Who to Refer for Speech Therapy at 4 Years of Age Versus Who to "Watch and Wait"?

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Objective To examine predictors of speech disorder resolution versus persistence at age 7 years in children with speech errors at age 4 years.

Study design Participants were drawn from a longitudinal, community cohort. Assessment at age 4 years (N = 1494) identified children with speech errors. Reassessment at age 7 years allowed categorization into resolved or persistent categories. Logistic regression examined predictors of speech outcome, including family history, sex, socioeconomic status, nonverbal intelligence, and speech error type (delay vs disorder).

Results At age 7 years, persistent errors were seen in over 40% of children who had errors at age 4 years. Speech symptomatology was the only significant predictor of outcome ($P = .02$). Children with disordered errors at age 4 years were twice as likely to have poor speech outcomes at age 7 years compared with those with delayed errors.

Conclusions Children with speech delay at age 4 years seem more likely to resolve, and this might justify a "watch and wait" approach. In contrast, those with speech disorder at age 4 years appear to be at greater risk for persistent difficulties, and could be prioritized for therapy to offset long-term impacts. *(J Pediatr 2017;185:200-4).*

Developmental speech disorders are common in preschool children, with estimates from community cohort studies suggesting 3.5% to 5% of 4-year-olds are affected.[1-3] Children with this condition have no known cause for their impairment (eg, no diagnosis of brain lesion, intellectual impairment, craniofacial or genetic disorder, or hearing impairment). The presence of a developmental speech disorder may be debilitating in itself, with associated psychosocial impacts, literacy difficulties, and restricted educational and employment outcomes longer-term.[4-7] Not surprisingly, given the prevalence of these disorders, parents frequently seek help from general practitioners (GPs) and pediatricians on this issue. However, population-based study of predictors of speech disorder recovery versus persistence into the middle school years to guide referral practice is lacking.[2]

A range of sociodemographic and early developmental factors are commonly hypothesized to predict speech outcome, including sex, socioeconomic status, maternal education, early language skills, early feeding skills, intelligence, and family history.[1,3,8-14] These findings are based largely on clinical studies[8-11] or derived from preschool children.[1] One population-based cohort has examined speech outcome into the middle school years; the Avon Longitudinal Study of Parents and Children (ALSPAC).[3,13,14] Methods involved resource-intensive transcription and analysis of connected speech.[3,13,14] Further to sociodemographic and cognitive-linguistic factors, a promising area of investigation for prediction of speech outcomes has been the predictive value of specific speech symptomatology (eg, type of speech error).[8,15] "Proportion" of speech errors was most predictive of poor outcomes at 8 years in the ALSPAC cohort.[14] Rating the proportion of errors alone is nonspecific for a child's speech diagnosis and does not inform intervention.

The most commonly used clinical diagnostic system denotes speech error patterns as being "delayed" or "disordered."[16] Speech error diagnosis is critical for intervention planning and prognosis.[17] A delay is an error that occurs in typical speech development but that is delayed relative to 90% of peers (eg, patterns that reduce consonant clusters such as "bue" for "blue." This error would be appropriate until 3 years 11 months but is considered as delayed in a 4-year 5-month old child).[18] Disordered speech is characterized by atypical errors seen in less than 10% of the typical population at any age (eg, backing sounds to more posterior mouth position such as "keddy" for "teddy"; removing initial consonants such as "og" for "dog").[18] Critically, the predictive value of speech error

ALSPAC	Avon Longitudinal Study of Parents and Children
GFTA-2	Goldman-Fristoe Test of Articulation-Second Edition
GPs	General practitioners
PCC	Percent consonants correct
SES	Socioeconomic status
SEIFA	Socioeconomic Indexes for Areas

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type (delay vs disorder) to inform prognosis remains unexplored in a longitudinal population or community cohort sample. Findings might guide practitioners regarding referral to speech therapy. Thus, this study examined children with speech errors at age 4 years, drawn from a longitudinal community cohort study of language and literacy, to determine predictors associated with speech recovery vs persistence at age 7 years.

