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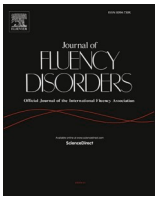

A prospective 14-year follow-up study of the persistence and recovery of stuttering

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ABSTRACT

Purpose: To document the trajectory of early childhood stuttering longitudinally for 14. years with a consideration on the features of overt and covert stuttering related to recovery status.
Method: Thirty-eight participants were observed longitudinally at three different time points: early childhood (Occasion 1), middle childhood (Occasion 2), and late adolescence (Occasion 3). Data collection involved speech samples and reports of stuttering experiences. Recovery on Occasion 3 was estimated through analysis of speech samples, parent and expert judgments, and self- judgement. Two categories of persistence were used: persistent-subjective (no observable stuttering) and persistent-objective (observable stuttering).
Results: The recovery rate was 65.6 %. The majority of the participants showed minimal disfluent speech with 88 % showing less than 1 % syllables stuttered and 97 % showing less than 3 % syllables stuttered in the collected speech samples. All participants classified as persistent reported covert symptoms of stuttering. No relapses in recovery were observed between Occasion 2 and Occasion 3. Late recovery was only observed for those classified as persistent-subjective on Occasion 2. About 64 % of the participants showing observable stuttering (persistent-objective) on Occasion 2 showed no observable stuttering (persistent-subjective) on Occasion 3.
Conclusions: Children continue to recover from early childhood stuttering as they age.The inclusion of self-reports adds to the understanding of recovery especially concerning the covert stuttering behaviours. The presence of overt symptoms of stuttering in the speech samples of children aged 7 to 13 years seems to be associated with the likelihood of late recovery of stuttering

1. Introduction

Childhood stuttering is a neurodevelopmental disorder that usually emerges when children are young, around three years of age (Yairi & Ambrose, 2005). The rate of recovery is high among young children who stutter, especially within a few months of onset (Andrew & Harris, 1964; Yairi & Ambrose, 2005). The reported recovery rate varies greatly across studies. Einarsdóttir et al. (2020) examined 23 studies on recovery of stuttering and found a mean recovery rate of 58.7 %, but with a range of 6.3 % to 94.0 %. The great variance in the reported rate of recovery has many influences, including differences in participant characteristics (clinical or population samples, age of inclusion) and methodology (treatment protocol, follow-up period, inclusion criteria, definition of

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recovery, measurement of recovery). When looking at prospective longitudinal studies of stuttering, the majority of these studies followed the participants only during their preschool years and/or early school years (Einarsdóttir et al., 2020). Since the time that this review was conducted, only two studies reporting recovery rate of stuttering among preschoolers have been published (Singer et al., 2022; Walsh et al., 2020). Similarly, to the studies examined in Einarsdóttir (2020), these studies followed children's recovery over the period of at least 2 years and the reported mean recovery rate was 34 % (Singer et al., 2022) and 61 % (Walsh et al., 2020). Few studies have documented the recovery of stuttering, during the middle school years (9–12 years of age) or early adolescence (13–15 years of age) (Andrew & Harris, 1964; Einarsdóttir et al., 2020; Franken et al., 2018; Howell & Davis, 2011; Howell et al., 2008, 2010). Andrews and Harris (1964) is the only study that has provided detailed documentation of the trajectory of stuttering from early childhood to adolescence. However, the usefulness of this study for describing long term recovery is obscured by an unclear criteria for both inclusion and recovery in this study.

The aim of the study reported in this paper is to examine the trajectory of recovery and persistence of stuttering in a longitudinal study of 14 years, spanning from childhood to early adulthood. This study seeks to examine whether individuals who experience recovery prior to puberty remain recovered, or if symptoms of stuttering re-emerge. Persistence is also estimated to understand the extent to which changes in stuttering occur after puberty. Within this study both overt and covert symptoms are considered and their relationship with trajectories of recovery and persistence explored. Concurrently, this study examines the differences in daily communication between recovered and persistent stutterers and explores negative thoughts which is indication of anxiety about speaking in people with a history of stuttering in early childhood.

1.1. Defining recovery of stuttering in early childhood or adolescence/adulthood

The definition used to estimate recovery rate of early stuttering is likely to impact the calculated recovery rate (Einarsdóttir et al., 2020). Defining the recovery of stuttering is not a straightforward process (Neuman et al., 2019; Tichenor & Yaruss, 2020). The definition of recovery developed by Yairi and Ambrose (1999) is frequently used in early childhood studies on recovery of stuttering (see Einarsdóttir et al., 2020; Yairi & Ambrose, 2005). Recovery under this definition is based on (a) clinicians' and parents' general judgments that the child is not stuttering, (b) stuttering severity being less than 1 on an 8-point severity rating scale, (c) stuttering-like disfluencies (SLD) being fewer than 3 per 100 syllables (based on clinical observation), and (d) no stuttering being present for a minimum of 12 months. This definition focuses solely on observation of overt stuttering behaviours and the judgement of the child's speech by clinicians and parents.

The definition of recovery of stuttering beyond early childhood is more complex. For example, some individuals who stuttered in early childhood may later show only minimal overt symptoms of stuttering. Many people who stutter have learned strategies, with age or through treatment, to cope with overt stuttering behaviours. These strategies can include using reduced speech tempo (Neumann et al., 2019), inserting pauses when speaking (Neumann et al., 2019) or covering their stuttering by changing words or sentences (Constantino et al., 2017). Therefore, in controlled situations overt and observable speech may not accurately reflect how a person actually speaks, particularly under stressful circumstances (Finn et al., 2005). It is known that many people who self-identify as being recovered use strategies, such as these, to sound more fluent (Constantino et al., 2017; Neumann et al., 2019; Sønsterud et al., 2022). These people might select or replace words when talking to avoid stuttering or refuse to speak in some social situations (Boyle et al., 2018). It is debated whether these individuals who have minimal observable stuttering (overt stuttering) and/or who use techniques to minimize potential stuttering (covert stuttering) should be classified as recovered or persistent (Finn et al., 2005; Neumann et al., 2019).

Given these complexities, there is a need to broaden the definition of recovery as put forward by Yairi and Ambrose (1999) for pre-pubescent children. This broader definition should include self-report and should consider both overt (receptions, prolongations and blocks) and covert signs of stuttering (speech avoidance, speech strategies or negative emotions). In line with this, self-report regarding recovery of stuttering has been emphasized by several authors (Brocklehurst, 2013; Finn et al., 2005; Tichenor and Yaruss, 2020). Self-report can provide valuable information on communication in daily situations and the use of strategies to enhance fluency. Further, self-report is necessary as stuttering fluctuates from moment to moment, over time, and across situations. Therefore, only the individual who stutters can truly know about the persistence or recovery of their stuttering (Constantino et al., 2016). Based on the literature, self-report of recovery should include detailed questions about stuttering, the tendency to stutter, the use of strategies to manage stuttering, general emotions and communication in daily life (Brocklehurst, 2013; Constantino et al., 2017; Finn et al., 2005; Tichenor and Yaruss, 2020).

