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Natural History of Stuttering to 4 Years of Age: A Prospective Community-Based Study



WHAT'S KNOWN ON THIS SUBJECT: Stuttering is extremely common, with 8.5% of children affected by age 3 years in a prospective community-ascertained cohort of Australian children. The natural history and comorbidities of early stuttering are uncertain at the population level.



WHAT THIS STUDY ADDS: The cumulative incidence of stuttering was 11% by 4 years. Stuttering children were similar in temperament and social-emotional development but had better verbal and nonverbal skills than their peers. Recovery from stuttering was low; 6.3% 12 months after onset.

AUTHORS: Sheena Reilly, PhD,^{a,b} Mark Onslow, PhD,^c Ann Packman, PhD,^{a,c} Eileen Cini,^a Laura Conway,^a Obioha C. Ukoumunne, PhD,^d Edith L. Bavin, PhD,^e Margot Prior, PhD,^f Patricia Eadie, PhD,^g Susan Block, PhD,^h and Melissa Wake, MD^{a,b}

^aMurdoch Childrens Research Institute, Victoria, Australia; ^bThe University of Melbourne Department of Paediatrics at the Royal Children's Hospital, Melbourne, Australia; ^cAustralian Stuttering Research Centre, The University of Sydney, Sydney, Australia; ^dPenCLAHRC, University of Exeter Medical School, University of Exeter, Devon, United Kingdom; ^eSchool of Psychological Science, and ^fSchool of Human Communication Sciences, La Trobe University, Melbourne, Australia; ^gSchool of Behavioural Sciences, The University of Melbourne and The Royal Children's Hospital, Melbourne, Australia; and ^hDepartment of Audiology and Speech Pathology, The University of Melbourne, Melbourne, Australia;

KEY WORDS

stuttering, longitudinal study, risk factors, child, preschool, epidemiological studies

ABBREVIATIONS

CI—confidence interval
ELVS—Early Language in Victoria Study
SEIFA—Socio-Economic Indexes for Areas

Dr Reilly conceptualized and designed the study, drafted the initial manuscript, and approved the final manuscript as submitted; Dr Reilly had full access to all of the data in the study takes responsibility for the integrity of the data and the accuracy of the data analysis and is the guarantor; Drs Onslow, Packman, Eadie, Bavin, Wake, Prior, and Block contributed to planning, reviewing, and editing the manuscript; Dr Ukoumunne provided statistical advice and assisted with analysis; and Ms Conway and Ms Cini analyzed the data.

(Continued on last page)

abstract

FREE

OBJECTIVES: To document the natural history of stuttering by age 4 years, including (1) cumulative incidence of onset, (2) 12-month recovery status, (3) predictors of stuttering onset and recovery, and (4) potential comorbidities. The study cohort was a prospective community-ascertained cohort (the Early Language in Victoria Study) from Melbourne, Australia, of 4-year-old children ($n = 1619$; recruited at age 8 months) and their mothers.

METHODS: Outcome was stuttering onset by age 4 years and recovery within 12 months of onset, defined using concurrent monthly parent and speech pathologist ratings. Potential predictors: child gender, birth weight, birth order, prematurity, and twinning; maternal mental health and education; socioeconomic status; and family history of stuttering. Potential comorbidities: preonset and concurrent temperament, language, nonverbal cognition, and health-related quality of life.

RESULTS: By age 4 years, the cumulative incidence of stuttering onset was 11.2% (95% confidence interval [CI]: 9.7% to 12.8%). Higher maternal education ($P = .004$), male gender ($P = .02$), and twinning ($P = .005$) predicted stuttering onset. At outcome, stuttering children had stronger language (mean [SD]: 105.0 [13.0] vs 99.6 [14.6]; mean difference 5.5, 95% CI: 3.1 to 7.8; $P < .001$) and nonverbal cognition (mean [SD]: 106.5 [11.4] vs 103.9 [13.7], mean difference 2.6, 95% CI: 0.4 to 4.8; $P = .02$) and better health-related quality of life but were otherwise similar to their nonstuttering peers. Only 9 of 142 children (6.3%; 95% CI: 2.9% to 11.7%) recovered within 12 months of onset.

CONCLUSIONS: Although stuttering onset is common in preschoolers, adverse affects are not the norm in the first year after onset. *Pediatrics* 2013;132:460–467

The disorder of stuttering can have major long-term impacts on quality of life, causing educational and occupational underachievement, psychiatric illness including social phobia, and a lifetime of impaired communication.¹⁻⁷ There is limited information about onset and recovery rates to inform practitioners about evidence-based service delivery for preschool children who stutter or to counsel their parents about the early stages of the disorder.