Methods

Participants were recruited from a community cohort study, the Early Language in Victoria Study. Between September 2003 and April 2004, 1910 infants aged 7 to 10 months were recruited into Early Language in Victoria Study from 6 local government areas in Melbourne, Australia.[19] The government areas were selected from the census-based Socioeconomic Indexes for Areas (SEIFA) Index for Relative Socioeconomic Disadvantage, and included 2 from each of 3 tiers, representing low, middle, and high socioeconomic status (SES) communities. A higher SEIFA scores indicates greater advantage. Primary recruiters were a universal nursing service for families with infants through 6-year-olds, the Victorian Maternal and Child Health service. All eligible families were invited to participate. A small number of participants were recruited via local newspapers and at hearing screening sessions also offered by the universal nursing service. Children with known disabilities (eg, genetic syndromes, hearing loss) were excluded. At baseline, parents were required to have adequate English skills to complete written questionnaires. For further recruitment details see Reilly et al.[19] Ethics approval was obtained from the Royal Children's Hospital Human Research Ethics Committee (HREC#23018). Written consent was obtained from all parents.

At age 4 years, all eligible participants received face-to-face assessment of speech and language (N = 1494). Children with variables that could confound speech performance were excluded (ie, neurodevelopmental disabilities, genetic conditions, craniofacial disorders, or a non-English speaking background). Four years of age is a critical time for examining outcomes because it is a common age for GP or pediatrician referral of children with speech disorder.[2] Speech sound errors were identified in 160/1494 (11%), and each child was eligible for repeat assessment at age 7 years. Speech assessment data at ages 4 and 7 years were available for 93 participants. Reasons for loss to follow-up included participants declined further follow-up (54), lost to contact (4), unavailable for assessment (4) failure to complete the assessment, (1) and unusable audio recordings (4). No statistically significant differences were found at age 7 years between participants (n = 93) and nonparticipant (n = 67) groups on demographic variables including SEIFA, sex, family history, and nonverbal IQ (**Table I**; available at www.jpeds.com).

Procedures

Outcome. At ages 4 and 7 years, participants' speech production was examined using the standardized Goldman Fristoe Test of Articulation-Second Edition (GFTA-2)[20] Sounds-in-Words subtest, one of the most commonly used standardized speech tests in clinical practice.[21] Research assistants were formally trained in the GFTA-2 procedures and followed a set administration protocol. The Diagnostic Evaluation of Articulation and Phonology Inconsistency subtest was also administered at age 4.[22] Both the GFTA-2[20] and DEAP[22] require children to name single words in response to picture stimuli. Two experienced speech pathologists examined participants' productions of all words across both time points. Percent consonants correct (PCC) ratings were determined as a measure of intelligibility and severity of speech disorder at ages 4 and 7 years. At age 4 years, speech errors were independently categorized as delayed or disordered by 2 authors and confirmed with consensus rating. Mean agreement was 96.8%. Delay and disorder were defined as per Dodd et al.[18] At age 7 years, data were analyzed to determine whether speech errors had persisted or resolved.

Predictor Variables. Predictors commonly reported in the literature were examined, including sex, SES, nonverbal IQ, speech symptomatology (proportion denoted by percent consonants correct; error type of delay versus disorder), language ability, family history of speech disorder, and having had speech therapy[1,3,8-14] at some time between 4 and 7 years of age.

Statistical Analyses

Univariate group comparisons were conducted across resolved and persistent groups (**Table II**). Logistic regression was conducted with Stata v 13.1 software (StataCorp, College Station, Texas) to identify predictors of outcome at age 7 years. Predictors with statistically significant association ($P < .05$) from the univariate comparisons were included in the multivariable (adjusted) model, along with sex, nonverbal IQ, and SEIFA disadvantage index as covariates (**Table II**). Exploratory post hoc analyses were then conducted to determine whether any of the predictor variables significantly influenced resolved or persistent status in the "delayed" and "disordered" diagnostic groups at age 7 years (**Tables III** and **IV**; available at www.jpeds.com). This exploratory analyses requires conservative interpretation because of the risk of false positives attributable to the number of variables examined relative to the small sample size of the disordered group.

