THE NATURAL HISTORY OF STUTTERING ONSET AND RECOVERY: DATA FROM A LONGITUDINAL STUDY

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The natural history of stuttering onset and recovery: data from a longitudinal study

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ABSTRACT

A large longitudinal epidemiological study in Australia allows to bring recent figures on the number of Children Who Stutters (CWS) and on the predictors of the persistence or recovery of the stuttering. The research is based on a rigorous methodology and controls several intrinsic factors to the child and relative to his or her family.

KEY WORDS: CHILDHOOD, STUTTERING, STAMMERING, ONSET, RECOVERY.

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The authors acknowledge the families and children who participated in the study and the team of investigators comprising Professors Onslow, Wake, Packman, Bavin and Prior and Doctors Eadie, Bretherton, Block and Ukoumunne. Other team members include Ms Cini and Ms Conway.

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Little is known about the epidemiology and natural history of stuttering. Prior to the Early Language in Victoria Stuttering Study, there had been 2 prospective studies of early childhood stuttering based on population\(^1\) samples. Andrews and Harris (1964) documented the cumulative incidence of stuttering in their 1000-family study in Newcastle-Upon-Tyne in England. In that city, between May and June in 1947, all children born were assessed annually up to 6 years. Andrews and Harris reported the cumulative incidence of stuttering to be 3.5\% by 5 years. By 6 years, 42\% (18 of 43 children) had recovered naturally. All cases of stuttering were confirmed by health visitors. No details were provided regarding how long after stuttering onset each child was assessed. Mansson (2000) recorded the cumulative incidence of stuttering by 3 years of age in a cohort of 1,042 Danish children born on the island of Bonholm. One month prior to each child’s birthday, parents were sent a letter offering their child a free speech and language screening. During the screening, children participated in a play session during which their speech was examined for evidence of stuttering behaviours. At this time, 5\% of children had stuttered. Cases of stuttering onset prior to 3 years however were not assessed. At 5 years, all previously confirmed cases of stuttering were reassessed to determine whether these children had continued to stutter. Additionally, researchers accessed all kindergarten and school reports for reports of new stuttering cases and only two additional cases were found hence the cumulative incidence of stuttering remained at 5\% at 5 years. Further, 71\% of the children identified as stuttering at 3 years, had recovered naturally by 5 years.

Many studies, such as those from Illinois, (Yairi & Ambrose 2005). have made valuable contributions to the literature however, incidence rates have been based either on participants recruited after stuttering onset or on general health professionals’ assessment and identification of stuttering cases which were not verified by trained speech pathologists. The Early Language in Victoria Stuttering Study (ELVS Stuttering) was developed to address these methodological issues.

**OVERVIEW OF THE EARLY LANGUAGE IN VICTORIA STUDY**

ELVS Stuttering commenced in Melbourne, Australia in 2005 and was embedded within the framework of the Early Language in Victoria Study (ELVS) which commenced in 2003. ELVS is a multi-disciplinary study involving speech pathology, epidemiology, paediatrics, linguistics and psychology. ELVS is based at the Murdoch Childrens Research Institute in Melbourne, Australia. Researchers at The Royal Children’s Hospital, Murdoch Childrens Research Institute, La Trobe University, University of Sydney and the University of Melbourne collaborated throughout the study. ELVS has been funded by two National Health and Medical Research Council Project Grants (Identification Numbers

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\(^1\) By population study we mean a sample of children derived from the community rather than samples that are recruited from clinics.
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237106 and 436958) and the Australian Research Council Discovery grants (Identification Numbers DP0557291 and DP0984833).

Objectives

The ELVS Stuttering Study sought to address the limitations of previous studies by establishing a large, community cohort of Australian children recruited prior to stuttering onset. The broad objective of ELVS Stuttering was to document the epidemiology and evolution of stuttering during the preschool years. Since its establishment, ELVS Stuttering has addressed some specific aims including documenting the cumulative incidence of stuttering up to 4 years of age; examining predictors of stuttering onset by 3 years and measuring recovery from stuttering 12 months post onset at 4 years of age.