Recently, we reported 8.5% of our large Australian community-ascertained sample to have commenced stuttering by age 3 years.⁸ This rate was higher than previously reported,⁹⁻¹¹ presumably because our study began in infancy before speech development, and we identified many early cases missed by other studies that did not assess children until they were 3 years of age or older.⁹ Although many antecedents and risk factors for stuttering have been proposed,¹¹⁻¹⁴ we did not find early stuttering onset to be associated with any social or environmental factors. Only 3.7% of the variation in stuttering onset was explained in our multivariable models.⁸ In particular, shy temperament was not a risk factor for stuttering onset. Furthermore, children who started to stutter early tended to have larger expressive vocabularies.⁸

Early intervention is efficacious and highly desirable for those destined for persistent stuttering.^{15,16} However, it is unlikely that there would ever be sufficient health care resources to treat all the 8.5% of children who are stuttering by age 3 years, nor that such treatment would be needed because many children recover naturally.^{11,13} To optimize service delivery, therefore, currently absent information is needed about recovery from early stuttering and its predictors. Recovery rates vary between studies according to when recovery was determined and sample

ascertainment.^{9-11,13,14,17} In a community-ascertained study in the United Kingdom, 42% of children were deemed recovered by 6 years of age and 79% by 16 years of age.⁹ A Danish community study reported 71.4% of children to have recovered by 5 years of age.¹⁰ Other studies where children were referred from the clinic or the community, such as the well-known Illinois studies,^{11,13,14} reported recovery rates of 9% at 1 year after onset rising to 72% at 4 years after onset. Potential recovery predictors include being female,^{11,13} having better speech and language skills,^{11,13,17} a decrease in stuttering over time,^{11,13,17} and having a family history of recovery.^{13,14,17,18} Currently, little is known about the social and emotional development of children who stutter, with available information drawn from clinic-referred samples of older children or adults.

Population data are urgently needed to inform practitioners about those children in whom stuttering is most likely to persist and require treatment. Here, we present such data for children with onset up to age 4 years in a prospective community-ascertained sample of children growing up in Melbourne, Australia, on whom we have previously reported stuttering onset to 3 years.⁸ We have repeatedly collected data about communication, language development, and stuttering from those children.^{8,19,20} We aimed to document the natural history of stuttering in those children: (1) cumulative incidence of stuttering onset, (2) 12-month recovery status, (3) predictors of onset and recovery, and (4) potential comorbidities.

METHODS

Overview of the Early Language in Victoria Study

The Early Language in Victoria Study (ELVS) commenced in 2002 in Melbourne (population 3.9 million in 2008),

Australia.^{8,19,20} A community sample of 1910 infants aged 7.5 to 10.0 months was recruited from 6 local government areas between September 2003 and April 2004. Infants were selected by ranking 31 Melbourne local government areas according to the Australian census-based Socioeconomic Indexes for Areas (SEIFA) Index for Relative Socioeconomic Disadvantage, then dividing into tertiles and selecting 2 noncontiguous areas from each.²¹

Previous publications detail the recruitment processes.^{8,19,20} Briefly, infants were recruited from the Victorian Maternal and Child Health service, which is a state government nursing service for families with children aged 0 to 6 years, supplemented by recruitment from 7- to 9-month hearing screening sessions and local newspaper publicity. We excluded children with a serious disability or developmental delay, such as Down syndrome or cerebral palsy. Data were collected annually by questionnaire from age 1 year, plus face-to-face assessments at age 4 years.

Procedures

After they returned the 2-year-old questionnaire, all ELVS parents were sent (1) an invitation letter to the nested Stuttering Study,⁸ (2) a fridge magnet defining and illustrating stuttering behaviors (see Appendix), and (3) an "opt-out" letter. Every 4 months for 12 months, a reminder letter was sent to all families who had not opted out, asking them to telephone if their child displayed any stuttering behaviors.

When a parent telephoned, a speech pathologist clarified the nature of the child's speech disruptions and arranged a 45-minute home visit unless it was clear that the speech behaviors described were not stuttering. At the home visit, parents clarified the onset and characteristics of stuttering and gave a detailed family history of stuttering.

During a video-recorded 25-minute play session, parents were then instructed to play with their child as they normally would, using a standard set of toys, pictures, and questions.

Measures

The following main outcome measures were established.

Stuttering Onset

Stuttering onset (age 2–4 years) was the main outcome measure. Stuttering presence was verified and stuttering severity rated using the Lidcombe Program 10-point stuttering severity scale (1 = “no stuttering,” 2 = “extremely mild stuttering,” and 10 = “extremely severe stuttering”).^{15,16,22} If there was uncertainty, a consensus panel of clinical experts in stuttering independently determined presence or absence of stuttering.⁸

Stuttering Recovery

After verification, the speech pathologist arranged monthly home visits for 12 months, during each of which the video-recorded play session was repeated. The parent and speech pathologist both assigned stuttering severity scores at each of these sessions. Children were classified as “recovered” if, for the final 3 visits, parent and speech pathologist severity ratings concurred in including at least 2 scores of 1 and no scores >2. Children were classified as “not recovered” if neither the parent nor the speech pathologist severity ratings met these criteria. Children were classified as “recovery status not determined” if parent and speech pathologist ratings were discordant for recovery or if the data were missing.