Results

At age 7 years, just under 60% of all children with speech errors at age 4 years had resolved (**Table V**). When examined according to type of speech error, over two-thirds (69.6%) of children with speech delay had resolved errors by age 7 years. By contrast, only two-fifths (40.5%) with disordered speech at age 4 years had resolved errors at age 7 years. Children with persistent errors at age 7 years did show some positive change in PCC over time but did not increase these proxy intelligibility ratings to an age appropriate level.[26] Children with delayed speech at age 4 years had a mean PCC of 77 (SD = 12), and

Table II. Results of association between predictors and resolved status at age 7 years (n = 93)

Predictors	Among those resolved	Among those with persistent errors	OR	95% CI	*P* value	aOR*	95% CI	*P* value
Male %	50.0 (27/54)	56.4 (22/39)	0.77	0.34, 1.77	.54	0.62	0.25, 1.53	.30
PCC at age 4 y, mean (SD)	80.17 (9.0)	74.9 (11.9)	1.05	1.01, 1.10	.03	1.04	0.99, 1.08	.12
Delay at age 4 y, compared with disorder %	72.2 (39/54)	43.6 (17/39)	3.36	1.41, 8.02	.006	3.01	1.18, 7.68	.02
SEIFA, mean (SD)	1040.0 (57.4)	1043.1 (54.5)	1.00	0.99, 1.01	.79			
Nonverbal IQ KBIT-2[23] at age 4 y, mean (SD)	107.6 (11.0)	105.3 (13.9)	1.02	0.98, 1.05	.38			
CTOPP-2[24] at age 5 y z score, mean (SD)	0.1 (0.9)	-0.2 (1.1)	1.30	0.80, 2.13	.29			
Family history at baseline, %								
No speech/language/reading problems	77.8 (42/54)	64.1 (25/39)	Ref					
Yes speech/language/reading problems	22.2 (12/54)	35.9 (14/39)	0.51	0.20, 1.28	.15			
Sought help for speech/seen by SLP	51.9 (28/54)	71.8 (28/39)	0.42	0.18, 1.02	.06			
CELF-IV[25] core language score at age 4 y, mean (SD)	100.7 (12.2)	96.1 (14.4)	1.03	0.99, 1.06	.10			

CELF-IV, Comprehensive Evaluation of Language Fundamentals-Fourth Edition; *CTOPP-2*, Comprehensive Test of Phonological Processing-Second Edition (blending words and sound matching); *KBIT*, Kaufman Brief Intelligence Test; *SLP*, speech-language pathologist.
CTOPP blending words and sound matching raw scores measured at age 5 years.
*Multivariate results of association between predictors (PCC, delay at age 4 years) and covariate (sex) and resolved status

this increased to 92 (SD = 5) at age 7 years. Children with speech disorder at age 4 years had a mean PCC of 69.5 (SD = 11), and this improved to an average PCC of 88 (SD = 8).

Univariate factors revealed significant differences between resolved and persistent groups at age 7 years on 2 variables only: percent consonants correct (ie, proportion of errors, $P = .03$) and type of speech error at age 4 (ie, delay compared with disorder, $P = .006$) (**Table II**). The variable of "sought help for/seen by speech-language pathology" approached significance ($P = .06$) but was not included in the multivariable analysis. Data were limited on whether children were assessed or received therapy, and no detail was provided on what type of therapy was applied. Eleven children (5 disordered and 6 delayed) attended speech pathology services for assessment and/or intervention between the ages of 4 and 7 years. Five of these children, all with delayed speech at age 4 years, had resolved by 7 years. The remainder of cases had persistent errors at age 7 years.

In the multivariable logistic regression analyses, only "type of error" (delay vs disorder) was a statistically significant predictor of speech error recovery vs persistence ($P = .02$, **Table I**). Further adjustment for IQ or SEIFA disadvantage index did not significantly change the results.

Exploratory post hoc analysis was conducted to explore predictors of resolved vs persistent status for the speech delay and speech disorder groups separately at 7 years of age. None of the variables examined predicted outcomes for either the delayed or disordered groups (**Tables III** and **IV**).

Table V. Speech outcomes for children assessed at age 4 and 7 years (n = 93)

Delay at age 4 y = 56 (60.2%)		Disorder at 4 = 37 (39.8%)	
Resolved at age 7 y	Persistent at age 7 y	Resolved at age 7 y	Persistent at age 7 y
39 (69.6%)	17 (30.4%)	15 (40.5%)	22 (59.5%)

Discussion

Our findings demonstrate that speech errors do not resolve for all children by 5 years of age, as previously suggested.[27] In fact, 40% of children with speech errors at age 4 years had persistent speech errors at age 7 years. The type of speech error (ie, delay vs disorder) at age 4 years remained the only significant predictor of whether a child belonged to the recovered compared with persistent outcome group at age 7 years. Other variables previously suggested predicting outcome, such as sex, SES, positive family history, language development, and nonverbal IQ were not significant. Our findings agree with another population-based speech outcome study that also covaried for sex and social status.[14] That study identified the symptom-based predictor of "proportion of speech errors" as significant, but not speech diagnosis as studied here.[14] Proportion of errors is time-consuming to calculate, nonspecific to a child's diagnosis, and hence, uninformative for intervention planning. Proportion of errors did not remain a significant predictor in our analyses. We have contributed new knowledge that type of error is more informative than proportion for outcome prediction and hence informing referral guidelines. Technically, we found that type of error is informative for predicting not only the presence of persistent speech errors at age 7 years, but also for resolution.