Recruitment of Participants for ELVS

ELVS has a prospective longitudinal observational design. Between September 2003 and April 2004, potential participants were recruited from 6 of the 31 metropolitan Local Government Areas (LGAs) in Melbourne, Australia. The Australian Bureau of Statistics developed the Socio-Economic Indexes for Areas (SEIFA) Index of Disadvantage which was derived from 2001 Australian census data. The SEIFA Index of Disadvantage considers characteristics such as high unemployment, low educational attainment and low income. Lower scores reflect areas which have greater disadvantage compared to other geographic areas. Using this index, the 31 metropolitan LGAs were stratified into 3 tiers. Two non-contiguous LGAs were then selected from each tier. In order to ensure that relatively even numbers of participants were recruited from each tier, the annual number of births and ranges of SEIFA indices in each LGA were considered during the selection process. The final sample covered a broad range of areas across Melbourne from disadvantaged to advantaged.

Participants were recruited using three methods: maternal and child health (MCH) nurses, MCH hearing screening visits, and media publicity. MCH Nurses were asked to approach the parents of every infant who attended their routine 8-month-old visit or hearing screening session between September 2003 and January 2004. MCH Nurses obtained written consent from interested parents and then the ELVS Research Team contacted these parents via telephone and letter to discuss the details of the study. Informed written consent was obtained from all parents who agreed to participate. Additionally, advertisements were included in suburban and metropolitan newspapers within the selected LGAs. Interested parents were asked to contact the ELVS Team directly. More detailed descriptions of the methodology can be found in Reilly et al. (2006) and Reilly et al. (2007).

Selection Criteria

Only infants who were living in one of the selected LGAs and who were aged between 7.5-10 months during the recruitment phase were eligible for participation. Children who had a developmental, congenital, physical or intellectual disability were excluded from the study. Additionally, children whose
parents could not speak and understand English at an adequate level for recruitment and completion of questionnaires were excluded. In total, 1,910 children were recruited into ELVS at 8 months of age.

Parent-Report Questionnaires
Following recruitment, the first wave of data collection commenced. The return rate for these 8-month-old questionnaires was 82.2% (1,910 of 2,325 questionnaires were completed and returned). A parent-report questionnaire was distributed to all eligible and consenting parents. The questionnaire measured a range of early communication and language skills as well as other child, family and environmental measures. Data included information relating to communication, language and speech development, general development and health, literacy and phonological awareness skills, socio-demographic characteristics, parent-child interaction, family history of speech and language difficulties, child behaviour and temperament, feeding, hearing, voice and cognition. Additionally, data relating to parents was also collected. Maternal vocabulary, maternal mental health, family stress factors, literacy and parental education were all measured. These parent-report questionnaires were completed annually around the time of the child’s birthday.

Predictor Measures
Many of the variables examined as predictors of onset were obtained from the 8-month-old questionnaire. These were gender, twinning, birth weight, premature birth status, birth order, maternal education level, family history of speech, language, reading and stuttering difficulties. Each child’s socio-economic status was calculated based on their SEIFA Index of Relative Disadvantage score.

Remaining predictor variables were collected from the 2-year-old questionnaire. Vocabulary scores were obtained from the MacArthur-Bates Communicative Development Inventories (CDI). Children who received a vocabulary production score below the 10th percentile were classified as late talkers. The Communication and Symbolic Behaviour Scales (CSBS) Infant-Toddler Checklist was used to measure children’s communicative behaviours up to 2 years of age. One of the 5 temperament dimensions approach/withdrawal from the Australian Temperament Project Short-Form (Toddler version) was also included in the questionnaire. Parents were asked to assign a score from 1 (almost never) to 6 (almost always) which reflected how often they observed their child exhibiting the behaviour described. Children who were more shy/withdrawn received higher scores. The Nonspecific Psychological Distress (Kessler-6) was used to measure maternal mental health.

Recruitment of Participants for ELVS Stuttering
At 2 years of age, ELVS participants who had returned their annual questionnaire were eligible for inclusion in ELVS Stuttering. Parents of eligible participants were contacted using a recruitment pack. Recruitment packs were mailed to participants and contained 3 documents.
Additional parent information statement

This letter provided details of the ELVS Stuttering study, what participation in the study would involve, the benefits of participating, and instructions for participants to follow if they did not want to participate in the study.

Opt-out letter

Parents who elected not to participate in ELVS Stuttering were asked to sign and return an opt-out letter to the ELVS Research Team using a self-addressed reply paid envelope which had been included in the recruitment pack. Those parents who return the opt-out form were not contacted again in relation to ELVS Stuttering however they did continue to participate in the original ELVS Study.