Predictors and Risk Factors for Stuttering Outcomes

A literature review, incorporating the US Preventive Services Task Force

systematic review examining preschool speech and language delay (including stuttering),²³ identified 12 putative risk factors: male gender (1), perinatal factors (2–4; twin birth, preterm birth, and birth weight), birth order (5), family history of speech and language problems (6), parental education (7), maternal age (8), and minority status, and (9) indicated by non-English speaking background. Socioeconomic status (10) was indicated by the SEIFA Index of Relative Disadvantage²¹ at the smallest geographic unit possible (census collection district). SEIFA scores are standardized for the Australian population to a mean of 1000 (SD 100), with lower scores indicating greater disadvantage.²¹ Maternal mental health (11) was measured at child age 12 months with the Nonspecific Psychological Distress Scale,²⁴ dichotomized as “likely mental health problem” (a score of ≥ 4 of a possible 24) or “no mental health problem” (score <4). Maternal vocabulary (12) was measured at Wave 2 (age 12 months) using the written 44-item modified version of the Mill Hill Vocabulary Scale,²⁵ tallying correct answers to provide a raw quantitative score with a possible maximum of 44.

At age 24 months, parents completed the Words and Sentences version of the MacArthur-Bates Communicative Development Inventory.²⁶ With author permission, we substituted 24 vocabulary items to accommodate Australian usage (eg, “footpath” for “sidewalk”). Raw (quantitative) scores and percentile ranks were calculated for vocabulary production. Children were classified as “late talkers” if their scores were below the 10th centile based on gender-specific norms (<119 words for girls and <79 words for boys).²⁶ Parents also completed the Communication and Symbolic Behavior Scales Infant-Toddler Checklist, yielding a standardized total score (normative mean 100, SD 15).²⁷

Additional putative stuttering-specific predictors of recovery were (1) stuttering severity after onset, calculated as the mean of the severity ratings for the first 3 monthly visits after onset,²⁸ calculated separately for research assistant and parent ratings; (2) parent report of the frequency (“at least sometimes” vs “less than sometimes”) of repetition of sounds, word parts, words and phrases; (3) rapidity of stuttering onset (within 1 day vs over a longer period); and (4) whether stuttering reportedly occurred episodically or continuously.

Potential Comorbidities (4 Years)

Research assistants individually assessed all children at age 4 years using the Australian adaptation of the Clinical Evaluation of Language Fundamentals-Preschool, Second Edition.²⁹ This test yields a standardized composite score and receptive and expressive scores, each with a mean of 100 and SD of 15. The Matrices subtest of the Kaufman Brief Intelligence Test, Second Edition³⁰ provided an estimate of nonverbal cognitive abilities.

Parents reported on their children’s behavior, temperament, and health-related quality of life in the 4-year-old questionnaire. The 25-item Strengths and Difficulties Questionnaire, a behavioral screening questionnaire for 4- to 10-year-olds,³¹ yields 4 subscale scores plus a Total Difficulties score. From the 30-item Short Temperament Scale for Children,³² we derived the Temperament Easy-Difficult Scale from the mean score of the approach, inflexibility, persistence subscales. The 23-item Pediatric Quality of Life Inventory Parent-Proxy Report³³ incorporates physical, emotional, and social well-being as well as a preschool dimension, generating Physical and Psychosocial summary scores and a Total scale score.

Sample Size and Analysis

After allowing for loss to follow-up in the main ELVS study, we anticipated that 1530 children would provide data up to 4 years of age in the ELVS stuttering substudy. We assumed that ~5% of these children would stutter by 4 years, giving 75 stutterers and 1455 non-stutterers. This sample size is large enough to detect a difference of a third of a SD (effect size 0.33) on continuous outcomes between the stuttering children and the typically developing fluent children with 80% power at the 5% level of significance.

The cumulative incidence of stuttering onset and 12-month recovery status were reported as percentages (Aims 1 and 2). Logistic regression was used to describe relationships between putative child, family, and/or environmental factors (potential predictors) and the 2 stuttering outcomes (Aim 3), presenting crude (unadjusted) odds ratios and adjusted odds ratios from models in which all potential predictors are used simultaneously. Scores for children born prematurely, defined as <36 weeks' gestation, were age-corrected before analysis. *t* tests were used to compare the stuttering and nonstuttering groups on potential comorbidities at age 4 years (Aim 4). Analyses were implemented using Stata.³⁴

Ethical approval was obtained from the Royal Children's Hospital Human Research Ethics Committee (# 23018). All parents provided written, informed consent.