Of greatest clinical relevance, delayed speech errors at age 4 years were more likely to resolve, whereas children with disordered speech at age 4 years were twice as likely to have persistent speech errors. This finding has implications for policy and practice, regarding waitlist prioritization. Our data call us to question whether the "watch and wait" approach should be universally applied to all preschool children. Rather, these data suggest an efficient model may guide children with disorder at age 4 years to be fast-tracked for speech therapy and those with delayed errors could remain on a watch-and-wait list with 6-monthly surveillance (**Table VI**; available at www.jpeds.com). Although replication of findings is warranted, we provide some

empirical data for the GP/Pediatrician speech sound disorder referral guidelines proposed by Reilly et al.[2]

Reilly et al[2] suggested that children who were unintelligible to a parent or significant other at ≥3 years of age should be referred to speech therapy. Our data support this guideline. Children with disordered speech are less intelligible because the patterns they are using are aberrant and, hence, less predictable. By contrast, delayed error forms are more predictable and interpretable because these error patterns occur in typically developing younger children and are commonly heard in society. As further support for our findings, a recent study noted that being unintelligible to familiar others at 38 months was a risk factor for poorer speech outcome at age 8 years.[3] Here we refine and extend the recommendations of Reilly et al[2] by providing empirical data to inform triage and prioritization for speech intervention at 4 years of age (**Table VI**).

This longitudinal community cohort study examined predictors of speech recovery vs persistence, and used valid clinical assessments and diagnoses of speech disorder; ensuring our findings are generalizable to clinical practice. The only other population based studies on the ALSPAC cohort used labor-intensive transcription and analysis of connected speech.[3,13,14] Analysis of connected speech is arguably more sensitive for detecting errors, but also exponentially more time-consuming and not feasible for clinical practice. Further population-based studies are required to compare the differences, if any, in the ability of single word vs connected speech samples to predict later speech outcome. It is possible that our single measure of speech could be less sensitive to speech conditions in connected discourse and may have underestimated prevalence or persistence. A further limitation included attrition from ages 4 to 7 years, although no differences were identified between responders and nonresponders at age 7 years.

The ultimate goal is earlier prediction, yet whether speech errors at age 2 or even 3 years can reliably predict later outcome remains unexplored. We examined predictors of children at 4 years of age when their speech sound systems are known to be more stable[18] and at a time when children are commonly referred for speech therapy assessment and or intervention.[2] Further, although those with disordered errors were at greatest risk for poor longer-term outcomes, a significant proportion, one-third, of the delayed group did not resolve. None of the factors explored here explained these outcomes in the delayed group. Broader demographic, genetic, environmental, and symptom-based predictors should be examined in children who are delayed vs disordered to understand better resolved vs persistent status at age 7 years in each of these categories. In particular, data are critically required to better understand how many cases experienced natural resolution or persistence, vs potential influences of therapy.

Future work should explore the predictive value of more specific treatment variables not available here, such as what type and how much speech intervention was received, delivered by whom (eg, speech therapist or speech therapy assistant). ■

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Table I. Comparison of participants followed-up (n = 93) and opted-out (n = 67) at age 7 years

Factors	Among followed-up (mean [SD] or % [n/N])	Among dropped-out (mean [SD] or % [n/N])	*P* value
SEIFA disadvantage at baseline	1041.3 (55.9)	1036.4 (56.7)	.58
Nonverbal IQ (Kaufman-Brief Intelligence Test) at age 4 y	106.6 (12.3)	103.3 (13.1)	.10
Family history at baseline			.43
No speech/language/reading problems	72.0 (67/93)	77.6 (52/67)	
Speech/language/reading problems only	28.0 (26/93)	22.4 (15/67)	
Sex			.61
Male	52.7 (49/93)	56.7 (38/67)	
Female	47.3 (44/93)	43.3 (29/67)	

Table III. Results of association between predictors and resolved status at age 7 years among the delay cohort (n = 56)