1.2. Longitudinal studies of recovery in early childhood to adolescence

Information on the trajectory of recovery or persistence from early childhood to adulthood is limited, primarily due to a lack of studies. Most prospective studies on the recovery of stuttering have followed participants from early childhood for two to six years (Einarsdóttir et al., 2020; Singer et al., 2020; Walsh et al., 2020). Only three studies have followed the participants for more than six years (Andrews & Harris, 1964; Einarsdóttir et al., 2020; Franken et al., 2018). Andrews and Harris (1964) recruited a population sample of children and followed them from birth to 16 years. There were 43 children who stuttered within this group. By the age of 16, 34 of their participants were classified as recovered, resulting in a reported recovery rate of 79.1 %. About one third of the participants ($n = 13$) started to stutter after 6 years of age. The recovery occurred at different ages (see, Figure 3 in Andrews and Harris): 42 % recovered before 6 years of age, 37 % recovered between 6 and 12 years of age and none recovered after 12 years of age. However, these results need to be interpreted with caution because of the unclear criteria for inclusion and recovery used in this study, as well as

the high attrition rate of 33.2 % (Ingham, 1976). Andrews and Harris' (1964) criteria for inclusion and recovery were based on reports from health visitors, parents and sometimes clinicians with no objective measurement of speech made and no self-report of stuttering or recovery documented. Nonetheless, their study indicated that for some children recovery does occur during the school years. Franken et al. (2018) followed a clinical sample of 15 preschool-aged children with a mean age of 3.9 (Range 2;6 to 5;0) for 9 years. The recovery rate was 73 % (11 of the 15 children) without self-report and 60 % (9 of the 15 children) when self-report was added to the criteria of recovery. Similar results emerged from Einarsdóttir et al. (2020), who conducted a longitudinal study of 38 children from the preschool years (2–5 years of age) to the school years (7–13 years of age). The recovery rate was 71.1 % without self-report, but this dropped to 55.3 % when children's self-report was added to the criteria for recovery. Including self-report as part of the recovery criteria impacted the reported recovery rate in these two studies (Franken et al., 2018; Einarsdóttir et al., 2020), resulting in a decrease from around 70 % (71.1–73.0) to approximately 55 % (55.3–60.0). Adding self-report to the criteria in these studies meant that they were sensitive to covert symptoms of stuttering and variability in stuttering, demonstrating that the inclusion of these factors lowered the reported recovery rate.

In summary, longitudinal research on the recovery of stuttering from early childhood to adolescence is limited, but there are indications that recovery can continue after puberty. Overall, more research, especially longitudinal studies, is needed to better understand the trajectory of stuttering, the likelihood of recovery and the rate of recovery in different ages. Self-report provides valuable information on the subjective experience of participants and decreases the rate of recovery compared to when recovery is based on observable stuttering behaviour alone.

1.3. The recovery of stuttering during and after the middle childhood years

There have been three prospective studies that have examined recovery within the middle childhood years (from 8 years to 12–14 years) conducted by Howell and colleagues (Howell & Davis, 2011; Howell et al., 2008, 2010). Their criteria for recovery were based on researcher, parents and self-judgements, and measurement of speech. Participants in all studies received treatment during the studies, so these reported recovery rates describe participants' response to the intervention as well as being a measurement of recovery. Howell and Davis (2011) followed a large sample of participants (N = 132) for six years, the reported recovery rate was 52.3 %. Howell et al. (2008) followed 76 children for four years reporting a 53.9 % recovery rate. Howell et al. (2010) followed 26 children for four years in which the recovery rate was 46.2 %. Some of the participants classified as recovered had mild stuttering based on the reported measurement scores of stuttering. Nevertheless, all studies by Howell and colleges reported an association between more stuttering severity at the beginning of the study (i.e., 8 years) and later recovery (12 years or more) whereby less severe stuttering at the beginning of the study was associated with a higher likelihood of recovery.

Retrospective studies are another means that has been used to examine late recovery among adults, relying on self-report. Late recovery generally refers to recovery after puberty or in adolescence or adulthood (Finn, 1997; Finn et al., 2005; Neumann et al., 2019). Findings from such studies suggest that recovery continues with age, but also that the participants experience different degrees of recovery. Some participants recover completely, while others continue to have a tendency to stutter (Finn, 1997; Finn et al., 2005; Neumann et al., 2019). Finn et al. (2005) interviewed 15 adults self-reported to have recovered from stuttering without treatment. It came to light in the interviews that seven of the speakers were completely recovered but eight reported that they stuttered occasionally. Those who had tendency to stutter, for instance under demanding speaking conditions, reported using speech modification techniques to maintain fluent speech. Neuman et al. (2019) investigated these fluency inducing methods in more detail with 110 participants living in eight countries. All participants reported that they had recovered from stuttering after 11 years of age. The participants reported using different methods to gain more fluent speech although they considered themselves to be recovered.

In summary both prospective and retrospective studies have reported recovery beyond early childhood. The definition of recovery for these participants must consider both overt symptoms of stuttering and covert symptoms (Brocklehurst, 2013; Finn et al., 2005; Neumann et al., 2019; Tichenor and Yaruss, 2020). Therefore, self-report is a key component in defining late recovery as well as measurements of speech performance and consideration of covert stuttering (Brocklehurst, 2013; Finn et al., 2005).

1.4. Recovery, daily communication, and negative thoughts

Stuttering in adulthood is reported to have a major impact on a person's social communications and daily life (Bloodstein et al., 2021; Boyce et al., 2022; Craig et al., 2009; Yaruss & Quesal, 2006). It has been well documented that people who stutter can experience negative reactions in their environment, such as negative, impatient, and/or critical communication partners which can have serious negative consequences, including reduced quality of life and increased anxiety (Bernard et al., 2022; Blood & Blood, 2007; pp. 3; Boyce et al., 2022). Adults who stutter employ strategies to avoid or hide stuttering in daily communication. This avoidance behaviour, or covert symptoms of stuttering, are associated with avoiding specific situations, activities or even speaking where stuttering might occur (Constantino et al., 2017; Sønsterud et al., 2022).

People who stutter are likely to experience more difficulty communicating in some speaking situations. Situations known to be difficult include public speaking, taking part in group discussions or meetings, and speaking under time pressure (Bloodstein et al., 2021; Yaruss & Quesal, 2006). Blood et al. (2001) compared 39 people who stuttered (aged 13 to 18 years) to 39 people who did not stutter using two standardized communication measures: the Personal Report of Communication Apprehension (PRCA-24) and the Self-Perceived Communication Competence (SPCC) scale. Participants who stuttered reported having significantly poorer communication skills and greater fear of communicating in group discussions and personal conversations than did the comparison group. Stuttering severity was also identified as a contributing factor, with participants who stuttered severely being more likely to believe

they were less competent speakers. In the validation study of the Overall Assessment of Speaker's Experience of Stuttering (OASES; Yaruss & Quesal, 2006, 2008) conducted by Yaruss (2010), the majority of participants reported that stuttering had a negative impact on both their communication and their quality of life.