RESULTS

Of the 1910 ELVS children, 1619 (85%) participated in the stuttering study, with 234 opting out and the remaining 57 already lost by the 2-year-old follow-up. Stuttering onset was confirmed in 181 children by age 4 years (cumulative incidence 11.2%; 95% confidence interval CI: 9.7% to 12.8%), with onset

noticeably slowing after 3.6 years (Fig 1).

Recovery status could not be reliably established for 39 of the 181 children because parent and/or researcher ratings were missing at ≥ 1 of the 3 times of interest. Of the remaining 142, only 9 children (6.3%; 95% CI: 2.9% to 11.7%) were classified as recovered within 12 months of stuttering onset.

Table 1 describes the characteristics of children with and without stuttering onset by age 4 years, whereas Table 2 summarizes the relationships between the putative predictors and stuttering onset. In the adjusted model, being a twin, male gender, having a mother educated to degree level, and higher Communication and Symbolic Behavior Scales score at 2 years all predicted higher rates of stuttering onset. The square Pearson correlation measure³⁵ indicated that 3.3% of the variability in stuttering onset status was explained by the model. Although as previously reported a higher expressive vocabulary score at 2 years predicted stuttering onset by 3 years,⁸ it did not predict cumulative onset by 4 years.

Rates of recovery within 12 months were higher in boys than girls (10% [9 of 88] vs 0% [0 of 54]; $P = .01$) and in those who did not repeat whole words at onset than those who did (15% [6 of 41] vs 3% [3 of 101]; $P = .02$). Compared with children who did not recover, the mean parent stuttering severity score of those who recovered, across the first 3 home visits, was 0.8 U lower (95% CI: 0.1 to 1.5; $P = .03$). There was no evidence at the 5% level of significance that any of the other putative predictors were associated with recovery status. Of the 9 children who recovered, 4 sought professional help (3 from a speech pathologist) during the previous 12 months.

Table 3 shows that, at age 4 years, the stuttering and nonstuttering groups had similar scores on the Strength and Difficulties Questionnaire temperament scales (Easy-Difficult and Approach/Withdrawal). However, stuttering children had higher scores than their nonstuttering counterparts on receptive and expressive language scores, nonverbal cognition and the social and preschool dimensions of health-related quality of life.

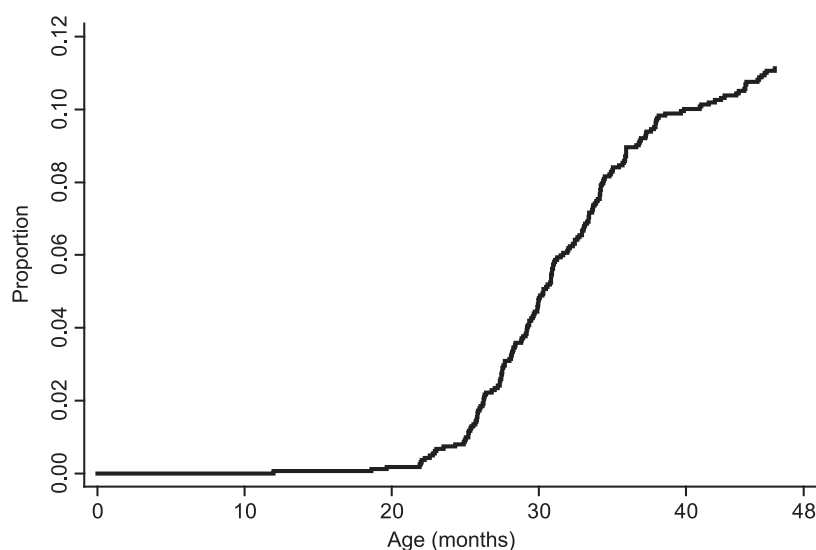


FIGURE 1

Proportion of study participants whose parents reported they stuttered by a given age. Data shown up to age 4 years.

TABLE 1 Characteristics of Children With Stuttering Onset Compared With Nonstuttering Children