Predictors	Among those resolved	Among those not resolved	OR	95% CI	*P* value
Male %	53.9 (21/39)	70.6 (12/17)	0.49	0.14, 1.64	.25
PCC at age 4 y, mean (SD)	81.7 (9.0)	78.1 (10.3)	1.04	0.98, 1.10	.20
Delay at age 4 y, compared with disorder %	n/a				
SEIFA, mean (SD)	1039.8 (56.7)	1053.6 (41.1)	0.99	0.98, 1.01	.37
IQ KBIT[23] at age 4 y, mean (SD)	107.3 (11.4)	105.9 (17.4)	1.01	0.97, 1.05	.73
CTOPP[24] z score,* mean (SD)	0.1 (0.9)	-0.1 (1.1)	1.20	0.59, 2.45	.61
Family history at baseline, %					
No speech/language/reading problems	79.5 (31/39)	64.7 (11/17)	Ref		
Yes speech/language/reading problems	20.5 (8/39)	35.3 (6/17)	0.47	0.13, 1.67	.25
Sought help for speech/seen by SLP	56.4 (22/39)	70.6 (12/17)	0.54	0.16, 1.83	.32
CELF-IV[25] core language score at age 4 y, mean (SD)	100.6 (12.1)	94.9 (15.0)	1.03	0.99, 1.08	.14

CELF-IV, Comprehensive Evaluation of Language Fundamentals-Fourth Edition; *CTOPP-2*, Comprehensive Test of Phonological Processing-Second Edition (blending words and sound matching); *KBIT*, Kaufman Brief Intelligence Test; *SLP*, speech-language pathologist.
*CTOPP blending words and sound matching raw scores measured at age 5 years.

Table IV. Results of association between predictors and resolved status at age 7 years among the disorder cohort (n = 37)

Predictors	Among those resolved	Among those not resolved	OR	95% CI	*P* value
Male %	40.0 (6/15)	45.5 (10/22)	0.80	0.21, 3.03	.74
PCC at age 4 y, mean (SD)	76.1 (7.8)	72.5 (12.7)	1.03	0.97, 1.11	.34
Delay at age 4 y, compared with disorder %	n/a				
SEIFA, mean (SD)	1040.4 (61.2)	1035.1 (62.7)	1.00	0.99, 1.01	.79
IQ KBIT[23] at age 4 y, mean (SD)	108.4 (10.4)	104.9 (10.8)	1.03	0.97, 1.10	.32
CTOPP[24] z score,* mean (SD)	0.1 (1.1)	-0.2 (1.2)	1.35	0.67, 2.70	.40
Family history at baseline, %					
No speech/language/reading problems	73.3 (11/15)	63.6 (14/22)	Ref		
Yes speech/language/reading problems	26.7 (4/15)	36.4 (8/22)	0.64	0.15, 2.68	.54
Sought help for speech/seen by SLP	40.0 (6/15)	72.7 (16/22)	0.25	.06, 1.01	.05
CELF-IV[25] core language score at age 4 y, mean (SD)	101.1 (12.7)	97.0 (14.3)	1.02	0.97, 1.08	.36

*CTOPP blending words and sound matching raw scores measured at age 5 years.

Table VI. Red flags guiding referral to speech therapy for preschool children

Red flags	Yes	No
Child cannot be understood by parent/caregiver at age 3-4 y		
Child cannot be understood by significant other at age 3-4 y		
Child demonstrating disordered speech errors*		
• Backing sounds to more posterior mouth position (eg, "keddy" for "teddy")		
• Replacement of stop sounds (p,b,t,d) with fricatives or affricates (z ,s ,sh ,tch, dge) (eg, "zog" for "dog")		
• Initial consonant deletion (eg, "og" for "dog")		
• Medial consonant deletion (eg, "fe-a" for "feather")		
• Additional consonants (eg, "dunk" for "duck")		
• Replacing nasal sounds (m, n) with a non-nasal sound (eg, "tife" for "knife")		
• Replacing groups of sounds with a favourite sound (all sounds replaced typically by a d, n or h—eg, "hy heddy" for "my teddy")		
Child significantly affected by speech difficulty (eg, becoming withdrawn)		
Parent/caregiver anxiety extremely high over child's speech difficulty		
Referral decision	Yes if ≥2 areas of concern	No if 1 area of concern

*Based on Dodd et al[23] where error patterns occurred in less than 10% of English monolingual children in normative sample or observed in clinical population.