Social anxiety has also been frequently reported to be experienced by adults who stutter. Iverach et al. (2009) explored the prevalence of anxiety disorders among 92 adults seeking treatment for stuttering, compared to 920 age- and gender-matched controls. They found that compared to controls, the people in the stuttering group had an increased chance of meeting the diagnostic criteria for anxiety disorder according to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV; American Psychiatric Association, 1994) or the International Classification of Diseases (ICD-10, World Health Organization, 1993). In addition, a recent systematic review showed that children and adolescents who stutter exhibit increased anxiety symptoms compared to non-stuttering peers (Bernard et al., 2022). Boyle et al. (2022) investigated the self-reported impact of stuttering across the lifespan in a large cohort (N = 987) of participants aged between 7 and 93 years. About 90.4 % of the participants reported to be anxious due to stuttering. As negative thoughts are linked to social anxiety in general, it has been hypothesized that negative thoughts could be frequent among people who stutter, especially when they are in stressful social situations. Anxiety is often associated with covert stuttering, in that people who are trying to hide their stuttering may feel anxious about the possibility of unexpected stuttering. However, this is not always the case. Some persons who stutter may feel comfortable using strategies to sound more fluent and this is not linked with anxiety (Constantino et al., 2017; Sønsterud et al., 2022). Based on clinical experience, Sønsterud et al. (2022) reported that this subgroup seldom seeks therapy.

As stuttering is known to influence communication behaviour, self-evaluation of communicative control, and feelings of anxiety, examining late recovery of stuttering must consider these aspects, as well as whether overt or covert stuttering is present.

1.5. This study

This study utilizes data collected at a follow-up point 14 years after children were first assessed for stuttering (information on previous phases of this study is available in Einarsdóttir et al., 2020). Longitudinal studies examining the trajectory of stuttering over longer periods of time are lacking. There is currently little evidence to support understanding whether recovery of stuttering continues with age and, importantly, if those who were considered to have recovered from stuttering during their early years remain recovered. The study aims to fill in this gap in current knowledge. Data were collected to examine stuttering recovery/persistence when both objective and subjective methods were used to evaluate recovery. Further, participants' perspectives on their social communication and any strategies they use to enhance their fluency were examined in relation to recovery status. The following research questions were addressed:

1. What is the recovery rate of early stuttering in late adolescence (Occasion 3)?
2. What is the pattern of change in recovery status between middle childhood (Occasion 2) and late adolescence (Occasion 3)?
3. Are there differences in participants' communication in daily situations, negative thoughts and strategies used for modifying the speech based on their recovery status?

2. Method

2.1. Ethics and consent

This study received approval from the Icelandic bioethical committee and all participants provided informed consent prior participating in the study. In cases where the participant was under 18 years of age ($n = 10$), a parent also provided informed consent.

2.2. Participants

The characteristics of participants involved at each time point (Occasions 1, 2, and 3) are presented in Table 1. Inclusion criteria were: (a) child aged 24–71 months, (b) both parents and two SLPs agreed that the child stuttered, and (c) unambiguous moments of stuttering were identified in a video-recorded speech sample. Exclusion criteria were (a) signs of neurological disorder, (b) evidence of severe language impairment, and (c) bilingualism. Thirty-eight children were recruited to study. Participants (N = 38) were assessed at Occasion 1 (Occ-1) during their preschool years (age 2–5 years). All 38 participants were assessed again at Occasion 2 (Occ-2) during middle childhood (age 7–13 years). Data on changes between these two Occasions are presented in Einarsdóttir et al. (2020). On Occasion 3 (Occ-3), 32 of the participants agreed to participate again. These participants were now in late adolescence or early adulthood (age 15–20 years). Attrition was the result of four participants (all male) declining to participate and two participants (also

Table 1
Participant characteristics on each Occasion of data collection.

Participant characteristics	Occasion 1 2004-2006	Occasion 2 2012	Occasion 3 2019
N	38	38	32
Age *M* (*SD*) *Range*	4;3 (1.1) *2;0 – 5;11*	10;9 (1.5) *7;10 – 13;3*	18;6 (1.5) *15;4 – 20;10*
Sex Male/Female	28/10	28/10	22/10

male) not attending their scheduled appointments.

2.3. Materials

2.3.1. Speech samples

Occasion 1. Children were video recorded while playing with the first author and a parent for 30–120 min, depending on how much the child was speaking. Three 1-minute video-recorded speech samples were selected for each child during which they were speaking in sentences and constantly in view. Three minutes of speech were analysed for each child.

Occasion 2. Speech samples were collected for children in two contexts: (a) conversation with an examiner for nine minutes (with 3 samples, each 3 min in length), and (b) reading a passage out loud (with 3 samples, each 3 min in length). Due to technical difficulties and reading difficulties experienced by some of the children, four of the conversation samples and 14 of the reading samples could not be analysed. As a result, 210 speech samples were analysed (see Einarsdóttir et al., 2020).

Occasion 3. As at Occ-2, samples of participants reading aloud and engaging in conversation were recorded on video. Participants self-selected the reading materials from a range of options provided by the investigators. The conversational speech samples were collected while participants discussed topics of interest, such as school, hobbies, and movies, with the investigator. Three samples of conversation (each three minutes) and three samples of reading aloud (each three minute) were collected, resulting in total of six samples from each participant. This led to a total of 192 samples that were analysed.

2.3.2. Parent-provided information

Occasion 1. Parents of participants were interviewed about their children's general developmental milestones. Relevant to the data presented in this paper, parents reported on children's stuttering history and language development. Detailed descriptions of this procedure are provided in Einarsdóttir and Ingham (2008, 2009).

Occasion 2. Parents completed a comprehensive questionnaire on their children's background, development of stuttering, and general academic and social development. Relevant to the data presented in this paper, parents reported on whether their children had recovered or still stuttered. If the child was still stuttering, or stuttering occasionally, the parents were asked to give a stuttering severity rating using a 5-point, qualitative Likert scale: 1 = borderline stuttering, 2 = mild stuttering, 3 = moderate stuttering, 4 = severe stuttering, 5 = very severe stuttering.

Occasion 3. Parents of all participants were interviewed via telephone. Parents were asked to report whether the participant had recovered or still stuttered. If the parent reported that the participant was still stuttering, or stuttered occasionally, the parent was asked to give a severity rating using the same Likert scale, as in Occ-2.

2.3.3. Participants-provided information

Occasion 1. Due to the young age of participants at Occ-1 no child-provided information was collected.