Variable	<i>n</i>	Stutterers ^a	<i>n</i>	Nonstutterers
Male gender, %	181	58.6	1438	49.9
Premature birth (<36 wk), %	181	3.3	1438	2.9
Twin birth, %	181	6.1	1438	2.2
Has older siblings, %	181	44.2	1434	49.9
Family history	181		1438	
No problem, %		76.8		74.8
Speech/language/reading problems only, %		14.9		19.7
Stuttering problems, % ^b		8.3		5.5
Birth wt, kg, mean (SD)	179	3.4 (0.5)	1414	3.4 (0.5)
SEIFA disadvantage score, mean (SD)	181	1047 (47)	1437	1035 (62)
Mother's education level	179		1423	
Did not complete Year 12, %		16.2		23.2
Completed Year 12, %		30.7		40.3
Degree/postgraduate, %		53.1		36.5
Maternal mental health score at 2 y, median (IQR)	176	2 (1, 4)	1293	3 (1, 4)
Temperament score at 2 y, mean (SD)	178	15.6 (4.6)	1311	15.7 (4.9)
CDI raw vocabulary score at 2 y, mean (SD); median (IQR)	179	284 (157); 261 (168, 398)	1295	261 (163); 247 (127, 377)
CDI percentile rank at 2 y, median (interquartile range)	179	40 (26, 68)	1295	38 (16, 63)
Late talker (score ≤10th percentile on MCDI) at 2 y, %	179	11.7	1295	19.9
CSBS total standardized score at 2 y, mean (SD)	174	107.7 (15.2)	1273	104.4 (14.6)
Language spoken to child—w1 (8 mo) to w4 (36 mo)	181		1438	
English only, %		97.8		93.2
Another language, %		2.2		6.8

CDI, MacArthur-Bates Communicative Development Inventory; CSBS, Communication and Symbolic Behavior Scales; IQR, interquartile range.

^a Proportion of participants with stuttering onset by 4 y (cumulative incidence) was 11.2% (95% CI 9.7% to 12.8%).

^b Children with a family history of stuttering may also have reported family histories of speech/language and/or reading problems.

DISCUSSION

This prospective, community-ascertained cohort study has clarified 3 important unknowns about the population epidemiology of early childhood stuttering. First, the cumulative incidence of stuttering onset by 4 years of age was, at 11.2% (95% CI: 9.7% to 12.8%), more than twice that previously reported and justifies our choice to ascertain onset prospectively and continuing stuttering by monthly visits. Second, these children showed little evidence of harm to their mental health, temperament, or psychosocial health-related quality of life and on average displayed better receptive and expressive language and nonverbal intelligence at outcome than their nonstuttering counterparts.

A strength of this study is that, uniquely, findings were derived prospectively from a community-ascertained cohort, with a broad range of putative risk and protective factors measured before stuttering onset. Parent reports of stuttering were validated with reliable,

clinical expert diagnoses. Rigorous inclusion criteria were adopted, and cases of ambiguous stuttering were excluded. Persistence and resolution were rigorously

monitored from ascertainment with a monthly standardized home visit; something never before attempted. Our low recovery rate in the first year after

TABLE 2 Logistic Regression of Variables Predicting Stuttering Status (Stuttering Onset vs Not Stuttering) by 4 Years

Variable	Unadjusted (<i>n</i> ranges from 1447 to 1619)				Adjusted (<i>n</i> = 1355)		
	N	OR	95% CI	<i>P</i>	OR	95% CI	<i>P</i>
Male gender	1619	1.42	1.04 to 1.94	.03	1.51	1.07 to 2.13	.02
Premature birth (<36 wk)	1619	1.14	0.48 to 2.72	.77	1.15	0.42 to 3.15	.78
Twin birth	1619	2.94	1.45 to 5.95	.003	3.26	1.44 to 7.40	.005
Birth wt, kg	1593	0.99	0.74 to 1.33	.96	0.97	0.67 to 1.40	.88
Has older siblings	1615	0.80	0.58 to 1.09	.15	0.81	0.58 to 1.14	.23
Family history	1619			.13			.07
No problem		ref			ref		
Speech/language/reading problems only		0.73	0.48 to 1.13		0.82	0.52 to 1.31	
Stuttering problem		1.47	0.82 to 2.62		1.90	1.01 to 3.57	
SEIFA disadvantage score (per 100 unit increase)	1618	1.44	1.08 to 1.93	.01	1.14	0.83 to 1.55	.43
Mother's education level	1602			<.001			.004
Did not complete Year 12		ref			ref		
Completed Year 12		1.09	0.68 to 1.75		0.99	0.59 to 1.64	
Degree/postgraduate qualification		2.08	1.34 to 3.22		1.75	1.08 to 2.85	
Maternal mental health score	1469	0.99	0.93 to 1.04	.60	1.01	0.95 to 1.07	.81
Temperament: approach/withdrawal at 2 y	1489	1.00	0.96 to 1.03	.78	1.01	0.98 to 1.04	.62
CSBS total score at 2 y (per 15 unit increase)	1447	1.26	1.07 to 1.47	.005	1.22	1.00 to 1.48	.05
CDI raw expressive vocabulary score at 2 y (per 100 unit increase)	1474	1.09	0.99 to 1.20	.08	1.05	0.94 to 1.19	.37

CDI, Communicative Development Inventory; CSBS, Communication and Symbolic Behavior Scales; OR, odds ratio; ref, reference.