Refrigerator magnet

A refrigerator magnet containing a checklist of examples of different speech behaviours which typify stuttering was also included in the recruitment pack.

Parents who did not opt out of the ELVS Stuttering Study were sent reminder letters every 4 months asking them to contact the ELVS Research Team if they noticed their child exhibiting any of the speech behaviours listed on the refrigerator magnet.

Assessment Procedures

Telephone contact

When a parent contacted the ELVS Research Team to report that their child was stuttering, a speech pathologist interviewed the parent over the phone to determine the nature of the speech difficulties being described. If the speech pathologist confirmed that the speech behaviours being reported by the parent typified stuttering or was unsure whether the behaviours were stuttering, a home visit was arranged. Home visits were arranged wherever possible, within 2 weeks of the initial telephone interview.

Home visit to confirm stuttering status

At the initial home visit, parents were interviewed by the speech pathologist about the onset of their child’s stuttering as well as the speech behaviours their child was exhibiting. A family history of stuttering was also obtained. After completing the interview, the speech pathologist videotaped a 25-minute play session between the parent and their child. Parents were told to “play with their child as they normally would” and restrict the number of questions they asked their child in order to minimise the number of 1-word utterances their child produced. An identical set of toys and books were used to stimulate conversation between each parent-child dyad. Following the play session, the parent and speech pathologist completed the stuttering severity scale. This was a 10-point scale, where a score of 1 represents no stuttering, a score of 2 represents extremely mild stuttering and a score of 10 represents extremely severe stuttering. Parents were asked to assign a score on this scale, which reflected their child’s stuttering severity during
the play session. Additionally, two stuttering specialists (Professors Onslow and Packman) reviewed the speech samples and completed the stuttering severity scale. Stuttering was confirmed when the parent, speech pathologist and both stuttering specialists assigned a score of 2 or more on the severity scale.

**Monthly home visits**

Participants who were confirmed as stuttering at the initial home visit then had 12-monthly home visits scheduled. At each of these visits, the speech pathologist attended the participants home to videotape a 25-minute play session between the parent and child. At the conclusion of each play session, the parent and speech pathologist completed the stuttering severity scale. In addition to this, parents were asked to assign a score using the severity scale, which reflected an average of their child’s stuttering severity over the past week.

At each monthly home visit, parents also completed a stuttering questionnaire. These questionnaires asked parents questions about their child’s stuttering over the past month. Data were collected about the following parameters:

- Type of stuttering behaviours exhibited by their child;
- Factors that parents felt had exacerbated or improved their child’s stuttering;
- Whether their child had displayed any awareness of their stuttering;
- Different reactions that any family members or friends had used toward their child

Figure 1 summarises the cumulative recruitment of participants reported and confirmed as stuttering from 2 to 4 years.

**RESULTS**

At the two year follow up, 1620 families of the original 1910 ELVS cohort participated in the ELVS stuttering study. Of the 291 non-participants, 57 were unavailable to participate and 234 opted not to be involved. Comparisons made between the participating and non-participating groups showed participants had more mothers with a degree or postgraduate qualification.

158 parents contacted the ELVS team to report their child had started to stutter by 3 years of age. During a subsequent home visit 137 of these children were confirmed as stuttering, equating to 8.5% of the sample. In 21 cases, a definitive diagnosis of stuttering could not be made, and children were classified by the panel as “non-stuttering” for the main analyses (Reilly et al., 2009). At 4 years of age, stuttering onset was confirmed in 181 children, with a cumulative incidence of 11.5%.

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2 Both Professors Packman and Onslow are investigators on the ELVS stuttering study and are from the University of Sydney.
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Recruited into ELVS Stuttering Study
N = 1,619

Confirmed as stuttering by 2-years
N = 12 (0.7% of participants)

Confirmed as stuttering by 3-years
N = 148 (9.1% of participants)

Confirmed as stuttering by 4-years
N = 181 (11.2% of participants)

Figure 1.
Cumulative Recruitment of the Stuttering Group

Nature of Onset
Stuttering onset was most common between 25 and 40 months with only a small minority of children starting to stutter prior to 24 months. 133 of the 137 children started to stutter when their utterance length increased to 3 or more words. Sudden onset was common, with 37% reporting stuttering starting within a day and 12.3% over 2-3 days. Gradual onset was reported in 27% of children over 1-2 weeks, 13.9% over 3-4 weeks and 5.8% over 5 weeks. Almost all parents (91.2%) reported an episodic or intermittent presentation of stuttering and in nearly three quarters (71%) of the sample, stuttering consisted of whole word repetitions.