Occasion 2. Participants were asked questions about whether they regarded themselves as someone who stuttered or someone who used to stutter, to estimate the severity of their stuttering (on a same 5-point scale their parents completed) and the variability of their stuttering. All participants were asked to evaluate the fluency of their speech, including overt (repetitions of sounds, syllables, prolongation) and covert (avoiding specific situations or words) stuttering symptoms (see Einarsdóttir et al., 2020; Supplementary material 2, section Fluency Self-Evaluation).

Occasion 3. Participants completed a custom designed written survey in five sections (I Stuttering behaviours and Variability, II Fluency Self-Evaluation, III Speech modification question, IV Communication in daily life, V Negative thoughts, see Supplementary material). The first three questions on stuttering behaviours (I) were the same as on Occ-2 but the last five questions were new. Questions on Fluency Self-Evaluation (II) were only answered by those who reported they stuttered or stuttered occasionally and included questions about overt stuttering symptoms (receptions/prolongation/secondary behaviours) and covered stuttering symptoms (avoiding words, using hesitations to avoid stuttering). These questions were the same as on Occ-2. All participants answered the same questions on speech modification techniques (III), communication in daily situations (IV) and negative thoughts (V). The questions on speech modification techniques were adapted, modified, and translated from Neuman et al. (2019). Five items from Neuman et al.'s list were selected based on these being the items that 29–40 % of the participants reported to have used to reduce disfluencies and being of high importance (>21 from Neuman et al. (2019)). Participants reported how often they used each technique on a 5-point Likert scale with the response options: 1 = *never*, 2 = *rarely*, 3 = *sometimes*, 4 = *often*, and 5 = *always*. Cronbach α was calculated for these questions and showed good internal consistency ($\alpha = 0.80$, 95 % CI [0.66:0.89]).

Questions on communication in daily situations (IV) were adapted and modified from Section III of OASES (Yaruss & Quesal, 2006). OASES was designed to provide an assessment of the experience and impact of stuttering. OASES-S has been translated into Icelandic and validated in the Icelandic context (Leósdóttir, 2014) and translations of the OASES for ages 13–18 (OASES-T) and for adults (OASES-A) were based on this translation. Since many of the participants were not stuttering the questions were modified to suit this situation. For example, *how hard is talking for you …* was changed to *how do you feel when talking…* Eleven of the 20 items were modified and used from the OASES-T. Responses were on a 5-point Likert scale (reversed scale from the OASES) where 1 = *very uncomfortable*, 2 = *uncomfortable*, 3 = *neutral*, 4 = *comfortable*, and 5 = *very comfortable.* Cronbach α was calculated and showed strong internal consistency ($\alpha = 0.87$, 95 % CI [0.79:0.93]) in responses.

Questions on negative thoughts (V) were drawn from the UTBAS-6 questionnaire (Iverach et al., 2016). UTBAS was designed to assess unhelpful thoughts and beliefs about stuttering (Iverach et al., 2011) and later the screening version UTBAS-6 was developed. The first four questions of the UTBAS-6 were translated into Icelandic (translated and back translated, final version made based on

agreement between the investigators) and modified to be suitable for both persistent and recovered groups. The first two questions were modified but the last two were just translated. Question 1) *I will never be successful because of my stutter* became *I will never be successful.* Question 2) *People will think I'm incompetent because I stutter* became *People will think I'm incompetent.* The responses were on a 5-point Likert scale 1 = *never*, 2 = *seldom*, 3 = *sometimes*, 4 = *often*, and 5 = *always*. Cronbach's α was calculated and showed a good internal consistency ($\alpha = 0.79$, 95 % CI [0.64:0.89]).

2.4. Procedure

2.4.1. Collection of interviews and speech samples

Occasion 1. Children were recruited for the study through referrals from speech-language pathologists, from the clinic of the first author, and through responses to media reports about the study (as described in Einarsdóttir & Ingham, 2008, 2009). Parents provided written consent. Investigators interviewed parents and collected speech samples from the children. For most of the children ($n = 35$), speech samples were video recorded during a clinical appointment with the investigators. The remaining speech samples were collected either at the child's preschool ($n = 2$) or at their home ($n = 1$). In all recordings the interlocutor was the first author, except for those made at the preschool where the children interacted with experienced preschool teachers who followed the study protocol for this data collection. The speech samples were recorded using a digital camera (Canon Elura 65).

Occasion 2. Parents of children who participated at Occ-1 were contacted to participate in a study on recovery of stuttering and all parents/children agreed to participate. Parents completed a questionnaire about their children. Children also answered a questionnaire on their stuttering. For most children, a video recorded speech sample was taken during a clinical visit ($n = 35$) using an iPad. Three children's samples were recorded over Skype on a desktop computer as they lived in rural areas and could not attend the clinic (more information in Einarsdóttir et al., 2020).

Occasion 3. Participants/parents who were involved at Occ-1 and Occ-2 were contacted again and asked to participate in a further follow up study. Participants also completed a questionnaire and provided speech samples. Speech samples for 25 participants were video recorded in a university setting using an iPhoneX. The remaining six participants were recorded during a Zoom meeting ($n = 6$). Three of these participants lived in rural areas, and three lived abroad. One participant living in a rural area was seen at his school. This sample was recorded using an iPhoneX. The parents were interviewed by telephone and answered questions on recovery, stuttering, and stuttering severity.

2.4.2. Measurement of speech performances

Examiners were trained to identify stuttering occurrences and evaluate stuttering severity using the training samples provided on the STUREN homepage for Icelandic samples (Einarsdóttir et al., 2014) (see www.sturen.vercel.app). The training utilized an Icelandic version of the Stuttering Measurement System (SMS) training program (Ingham et al., 2008). The SMS computer program was used to calculate the number of syllables, stuttering occurrences, and percentage of syllables stuttered (%SS) for each recording. Following the SMS manual, one stuttering occurrence was counted as one syllable. Each judge trained for approximately 15–20 hours before evaluating the samples (see Einarsdóttir et al., 2020). The following procedure was used for the longitudinal study on for Occ-1, Occ-2, and Occ-3: the first examiner independently listened to each speech sample and rated each one using SMS and the severity using the 11-point STUREN severity scale. For Occ-1 and Occ-2 the second independent examiner re-rated all the samples (100 %) and for Occ-3 the second independent examiner re-rated 66 of the 192 samples (34 %). The investigators met after all speech samples had been analysed, to review all samples and assign qualitative labels for the severity rating as follows: 0 'no stuttering', 1 'borderline stuttering', 2–4 'mild stuttering', 5–7 'moderate stuttering', and 8–10 'severe stuttering'. Differences on the severity scale were discussed until consensus was reached.

2.4.3. Criteria for recovery and persistence

Participants were classified as Recovered, or Persistent stutterers using the following criteria.

Recovered: (1) the participant reported recovery by answering "no" to the questions *do you still stutter* and *do you stutter under certain circumstances*; (2) one or both parents reported recovery and rated the severity of stuttering ≤ 1; (3) the investigators (the two first authors) agreed on the status of recovered and rated the severity rating of stuttering ≤ 1; and (4) both the reading and conversational speech samples showed ≤ 3 %SS.

