assistants as well as the funder of the study, The Research Council of Norway.

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Appendix A

Coding Categories for Speech Disfluencies

Category	Description with examples
Stuttering-like disfluencies	
Repetition of sounds	Repetition of sounds e.g., “B-b-balloon”
Repetition of syllables	Repetitions of syllables e.g., “Ba-ba-balloon”
Repetition of one-syllabic words	Repetitions of one-syllable words e.g., “I’m going to... to... school”
Prolongation of sounds	Stretching of a sound e.g., “A buuus”, “No, not thaaaaat”
Blocks	No production of sound at the beginning or within the word due to tension e.g., “Can you give me an... (tension) apple?”
Other disfluencies	
Interjections	The addition of a sound, word, or phrase unrelated to the utterance before, in between, or after words e.g., “Ehm”
Revisions	Rephrasing an utterance e.g., “Can I go – should we go home?”
Multisyllabic word and phrase repetitions	Repetitions of words or phrases e.g., “Cause,” “I went... I went home”

The following is a thorough description of how the coding categories are utilized.

Stuttering-Like Disfluencies

Repetitions of sounds, syllables, and monosyllabic words were defined as one or more subsequent repetitions of the sound, syllable, or monosyllabic word. In the current analysis, all subsequent repetitions were included with the following exceptions: (a) in the use of referencing language (e.g., referencing the images, such as “there, there, there”), (b) when the repetition of words reflected a natural/typical way of speech (e.g., “well, well”), (c) when the repetition conveyed meaning within the context of storytelling (e.g., “heeelp heeelp!”).

Prolongations were defined as the stretching of sounds. In the current analysis, all prolongations were coded with the following exceptions: (a) when the prolongation of sounds was judged to be a natural/typical way of speech (e.g., “nooo, I don’t think so”) or (b) when the prolongation reflected an exaggeration to convey meaning within the context of storytelling (e.g., “stooooop the buuuus!”).

Blocks were defined as all distinct “stops”—before or within words—that were characterized as tense. There were no exceptions to the rule. However, it must be noted that children with Down syndrome may exhibit a “staccato” pronunciation or subtle stops within words (commonly before consonants; e.g., “jum-pe”). This may be due to breathing patterns, the strain of trying to pronounce the sounds correctly, or because of how they have been stimulated to pronounce words correctly. These cases were not coded as blocks.

Other Disfluencies

Repetitions of words and phrases were defined as one or more subsequent repetitions of a multisyllabic word or phrase (two or more words). All subsequent repetitions were included with a few exceptions: (a) in the use of referencing language (e.g., referencing the images, such as “there and there and there”) or (b) when the repetition represented a natural way of speech (e.g., “come on, come on!”).

Interjections were defined as the filling of pauses with words unrelated to the utterance—such as “uhm” and “ehm”—before, in between, or after words. Affirmative sounds such as “m-mm” or expressions of dissatisfaction such as “ahh!” were not considered to be interjections.

Revisions were defined as the correcting or rephrasing of a word (e.g., “gage garage”) or utterance (e.g. “can I go – should we go home?”). There were no exceptions to this rule.

Appendix B

Overview of Standardized Measures of Language and Nonverbal Mental Ability

Test name	Reference	Items	Max. score	Target	Procedure
Picture naming	Wechsler Preschool and Primary Scale of Intelligence–Third Edition (WIPPSI-III) Wechsler, 2002	38	38	Measures the ability to verbally express words	The child is asked to name pictures when prompted by a set of single pictures one item at a time.
British Picture Vocabulary Scale - II (BPVS)	Dunn et al., 1997; Norwegian version by Lyster et al., 2010	144	144	Measures the understanding of words	The child is shown four pictures and is asked to point to the picture corresponding with the word spoken aloud by the test administrator.
Test for Reception of Grammar (TROG-R)	Bishop, 2003; Norwegian version by Lyster & Horn, 2009	80	80	Measures receptive grammar	The child is shown four pictures and is asked to point to the picture corresponding with the sentence read by the test administrator.
Grammatic Closure (ITPA)	Kirk et al., 1967; Norwegian version by Gjessing & Nygaard, 1975	33	33	Measures expressive grammar	The child is shown two pictures and is asked to complete a statement that corresponds with the items or events shown in the pictures.
Block Design	WIPPSI-III Wechsler, 2002	20	40	Measures nonverbal mental ability	The child is shown a design modeled and/or pictured and is asked to replicate the design using colored blocks within a certain amount of time.