TABLE 3 Four-Year-Old Outcomes of Children by Stuttering Status (Stuttering Onset vs Not Stuttering) Compared by *t* Tests

Variable	Stuttering		Never stuttering		Mean difference (stuttering minus never-stuttering)		
	<i>n</i>	Mean (SD)	<i>n</i>	Mean (SD)	Difference	95% CI	<i>P</i>
CELF-P2							
Core language score	161	105.0 (13.0)	1249	99.60 (14.6)	5.5	3.1 to 7.8	<.001
Expressive language composite	159	104.4 (14.0)	1219	99.7 (15.0)	4.6	2.2 to 7.1	<.001
Receptive language composite	160	103.1 (13.4)	1247	96.5 (14.9)	6.6	4.2 to 9.0	<.001
SDQ							
Emotional symptoms scale	165	1.6 (1.6)	1237	1.4 (1.5)	0.2	−0.05 to 0.5	.11
Conduct problems scale	165	1.6 (1.4)	1237	1.6 (1.5)	0.03	−0.3 to 0.2	.84
Hyperactivity scale	165	3.3 (2.4)	1237	3.3 (2.2)	−0.09	−0.4 to 0.3	.64
Peer problems scale	165	1.2 (1.3)	1237	1.4 (1.5)	−0.2	−0.4 to 0.1	.18
Prosocial scale	165	7.2 (2.0)	1237	7.5 (1.8)	−0.2	−0.5 to 0.1	.11
Total Difficulties scale	165	7.7 (4.7)	1237	7.8 (4.6)	−0.08	−0.8 to 0.7	.84
Temperament							
Approach/Withdrawal score	164	20.8 (6.0)	1234	20.1 (6.7)	0.7	−0.4 to 1.8	.19
Easy-Difficult score	163	67.9 (12.5)	1232	68.0 (12.7)	−0.04	−2.1 to 2.0	.97
PEDS-QL							
Physical functioning scale	165	88.0 (10.0)	1234	87.4 (10.6)	0.6	−1.1 to 2.3	.49
Emotional Functioning	165	73.4 (13.9)	1233	75.5 (14.5)	−2.1	−4.5 to 0.2	.08
Social Functioning	165	91.7 (11.0)	1234	87.2 (12.8)	4.5	2.5 to 6.6	<.001
School/Preschool	130	93.7 (11.2)	984	91.0 (13.2)	2.7	0.3 to 5.1	.03
Total scale score	165	85.9 (8.6)	1234	84.8 (9.5)	1.1	−0.4 to 2.7	.14
K-BIT Matrices subtest Standard Score	160	106.5 (11.4)	1247	103.9 (13.7)	2.6	0.4 to 4.8	.02

CELF-P2, Clinical Evaluation of Language Fundamentals—Preschool, Second Edition; K-BIT, Kaufman Brief Intelligence Test; PEDS-QL, Pediatric Quality of Life Inventory; SDQ, Strengths and Difficulties Questionnaire.

onset, at just 6.3%, was in line with the 9% 12-month recovery rates previously reported in the Illinois study.¹¹

The study also has limitations. Some cases may have been missed because parents opted out of the study at earlier waves, were lost to follow-up, or overlooked the stuttering (eg, if it was mild or if they had another child who had recovered from stuttering). More children may yet present beyond age 4 years, although, because incidence slowed markedly after ~3.5 years, these will likely be relatively few.⁸ For all these reasons, the true cumulative incidence may therefore be even higher than we report. Regarding cohort bias, we acknowledge that there were fewer children from disadvantaged families and more children from families with more highly educated mothers.⁸ One might speculate that the more highly educated mothers of stuttering children might be more vigilant and more likely to report mild and transient bouts of stuttering. If, however, the reverse is true and onset was

underreported by less well-educated mothers, this would likely result in an even higher onset percentage than we report. Finally, we could not examine predictors of recovery because only 9 children recovered; this will be the subject of future population-based research as the ELVS cohort ages.

As at age 3 years in this same cohort, stuttering onset by age 4 was associated with twinning, male gender (in line with previous studies) and higher maternal education.⁸ However, these factors jointly continue to explain little variation in stuttering onset in the regression models. The small number of twins in the cohort suggests caution in interpreting these findings because odds ratio estimates may be unstable.

The temporal relationships we report here are particularly novel and intriguing. To stutter, a child needs to be talking. Therefore, it makes sense that children with more developed expressive language skills would stutter earlier than those with relatively poorer expressive language skills. Yet expres-

sive vocabulary at age 2 years only weakly predicted stuttering onset by age 3 years,⁹ and it did not predict stuttering onset by 4 years. In contrast, stuttering onset strongly predicted *subsequent* language at age 4 years, with measured receptive and expressive scores approximately one-third of an SD better in stutterers than non-stutterers. Together, the high incidence, better subsequent language outcomes, and absence of harm to any of mental health (Strengths and Difficulties Questionnaire), temperament, or psychosocial health-related quality of life (Pediatric Quality of Life Inventory)³³ by age 4 years could suggest that the presence of stuttering improves those outcomes. Alternatively, perhaps stuttering might be a “byproduct” of rapid language development between 2 and 4 years of age, rather than of early precocious language development at age <2 years. This is a period in which a child’s motor speech system is challenged to keep pace with the phenomenal rate of language acquisition. Finally, we

should consider that this is a chance finding or that there is some unidentified bias in our community sample, although the strength of the associations we report suggest not, and our population design should reduce the likelihood of bias.