Persistent-Subjective: (1) the participant reported they still stuttered by answering "yes" to the question *do you still stutter* and/or *do you stutter under certain circumstances*; (2) one or both parents agreed on recovery and rated the severity of stuttering ≤ 1; (3) the investigators (the two first authors) agreed the status of recovered and rated the severity of stuttering ≤ 1; and (4) both the reading and conversational speech samples showed < 3 %SS, and a severity rating of ≤ 1.

Persistent-Objective: (1) the participant reported they still stuttered by answering "yes" to the questions *do you still stutter* and *do you stutter under certain circumstances*; (2) one or both parents agreed that the participant was stuttering; (3) the investigators agreed that the participant was stuttering; and (4) the reading and/or conversational speech samples showed > 3 %SS, and/or a severity rating of > 1.

2.5. Reliability

Measurement of speech performance. Intraclass correlation coefficients (ICC) were calculated to examine inter-rater reliability for number of syllables produced, number of stuttered syllables, %SS and the severity rating. The average ICC for number of syllables was

.92, with a 95 % confidence interval from .87 to .95 ($F(65,66) = 24$, $p < .0001$). The average ICC for number of stuttered syllables was .93, with a 95 % confidence interval from .89 to .96 ($F(65,66) = 29$, $p < .0001$). The average ICC for %SS was .94, with a 95 % confidence interval from .91 to .96 ($F(65,66) = 33$, $p < .0001$) and the average ICC for severity rating was .90, with a 95 % confidence interval from .85 to .94 ($F(65,66) = 19$, $p < .0001$). All ICCs are qualitatively classified as representing excellent agreement (Koo & Li, 2016).

2.6. Data analysis

Descriptive statistics are reported both as absolute numbers and proportions. Inferential statistical analyses (Wilcoxon non-parametric test) were used to calculate the significant difference between the recovered and persistent group in relation to communication in daily life, negative thoughts, variability of talking and the use of speech strategies.

3. Results

3.1. Recovery and recovery rates at occasion 3

The recovery rate was 65.6 % with 21 participants classified as recovered. This was based on the definition which included judgements of the investigators, parents, participants, measurements of speech samples and self-report. Eleven were classified as persistent, seven (22 %) of whom were *persistent-subjective* (showing no overt behaviours on stuttering on speech samples) and four

Table 2
The investigators, parents, and participants measurements of severity of stuttering (0–5) on Occ-2, and Occ-3.

	Occasion 2				Occasion 3			
P	Investigators Severity	Parent Severity	Participant Severity	Recovery*R/ PO/PS	Investigators Severity	Parent Severity	Participant Severity	Recovery*R/ PO/PS
P1	3	3	2	P0	2	2	3	PO
P2	1	0	0	R	0	0	0	R
P3	0	0	0	R	0	0	0	R
P4	0	0	0	R	-	-	-	-
P5	0	3	2	PO	2	2	2	PO
P6	1	3	2	PO	0	0	1	PS
P7	0	0	2	PS	0	0	1	PS
P8	1	0	0	R	0	0	0	R
P9	0	0	2	PS	0	0	0	R
P10	0	0	2	PS	0	0	0	R
P11	0	0	0	R	0	0	0	R
P12	1	0	0	R	0	0	0	R
P13	1	0	2	PS	0	0	0	R
P14	1	0	0	R	0	0	0	R
P15	2	3	3	PO	-	-	-	-
P16	1	2	2	PO	0	0	1	PS
P17	1	3	3	PO	2	3	3	PO
P18	3	2	3	PO	0	1	1	PS
P19	0	1	1	PS	0	0	0	R
P20	1	1	1	PS	-	-	-	-
P21	1	0	0	R	0	0	0	R
P22	0	0	0	R	-	-	-	-
P23	0	1	0	R	0	0	0	R
P24	0	0	0	R	-	-	-	-
P25	3	3	2	PO	0	0	1	PS
P26	0	0	0	R	-	-	-	-
P27	0	0	0	R	0	0	0	R
P28	0	0	0	R	0	0	0	R
P29	0	2	2	PO	2	1	3	PO
P30	0	0	0	R	0	0	0	R
P31	1	4	4	PO	0	0	2	PS
P32	0	3	3	PO	1	1	2	PS
P33	0	0	0	R	0	0	0	R
P34	0	0	0	R	0	0	0	R
P35	0	0	0	R	0	0	1	R
P36	1	0	0	R	0	0	0	R
P37	0	0	0	R	0	0	0	R
P38	0	0	0	R	0	0	0	R

Note. Investigator severity: 0 'no stuttering', 1 'borderline stuttering', 2-4 'mild stuttering', 5-7 'moderate stuttering', and 8-10 'severe stuttering'. Parent and participant severity: 0 = no stuttering, 1 = borderline stuttering', 2 = mild stuttering', 3 = moderate stuttering', 4 = severe stuttering 5 = very severe stuttering. *Classification of recovery: R = Recovered; PO = Persistent-Observed; PS = Persistent-Subjective. - = no data collected

(12.5 %) of whom were *persistent-objective.* See definitions of recovery and persistence in Section 2.4.3. Table 2 shows the severity of stuttering assessed by the investigators, parents and the participants on Occ-2 and Occ-3 including a severity rating, as described in Section 2.4.2. Fig. 1 shows the trajectory of observed %SS for each participant through the three Occasions.

3.1.1. Stuttering measurement on Occ-1, Occ-2, and Occ-3

At Occ-1, the mean %SS was 8.5 (*Range* 1.6–18), and every participant stuttered unambiguously on speech samples, showing %SS of at a least 1. At Occ-2, the mean %SS was 0.6 (*Range* 0–5) for the whole group; for the *recovered group* the mean %SS was 0.1 (*Range* 0–0.4); for the *persistent-subjective* group the mean %SS was 0.3 (*Range* 0–0.7); for the *persistent-objective* group the mean %SS was 1.6 (*Range* 0.3–4.9). Five participants showed %SS between 1 and 5. At Occ-3, the mean %SS was 0.7 (*Range* 0–3.6) for the whole group; for the *recovered group* the mean %SS was 0.1 (*Range* 0–0.8); for the *persistent-subjective* group the mean %SS was 0.2 (*Range* 0–1.0); for the *persistent-objective* group the mean %SS was 2.8 (*Range* 2.4–3.6). Only four participants stuttered unambiguously on Occ-3: P1, P5, P17, P29 with two (P5, P29) showing an increase in their stuttering from Occ-2. One participant (P32) showed borderline stuttering on the recording and reported occasionally stuttering while reading. He was classified as *persistent subjective.* No participant was stuttering moderately or severely on Occ-3.