Predictors
Boys, twins and children with mothers educated at a degree level or higher were all significantly associated with stuttering onset at 3 and 4 years of age. The children in the stuttering group at 3 years also had higher total scores on the CDI than the children who did not stutter. Specifically, an increase of 100 words on the vocabulary score corresponded to an increase of 17% in the odds of beginning to stutter (Reilly et al 2009). However, having a larger vocabulary was not associated with cumulative incidence at 4 years. Despite these findings, the overall the multivariable model showed a low predictive strength, accounting for only 3.7% of the total variation.

Mean shyness, birth weight and maternal mental health scores were all similar in the two groups. Late talkers and children with higher total scores on the CDI and CSBS at 2 years were represented in lower numbers in the stuttering group. None of the predictors measured were associated with recovery.
Case histories with the confirmed stuttering children revealed 51% had a positive family history of stuttering. Between 20-30% reported a family history of other speech, language and academic difficulties. Conditions such as epilepsy, intellectual impairment, language problems, motor difficulties and attention and concentration problems, occurred less frequently in the families (<20%). Detailed information on results can be found in Reilly et al. 2009.

Recovery
Over the course of the last 3, monthly home visits (as discussed above) children were classified as either “recovered”, “not recovered” or “recovery status not determined”. A child was considered “recovered” if both the parent and research assistant rated the child a severity rating of 1 for at least 2 of the 3 visits, and the child had no score greater than 2. If neither the parent nor research assistants’ severity ratings met these criteria the child was classified as “not recovered”. “Recovery status not determined” was assigned if the parent and research assistant ratings were discordant for recovery (Reilly et al., 2013). 9 children (6.3%) were classified as recovered within 12 months of stuttering onset, of whom all were boys. 4 of these children sought professional help and 3 saw a speech pathologist. The recovered group had a lower mean parent stuttering severity rate and fewer whole word repetitions at onset. As discussed by Reilly and colleagues (2013), the 95% confidence intervals suggested that the true recovery rate in the study population would not be greater than 12%, which is much lower than previously suspected. There was no evidence that any of the other potential predictors were associated with recovery status. For further details on recovery from stuttering at four years of age, see Reilly et al. 2013.

DISCUSSION
Findings of this prospective longitudinal study showed a cumulative incidence of stuttering of 8.5% at 3 years, and 11.5% at 4 years. These figures are significantly higher than previously reported in the research literature. Conversely 12 months after onset, the recovery rate of 6.3% was strikingly low.

4 of the 12 predictors measured prior to stuttering onset had a significant association with starting to stutter at age 3. Gender is a well-documented predictor of stuttering and this was confirmed in the present study; undoubtedly being a boy increases risk of stuttering onset. Twins and children with mothers with a degree or postgraduate qualification were higher in the stuttering group although these results should be interpreted with caution. As discussed by Reilly et al. 2009, the number of twins who started to stutter was small (N = 7). Twinning was a rarely occurring risk factor and the confidence intervals allowed for a wide range of effects. In regards to maternal education, it is feasible that mothers with higher education were better at identifying and reporting their child’s stuttering. In addition to this, 3 word utterances may have developed sooner in the children of these mothers. Notably, while having a larger vocabulary was a predictor of stuttering at 3 years, it was not at 4. It may have been that
stuttering simply presented earlier in those who used more complex utterances at a younger age. Nevertheless, the 12 predictors investigated accounted for very little of the variation in stuttering onset.