CONCLUSIONS

These findings from a community-ascertained cohort refute long-held views suggesting that developmental stuttering is associated with a range of poorer outcomes. If anything, the reverse was true, with stuttering predicting subsequently better language, nonverbal skills, and psychosocial health-related quality of life at 4 years of age. Future research with this cohort will support a more complete longitudinal

understanding of when and in whom recovery occurs. Current best practice recommends waiting for 12 months before commencing treatment, unless the child is distressed, there is parental concern, or the child becomes reluctant to communicate. It may be that for many children treatment could be deferred even longer. Treatment is efficacious¹⁵ but is both intensive (median of 15.4 one-hour clinical sessions followed by 10 one-hour clinical maintenance sessions)^{36,37} and expensive^{38,39}; this “watchful waiting” recommendation would therefore help target allocation of scarce resources to the small number of children who do not resolve and experience adverse outcomes, secure in the knowledge that delaying treatment by a year or more has been shown not to compromise treatment efficacy.^{36,37}

APPENDIX: INFORMATION PROVIDED TO PARENTS ABOUT THE ONSET OF STUTTERING

Children who stutter have trouble getting their words out. Stuttering is when children

- Repeat words or syllables over and over (eg, “can—can—can—I go)
- Make long, prolonged sounds (eg, “caaaaaaaaaaaaaan I go”)
- Have speech “stoppages” or “blocks” where no sounds come out

Stuttering can start quite suddenly, or it can begin gradually over days, weeks, or months.

If you think your child is stuttering, please contact the Early Language in Victoria Study team as soon as you notice it.

REFERENCES

1. Craig A, Blumgart E, Tran Y. The impact of stuttering on the quality of life in adults who stutter. *J Fluency Disord.* 2009;34(2):61–71
2. Bricker-Katz G, Lincoln M, McCabe P. A lifetime of stuttering: how emotional reactions to stuttering impact activities and participation in older people. *Disabil Rehabil.* 2009;31(21):1742–1752
3. Messenger M, Onslow M, Packman A, Menzies R. Social anxiety in stuttering: measuring negative social expectancies. *J Fluency Disord.* 2004;29(3):201–212
4. de Carle AJ, Pato MT. Social phobia and stuttering. *Am J Psychiatry.* 1996;153(10):1367–1368
5. Iverach L, Jones M, O'Brian S, et al. Screening for personality disorders among adults seeking speech treatment for stuttering. *J Fluency Disord.* 2009;34(3):173–186
6. Iverach L, O'Brian S, Jones M, et al. Prevalence of anxiety disorders among adults seeking speech therapy for stuttering. *J Anxiety Disord.* 2009;23(7):928–934
7. Iverach L, Jones M, O'Brian S, et al. The relationship between mental health disorders and treatment outcomes among adults who stutter. *J Fluency Disord.* 2009;34(1):29–43
8. Reilly S, Onslow M, Packman A, et al. Predicting stuttering onset by the age of 3 years: a prospective, community cohort study. *Pediatrics.* 2009;123(1):270–277
9. Andrews G, Harris M. *The Syndrome of Stuttering.* London, UK: Heinemann; 1964
10. Månsson H. Childhood stuttering: incidence and development. *J Fluency Disord.* 2000;25(1):45–57
11. Yairi E, Ambrose N. *Early Childhood Stuttering.* Austin, TX: Pro Ed; 2005
12. Büchel C, Sommer M. What causes stuttering? *PLoS Biol.* 2004;2(2):E46
13. Yairi E, Ambrose NG, Niermann R. The early months of stuttering: a developmental study. *J Speech Hear Res.* 1993;36(3):521–528
14. Ambrose NG, Cox NJ, Yairi E. The genetic basis of persistence and recovery in stuttering. *J Speech Lang Hear Res.* 1997;40(3):567–580
15. Jones M, Onslow M, Packman A, et al. Randomised controlled trial of the Lidcombe programme of early stuttering intervention. *BMJ.* 2005;331(7518):659–661
16. Onslow M, Jones M, Menzies R, O'Brian S, Packman A. Stuttering. In: Sturmeyp P, Hersen M, eds. *Handbook of Evidence-Based Practice in Clinical Psychology.* Hoboken, NJ: Wiley; 2012:185–207
17. Yairi E, Ambrose NG, Paden EP, Throneburg RN. Predictive factors of persistence and recovery: pathways of childhood stuttering. *J Commun Disord.* 1996;29(1):51–77
18. Kloth SAM, Kraaimaat FW, Janssen P, Brutton GJ. Persistence and remission of incipient stuttering among high-risk children. *J Fluency Disord.* 1999;24(4):253–265
19. Reilly S, Wake M, Bavin EL, et al. Predicting language at 2 years of age: a prospective community study. *Pediatrics.* 2007;120(6). Available at: www.pediatrics.org/cgi/content/full/120/6/e1441
20. Reilly S, Wake M, Ukoumunne OC, Bavin EL, Prior M, Cini E. Predicting language outcomes at 4 years of age: findings from the Early Language in Victoria Study. *Pediatrics.* 2010;126(6). Available at: www.pediatrics.org/cgi/content/full/126/e1530
21. Australian Bureau of Statistics. *Socio-Economic Indexes for Areas.* Canberra, Australia: Australian Bureau of Statistics; 2001
22. Teesson K, Packman A, Onslow M. The Lidcombe Behavioral Data Language of stuttering. *J Speech Lang Hear Res.* 2003;46(4):1009–1015
23. Nelson HD, Nygren P, Walker M, Panoscha R. Screening for speech and language delay in preschool children: systematic evidence review for the US Preventive Services Task