3.1.2. Stuttering background, variability and fluency self-evaluation

Approximately one-third ($n = 11$, 34.4 %), all classified as recovered, reported that they did not recall having experienced stuttering. Most participants ($n = 26$, 81.3 %) remembered receiving some form of treatment but only a small proportion ($n = 8$, 25.0%) could recall specific details of the treatment, such as where and when it occurred. The persistent groups reported experiencing significantly more variability in their speech fluency compared to the recovered group ($W = 60.5, p = 0.04$). The participants classified as persistent (both *persistent-subjective* and *persistent-objective*) answered questions where they evaluated their fluency while speaking. All reported selecting words when speaking to avoid stuttering and feeling that they could get stuck before and within words, resulting in them being unable to continue speaking. The majority ($n = 9$, 81.8%) reported that they repeated sounds and syllables and/or prolonged sounds, while 63.6 % ($n = 7$) reported using interjections or small words to avoid stuttering. Only one-third ($n = 4$, 36.4 %) reported that they were hesitant to say their own names or that stuttering had interfered with their studies. Even fewer ($n = 2$, 18.2 %) reported that stuttering influenced their choice of employment.

3.2. Changes in recovery status from middle childhood to late adolescence

The pattern of change in recovery status from middle childhood (Occ-2, 10;9 years) to late adolescence (Occ-3, 18;6 years) is

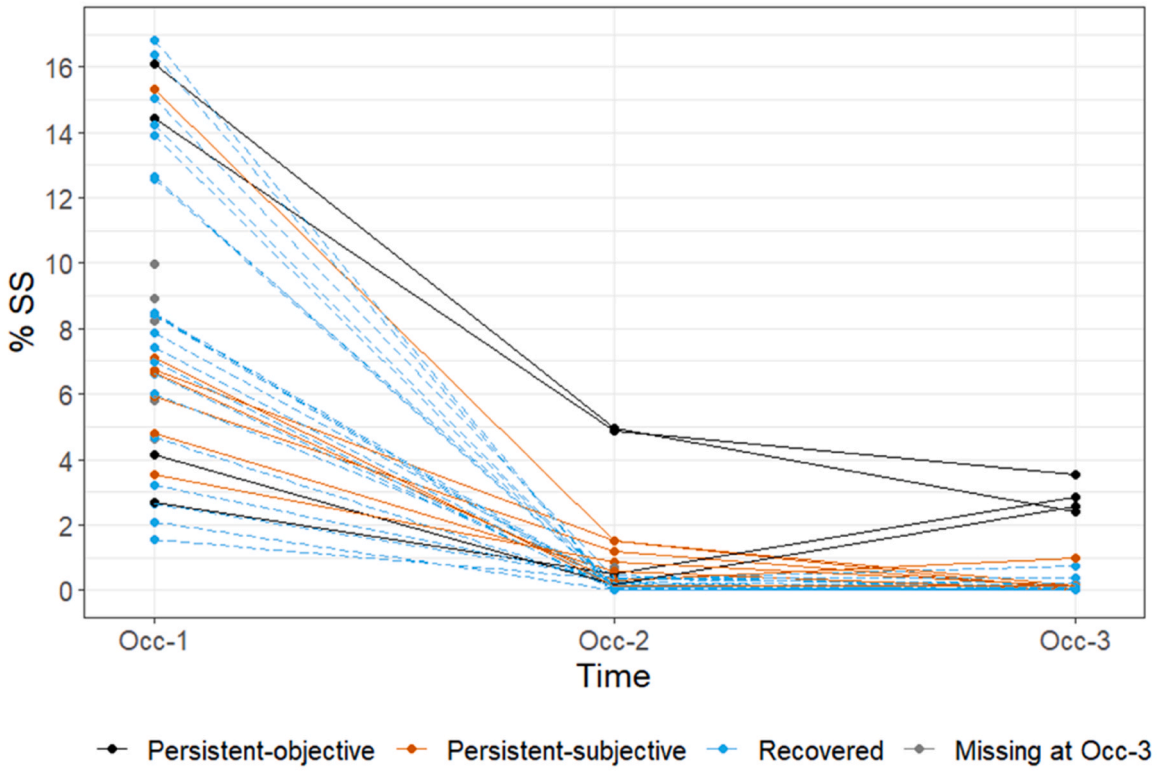

Fig. 1. The trajectory of the severity of stuttering measured with %SS through Occ-1, Occ-2, Occ-3.

visualized in Fig. 2. Those who were considered *recovered* during middle childhood (Occ-2) remained *recovered* in late adolescence (Occ-3) what indicates no relapses concerning the recovery before and after puberty. Seven participants were *persistent-subjective* at Occ-3. At Occ-2, these participants were classified as: *persistent-subjective* ($n = 1$) or *persistent-objective* ($n = 6$). The four participants who were classified as *persistent-objective* at Occ-3 were also all classified as *persistent-objective* at Occ-2. The six participants who did not participate on Occ-3 were classified on Occ-2 as *recovered* ($n = 4$), *persistent-subjective* ($n = 1$) and *persistent-objective* ($n = 1$) (see Table 2).

3.3. Speech modification techniques, communication in daily situations and negative thoughts

All participants answered questions concerning the use of speech modification techniques, communication in daily life and frequency of negative thoughts. Responses were on a 5-point Likert scale, with higher scores indicating better communication, but also higher frequency of negative thoughts and the use of speech modification techniques (see Table 3). The persistent groups reported utilizing speech modification techniques significantly more frequently than the recovered participants ($W = 15.5$, $p < 0.001$). The techniques reported included slowing down their speech rate, concentrate on their breathing and incorporating pauses into conversation.

The participants reported experiencing comfortable communication in daily situations (*recovered* ($M = 4.1$, $SD = 0.5$), *persistent-subjective* ($M = 4.2$, $SD = 0.5$) *persistent-objective* ($M = 3.2$, $SD = 0.7$)) with *persistent-objective* showing a slightly lower value. Regardless of recovery status the participants reported to be challenging speaking in front of large groups and under time pressure. No significant difference was found between the recovered and the persistent group (PS and PO) ($W = 142.5$, $p = 0.29$). Negative thoughts were not frequently reported ($M = 2.1$, $SD = 0.8$) and the difference between the recovered and the persistent group was not significant ($W = 109$, $p = 0.81$).

4. Discussion

This is the first study to longitudinally follow children with stuttering and monitor persistence/recovery from the preschool years until adulthood using detailed measurements of speech and self-report of speaking-related thoughts and behaviours. The recovery rate was 65.6 %. The few participants who showed overt behaviours of stuttering at the end of the study, were all rated to stutter at a level of mild severity. Analysis of speech samples revealed a decline in the severity of stuttering as participants grew older. All participants, classified as persistent (n = 11, 34.4 %) reported covert symptoms of stuttering such as selecting words and using speech modification strategies but only one-third (n = 4, 36.4 %) showed also overt behaviour. The seven participants who were classified as *persistent-subjective* on Occ-3 rated their stuttering as mild or borderline. Participants who recovered at the age of 7–13 (Occ-2) appear to experience complete recovery and no relapses were observed in this cohort. Late recovery was also observed, but only those who were classified as *persistent-subjective* on Occ-2 were considered to be recovered on Occ-3. There was a significant difference between those who were classified as recovered and those who were classified as persistent in the reported variability of speech skills and the use of speech modification techniques. On average, the group reported being satisfied with communication in daily situations, with no signs of frequent negative thoughts. There were no significant differences between recovered and persistent participants in their reported communication in daily situations and or negative thoughts.