While extremes in data must always be approached with caution, discrepancies need considered within the context of the methodological shortcomings discussed in reference to earlier prospective community studies investigating stuttering epidemiology (see Ingham, 1976; Mansson, 2000). A unique strength of the ELVS stuttering study research design was the recruitment of families prior to stuttering onset. This was crucial as it allowed us to clearly discern between predictors and effects of stuttering. This was particularly salient in regards to temperament, which has long been debated as possible cause of stuttering. In a recent review of the literature on temperament and stuttering, Kefalianos and colleagues discussed two hypotheses, namely if unusual temperament was a cause or effect of stuttering. The authors concluded that the only way the question would be answered was with “longitudinal studies with a cohort ascertained prior to the onset of stuttering” (Kefalianos, Onslow, Block, Menzies, & Reilly, 2012). The prospective design of ELVS stuttering contributed unique data to the literature by showing that temperament was not associated with stuttering onset. A further benefit of the prospective design was that it allowed for parent education on specific stuttering behaviours that might emerge, so parents knew what to look for. As a result, stuttering that was mild in its presentation or was only apparent for a short period before resolving may have been more readily identified in the present study (Reilly et al., 2009). Careful consideration to attrition was also important element. Potential bias was acknowledged and accounted for. Mothers with a degree or a post-graduate qualification were more highly represented in the participant group than the non-participant group. An analysis taking this factor into consideration showed only a 1% difference in cumulative incidence at 3 years (7.5%), which is still higher than previously reported and therefore did not alter conclusions. Finally, rigorous care was taken to ensure an accurate diagnosis of stuttering, which is integral to reliably establishing incidence.

Significance
Understanding the epidemiology of stuttering has both theoretical and clinical implications (Yairi & Ambrose 2005). Working towards understanding the cause of stuttering will help inform treatment practices. Findings relating to the timing of onset and the stage of language development favour the current, well-accepted multifactorial causal theory of stuttering, the Packman and Attanasio 3-factor causal model (P&A model). Packman 2012 discusses the role of possible neural processing deficits in stuttering, that become apparent after the first few years of life. This coincides with the age of stuttering onset seen in this study. Rather than impaired language, the P&A model identified increased language demands as a possible trigger for stuttering. This notion was also supported with findings showing that children with larger vocabularies at 3 started stuttering earlier.
Regarding the clinical significance, stuttering therapy for pre-schoolers often requires a considerable investment of time, money and resources from both families and service providers. With this in mind, determining who will benefit most from treatment from those who will recover naturally is crucial. Findings from this study showing only 6.3% of children recovered after 12 months suggest the current recommendation to allow six months for natural recovery may need to be revised. While clear risk factors for stuttering were not established, identifying factors that are not related to stuttering onset are of significant consequence. The experience of having a child start to stutter can be a worrying phenomenon for parents, particularly as onset can occur without warning, and where there was previously fluent speech and language development appeared normal (Yairi & Ambrose, 2005). Feelings of guilt, anxiety and uncertainty are common. Clinicians can reassure parents that stuttering in the preschool years is not unusual and does not mean their child is more shy or withdrawn. In addition to this, there was no strong evidence that a range of other social and environmental factors measured were related to stuttering onset (Reilly et al., 2009).

Future directions
Data collected from the ELVS stuttering cohort provides a unique opportunity to address a number of ongoing challenges. While continuing to document the natural history of stuttering in a community sample, the following questions will be addressed:

- Is there an association between language development and stuttering in children with persistent stuttering?
- Do any predictors associated with natural recovery emerge as the children age? Can we determine which children will benefit most from treatment and which ones will recover on their own?
- What is the relationship between anxiety and stuttering in children with stuttering? Does onset of anxiety occur in childhood and what is the trajectory of this disorder in stuttering?

Findings will inform clinical practice, help determine if specialised programs for sub-groups of children with stuttering are warranted and contribute to our overall understanding of the disorder of stuttering.

CONCLUSION
In order to address some of the long debated epidemiological questions about stuttering, there was a critical need for a prospective longitudinal study using a community cohort. Utilizing a rigorous methodology, including measuring putative risk factors prior to stuttering onset, the ELVS stuttering study succeeded in providing robust findings on onset, predictors and recovery. Onset of stuttering was higher at age 3 and 4 than previously reported. This combined the low recovery rate 12 months after onset, presents a difficult predicament
for clinicians when deciding if and when to intervene with stuttering therapy. It remains to be seen if more children will recover in the second and third year after onset and this will further influence the decision making process.

Some previously made assumptions about stuttering have been rejected, including those regarding temperament, language difficulties and a range of other social and environmental factors causing stuttering onset. It is of great importance to continue to monitor these factors in children in whom stuttering persists. As the ELVS data continues to be analysed, we can expect to shed further light on the trajectory and impact of stuttering and ultimately work towards achieving the best possible prognosis for children with this disorder.

**Reference List**