- Force. *Pediatrics*. 2006;117(2). Available at: www.pediatrics.org/cgi/content/full/117/2/e298
24. Kessler R, Mroczek D. Final version of our non-specific psychological distress scale [memo]. Ann Arbor, MI: Institute for Social Research, 1994
 25. Raven JC. *Mill Hill Vocabulary Scale*. Oxford, England: Oxford Psychologists Press; 1997
 26. Fenson L, Dale PS, Reznick JS. *The MacArthur Communicative Development Inventories: User's Guide and Technical Manual*. San Diego, CA: Singular Publishing Group; 1993
 27. Wetherby A, Prizant B. *Communication and Symbolic Behavior Scales*. Baltimore, MD: Paul H. Brookes Publishing Co; 2002
 28. Onslow M, Jones M, O'Brian S, Menzies R, Packman A. Defining, identifying, and evaluating clinical trials of stuttering treatments: a tutorial for clinicians. *Am J Speech Lang Pathol*. 2008;17(4):401–415
 29. Wiig EH, Secord WA, Semel E. *Clinical Evaluation of Language Fundamentals—Preschool: Australian Standardised Edition*, 2nd ed. Sydney, Australia: Harcourt Assessment; 2006
 30. Kaufman AS, Kaufman NL. *Kaufman Brief Intelligence Test Manual*. Circle Pines, MN: American Guidance Service; 1990
 31. Goodman R. The strengths and difficulties questionnaire: a research note. *J Child Psychol Psychiatry*. 1997;38(5):581–586
 32. Sanson AV, Smart DF, Prior M, Oberklaid F, Pedlow R. The structure of temperament from three to seven years: age, sex and demographic influences. *Merrill-Palmer Q*. 1994;40:233–252
 33. Varni JW, Seid M, Rode CA. The PedsQL: measurement model for the pediatric quality of life inventory. *Med Care*. 1999;37(2):126–139
 34. *Stata Statistical Software: Release 9.2*. College Station, TX: Stata Corporation; 2005
 35. Hosmer D, Lemeshow S. *Applied Logistic Regression*. New York, NY: Wiley; 2000
 36. Kingston M, Huber A, Onslow M, Jones M, Packman A. Predicting treatment time with the Lidcombe Program: replication and meta-analysis. *Int J Lang Commun Disord*. 2003;38(2):165–177
 37. Jones M, Onslow M, Harrison E, Packman A. Treating stuttering in young children: predicting treatment time in the Lidcombe Program. *J Speech Lang Hear Res*. 2000;43(6):1440–1450
 38. Skeat J, Wake M, Ukoumunne OC, Eadie P, Bretherton L, Reilly S. Who gets help for pre-school communication problems? Data from a prospective community study [published online ahead of print March 22, 2013]. *Child Care Health Dev*. doi:10.1111/cch.120-32
 39. Skeat J, Eadie P, Ukoumunne O, Reilly S. Predictors of parents seeking help or advice about children's communication development in the early years. *Child Care Health Dev*. 2010;36(6):878–887

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Address correspondence to Sheena Reilly, PhD, Murdoch Childrens Research Institute, Flemington Rd, Parkville, Australia. E-mail: sheena.reilly@mcrci.edu.au

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Natural History of Stuttering to 4 Years of Age: A Prospective Community-Based Study

Sheena Reilly, Mark Onslow, Ann Packman, Eileen Cini, Laura Conway, Obioha C. Ukoumunne, Edith L. Bavin, Margot Prior, Patricia Eadie, Susan Block and Melissa Wake

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