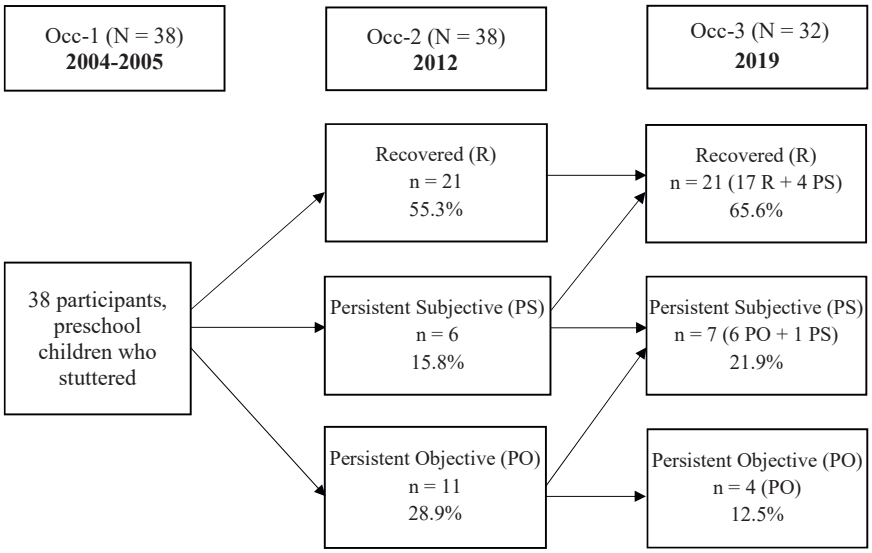

Fig. 2. The rate of recovery through the 14 years of follow-up.

Table 3
The reported use of speech modification techniques, experience of daily communication, and frequency of negative thoughts.

	Recovered (N = 21)		Persistent (N = 11)			
			PS		PO	
	M	SD	M	SD	M	SD
Speech modification techniques	2.1	0.6	3.1	0.4	3.6	0.3
Communication in daily situations (11 items)	4.1	0.5	4.2	0.5	3.2	0.7
Questions on negative thoughts (4 items)	2.1	0.8	1.9	0.7	2.5	1.0

Note. Higher score means better more frequent use of speech techniques, more comfortable communication experience, but also more frequent report on negative thoughts on 5- point Likert scale.

4.1. The recovery rate, self-report and covert stuttering behaviours

The result of this study showing the recovery rate of 65.6 % is in line with previous studies using similar approach for defining recovery, 60 % in Franken et al. (2018) and 55.3 % in Einarsdottir et al. (2020). This rate is lower than in previous longitudinal studies that did not use self-report for defining recovery. In Andrews and Harris (1964) the reported recovery rate was 79.2 %, in Franken et al. (2018) it was 73 % without self-report, and in Einarsdóttir et al. (2020) it was 71 %. In this study the reported recovery rate without self-report is 87.5 %. Earlier longitudinal research on childhood stuttering followed participants during the preschool years for shorter periods, often less than six years, showed much greater variations in recovery rate, ranging from 6.3 % to 94.0 % using different criteria for recovery (see Einarsdóttir et al., 2020). Participants in the current study who were fully recovered by the age of 7–13 did not relapse, and many did not remember ever stuttering. This indicates that recovery during the preschool or early school years may represent complete recovery.

It was clear in this study that self-reporting of recovery added to the understanding of recovery or persistence of stuttering. Many of those who report still stuttering and/or stuttering under certain circumstances did not have unambiguously stuttered speech in either the speech samples analysed or reported from their parents. The persistent participants, both *persistent-subjective* and *persistent-objective*, reported to use speech strategies to modify their speech, all reported that they selected or replaced words when speaking and the majority used interjections or small words to avoid stuttering. Both persistent groups reported using a range of techniques significantly more often to modify their speech than the recovered group. These techniques included reducing the speech tempo when speaking and trying to be more relaxed. The use of these speech modification strategies has been associated with covert symptoms of stuttering (Constantino et al., 2017; Neumann et al., 2019; Sønsterud et al., 2022; Yaruss & Quesal, 2006).

Recent studies on covert stuttering (Constantino et al., 2017; Sønsterud et al., 2022) have shown that the concept of covert stuttering is complex and not necessarily connected with anxiety or avoidance strategies for all participants, as previously thought. The persistent participants use speech strategies to gain more fluent speech and as in Constantino et al. (2017), Sønsterud et al. (2022) is doesnt necessarily seem to be linked with anxiety. One persistent subjective participant reported to pause briefly when experience stuttering, while another reported to change words "*just out of habit*". This is consistent with findings from studies where the participants identified themselves as recovered, although they report to stutter occasionally, possibly because they don't regard their stuttering as a problem (Finn et al., 2005; Neumann et al., 2019).

4.2. The recovery of stuttering from middle childhood to adolescence

Late recovery was observed in current study as in earlier retrospective studies on late recovery (Finn, 1997; Finn et al., 2005; Neumann et al., 2019) as well as in Andrews and Harris' (1964) study. Those who were classified as *subjective persistent* at Occ-2, all except one were classified as recovered in this study. The participants who showed overt stuttering behaviours on the speech samples or *objective-persistent* at Occ-2 either remained classified as *objective-persistent* (n = 4) or were re-classified as *subjective -persistent (n = 7)*. The two participants who were stuttering more than 3 %SS on the speech samples on Occ-2 continued to stutter what could suggest that severity on the speech sample during middle childhood could predict recovery. However, due to the small number of participants in this study the interpretation is limited. The severity of stuttering has been associated with recovery in middle childhood (Howell & Davis, 2011; Howell et al., 2008, 2010) and after 4 years of age (Walsh et al., 2020) and it was also one of the predictors of persistence in Singer et al.'s. (2022) study.

Studies on late recovery (Finn, 1997; Finn et al., 2005; Neumann et al., 2019) have reported on a group of participants who have tendency to stutter but seems to consider themselves as recovered or do not associate disability with their speech. Finn (1997) described a group who occasionally stuttered in emotional situations and were classified as partly recovered. In Neumann et al.'s (2019) study on late recovery of stuttering, only a quarter of the participants reported to experience complete recovery, the others experienced occasional stuttering. The classification of these individuals as recovered, persistent, partly recovered or partly persistent remains controversial. The experience of feeling recovered from stuttering could be associated with not seeing the stuttering as a problem and not experiencing being handicapped by the disorder, as Finn et al. (2005) suggested. It is possible that individuals achieving late recovery with residual stuttering do not experience disability or negative feelings associated with stuttering, as they may have developed coping mechanisms. However, some underlying disorder might persist.

4.3. Communication in daily life and negative thoughts

The reported communication in daily life was reported to be comfortable for the *recovered* group and the *persistent subjective* group but the *persistent objective* group estimated it on average as neutral. The lack of significant difference between the recovered and persistent groups in the reported communication in daily life might be explained by that the overall satisfaction with communication within the *persistent subjective* group. In Franken et al. (2018) the participants also showed on average good communication in daily life.

Participants all reported seldom or never having negative thoughts what is not in line with previous studies among people who stutter (Iverach et al., 2009, 2016). Once again, sample size likely played a role in these findings, and the low severity of stuttering among the persistent participants may have also impacted the results, as all were stuttering mildly. The questions used in the study were not validated and were modified from the original questionaries (OASES and UTBAS) to use for the purposes of this study.

4.4. Limitations

The participants cohort in this study was small, as is typical of many previous longitudinal studies on the recovery of early stuttering. A greater number of participants would make it possible to perform more comprehensive inferential analyses similar to those performed by Singer et al. (2022) and Walsh et al. (2021), using a combination of factors that could predict recovery and minimizing the impact of attrition. Although attrition rate was low on Occ-3 (15.8 %) there was a gender difference in the participation with all females participating across all three occasions while six males (21.4 %) did not participate on Occ-3. No attrition on Occ-3 would have been desirable for more robust findings. Additionally, the participants were only assessed once on each Occasion. Repeated measurements in different situations over time would be preferable to capture the inherent variability of stuttering across time and situation. Further, standardised questionnaires designed for people who stutter had to be adapted for this study, as no appropriate standardized questionnaire suitable for both the recovered and persistent groups was available. This adaptation might have impacted the measures' validity and reliability. Finally, this study did not control for the effect of treatment which may have impacted recovery but was outside the scope of this project.

4.5. Conclusion

Overall, this study contributes to our understanding of the long-term outcomes of early childhood stuttering. The finding shows that the recovery status in early childhood seems to be robust for those who have recovered. The trend is towards recovery and adjustment of speaking and communicating those who were classified as persistent in early middle childhood. Emphasizing the importance of including self-report measures in the recovery assessment, it is important to consider different degrees of recovery or persistence and the participants perception on their speech. The findings highlight the need for larger sample sizes and repeated measurements over time to grasp the factors contributing to recovery. Further research is needed to investigate the predictors of recovery and to explore additional factors that could enhance our understanding of effective treatment options for adolescents and adults who stutter.

Ethics statement

Ethical approval was obtained from the Icelandic bioethical committee 2019 registration number (VSN-19–123).

CRediT authorship contribution statement

Jóhanna Thelma Einarsdóttir: Writing – review & editing, Writing – original draft, Supervision, Resources, Project administration, Methodology, Funding acquisition. **Brynja Hermannsdóttir:** Writing – original draft, Methodology, Investigation. **Kathryn Crowe:** Writing – review & editing, Writing – original draft, Formal analysis, Data curation, Conceptualization.

Declaration of Competing Interest

The authors confirm that there is no conflict of interest to declare.

Data Availability

The authors do not have permission to share data.

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Appendix A

		Occasion 1			Occasion 2			Occasion 3		
P	SEX	AGE	%SS	SEV	AGE	%SS	SEV	AGE	%SS	SEV
P1	F	5	16.08	Mo	12	4.94	Mo	20	2.40	Mi
P2	M	5	1.55	Mi	12	0.32	Bo	20	0.76	N St
P3	M	5	3.21	Mo	12	0.33	N St	19	0.23	N St
P4	M	3	4.61	Mo	10	0.38	N St	-	-	-
P5	M	4	4.14	Mi	11	0.17	N St	19	2.57	Mi
P6	M	5	3.54	Mi	13	0.85	Bo	20	0.15	N St
P7	F	4	7.09	Mo	11	0.15	N St	19	0.15	N St
P8	F	5	2.64	Mo	12	0.39	Bo	19	0.39	N St
P9	F	5	12.67	Mo	13	0.09	N St	20	0.00	N St
P10	M	4	15.05	Se	12	0.38	Bo	19	0.15	N St
P11	M	4	4.67	Mi	11	0.14	N St	19	0.00	N St
P12	F	3	7.85	Se	11	0.22	Bo	18	0.00	N St
P13	F	4	12.56	Mo	11	0.26	Bo	18	0.09	N St
P14	M	3	8.40	Mo	10	0.01	Bo	18	0.00	N St
P15	M	4	5.80	Mi	11	0.70	Mi	-	-	-
P16	M	4	5.93	Mi	11	1.19	Bo	19	0.06	N St
P17	M	3	14.43	Se	11	4.89	Mo	18	3.55	Mi
P18	M	5	15.32	Se	12	1.51	Bo	20	0.00	N St
P19	M	5	13.92	Se	12	0.04	N St	20	0.00	N St
P20	M	4	8.25	Se	12	0.67	Bo	-	-	-
P21	M	4	6.01	Mi	10	0.44	Bo	17	0.00	N St
P22	M	4	8.44	Mi	10	0.00	N St	-	-	-
P23	M	2	6.62	Mo	7	0.09	N St	15	0.00	N St
P24	M	3	9.98	Se	8	0.00	N St	-	-	-
P25	M	4	6.74	Mo	10	1.50	Mi	17	0.15	N St
P26	M	2	8.92	Mi	8	0.09	N St	-	-	-
P27	F	2	8.42	Mo	8	0.08	N St	15	0.00	N St
P28	M	3	8.48	Mo	9	0.19	N St	16	0.00	N St
P29	M	5	2.69	Mi	11	0.50	N St	19	2.86	Mi
P30	M	3	7.43	Mo	9	0.09	N St	16	0.00	N St
P31	M	4	4.78	Mo	10	0.58	Bo	18	0.00	N St
P32	M	4	6.64	Mo	10	0.28	N St	18	0.97	Bo
P33	M	5	6.98	Mo	11	0.02	N St	18	0.00	N St
P34	F	2	2.08	Mi	8	0.00	N St	15	0.00	N St
P35	F	4	14.25	Mi	10	0.08	N St	17	0.04	N St
P36	M	5	12.58	Mo	11	0.05	Bo	18	0.00	N St
P37	F	2	16.38	Se	8	0.00	N St	16	0.00	N St
P38	M	3	16.81	Se	9	0.00	N St	16	0.08	N St

Note. M = male; F = female; %SS = percent of syllables stuttered; SEV = severity; NSt = not stuttering; Bo = borderline; Mi = mild; Mo = moderate; Se = severe; - = data not collected.

Appendix B. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.jfludis.2024.106058.

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