Predictors of Phrase and Fluent Speech in Children With Autism and Severe Language Delay

WHAT'S KNOWN ON THIS SUBJECT: Autism is a disorder that significantly affects language/communication skills, with many children not developing fluent language. The rate of spoken language acquisition after severe language delay and predictors of functional language, beyond comorbid intellectual disability, is less clear.

WHAT THIS STUDY ADDS: This study uses the largest sample to date to examine the relationship between key deficits associated with autism and attainment of phrase and/or fluent speech after a severe language delay, providing information to guide therapeutic targets and developmental expectations.

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abstract

OBJECTIVE: To examine the prevalence and predictors of language attainment in children with autism spectrum disorder (ASD) and severe language delay. We hypothesized greater autism symptomatology and lower intelligence among children who do not attain phrase/fluent speech, with nonverbal intelligence and social engagement emerging as the strongest predictors of outcome.

METHODS: Data used for the current study were from 535 children with ASD who were at least 8 years of age (mean = 11.6 years, SD = 2.73 years) and who did not acquire phrase speech before age 4. Logistic and Cox proportionate hazards regression analyses examined predictors of phrase and fluent speech attainment and age at acquisition, respectively.

RESULTS: A total of 372 children (70%) attained phrase speech and 253 children (47%) attained fluent speech at or after age 4. No demographic or child psychiatric characteristics were associated with phrase speech attainment after age 4, whereas slightly older age and increased internalizing symptoms were associated with fluent speech. In the multivariate analyses, higher nonverbal IQ and less social impairment were both independently associated with the acquisition of phrase and fluent speech, as well as earlier age at acquisition. Stereotyped behavior/repetitive interests and sensory interests were not associated with delayed speech acquisition.

CONCLUSIONS: This study highlights that many severely language-delayed children in the present sample attained phrase or fluent speech at or after age 4 years. These data also implicate the importance of evaluating and considering nonverbal skills, both cognitive and social, when developing interventions and setting goals for language development. *Pediatrics* 2013;131:e1128–e1134

Children with autism spectrum disorders (ASDs) comprise a heterogeneous group, with varying levels of behavior, communication, socialization, and intellectual ability. Abnormalities in communication/language functioning are considered a defining feature of ASD, as evidenced by inclusion in the fourth edition of the *Diagnostic and Statistical Manual of Mental Disorders*.[1] More importantly, acquisition of functional language by school age is associated with a more favorable prognosis for milder symptoms and better adaptive functioning in adulthood,[2–4] although a large proportion of children with ASD do not develop useful language during this period. For example, a recent longitudinal study[5] monitored speech development among 130 children with ASD from age 2 to 9 years who were not exclusively language delayed. They found that 24% of participants with autism and 59% with pervasive developmental disorder not otherwise specified obtained fluent speech (ie, ability to use complex utterances to talk about topics outside of the immediate physical context) by age 9 years. In contrast, 30% of those with autism and 4% with pervasive developmental disorder not otherwise specified were termed nonverbal (ie, using no or few consistent words) by age 9. With regard to predictors of language acquisition, early nonverbal intelligence and joint attention emerged as being the most salient, although additional behaviors associated with ASD were not examined (eg, repetitive behaviors, psychiatric comorbidities).

Recently, Pickett et al[6] conducted a comprehensive review of the literature to examine the age of speech onset and subject characteristics of severely language-delayed ("nonverbal") children with ASD in which only 167 individuals fitting these criteria were identified. The majority of children identified began speaking between age 5 and 7 years, and most gained only the ability to produce single words; however, ~30% achieved phrase speech. Characteristics of individuals who began to speak after age 5 years included an IQ of ≥50 and participation in (behavioral) intervention. Again, other behaviors associated with ASD were not examined.

Although recent research suggests that factors such as nonverbal intelligence and social responsiveness (ie, joint attention) may be important predictors of language outcomes, the complex question remains why a large number of children with ASD do not develop meaningful language during their preschool years. For instance, whereas repetitive behaviors have been mostly overlooked with regard to their association with language development, there is some evidence for an inverse relationship with nonverbal cognition and language development,[7,8] particularly in very young children.[9] How these and other ASD-related factors (eg, sensory behaviors, comorbid psychopathology) interact, particularly in children who exhibit significantly delayed language development, is unclear.

The purpose of the current study was to examine the rate of language acquisition in a large, well-characterized sample of children with ASD with a history of severe language delay (defined as children who are not putting words together into meaningful phrases by age 4 years, which could include children who are nonverbal as well as those using single words or occasional basic phrases without a verb). In addition, cognitive and behavioral traits related to the development of functional verbal communication among these children were evaluated. The second aim examined factors that predict the age at which children with severe language delays develop phrase speech. Specifically, we hypothesized greater impairments across autism characteristics and nonverbal cognitive development in children who remain nonverbal as compared with those who gain verbal communication. Second, given the known relationship between socialization and communication as well as cognition and language development, we predicted a stronger association between social and cognitive factors and language outcome than between repetitive and sensory behaviors and language outcome.

METHODS

Sample

Data used for the current study were gathered from the Simon Simplex Collection (SSC). Data collection and analysis were approved by the appropriate institutional review boards. The SSC is a multisite database project that gathers biological and phenotypic data of children with ASD (ages 4–18 years) whose families have (1) 1 child with ASD and (2) no first- through third-degree relatives diagnosed with or suspected of having an ASD. In addition, children were excluded from the SSC if there was history of (1) gestational age <36 weeks or birth weight <2000 g, (2) extensive pregnancy or birth complications, (3) confirmed fragile X or Down syndrome, (4) sensory or motor impairment precluding participation in standardized testing, or (5) severe nutritional or psychological deprivation. All probands met clinical cutoffs for ASD according to the Autism Diagnostic Interview–Revised (ADI-R)[10]; and the Autism Diagnostic Observation Schedule–Generic (ADOS).[11] For further details of the SSC collection, see www.sfari.org.

Data used for the present analysis were extracted on February 1, 2012. Although a similar, smaller subset of this SSC sample was examined by Mazurek et al,[12] their objectives, variables, and analyses were significantly different from the methodology used in the current study.

At the time of data analysis, the SSC data release (version 13) included a total of 2648 families. The initial sample derivation included children aged ≥8 years (n = 1456). The second derivation required the child to either have no phrase speech when assessed for enrollment in the SSC or an age of phrase speech onset at ≥4 years reported via the ADI-R item 10 (n = 588). Next, validation of speech presentation at the time of testing was determined by clinician observation via the ADOS module administered. There were some instances of discordance between parent report (ADI-R) and clinician observation (ADOS) of the current level of speech production. Given that retrospective parent report of developmental milestones is prone to errors or distortions of recall,[13] the final determination of group for discordant children was based on ADOS administration/clinician observation (ie, children who received an ADOS module 1 were considered as not having attained phrase speech [n = 84]; children who received an ADOS module 2 were dropped from analyses [n = 53], because it could not be determined when they achieved phrase speech).

This resulted in a total sample of 535 children. Of these 535 children, 163 were given an ADOS module 1 and considered to be severely language delayed or to have single words only/no phrase speech (NPS group). Children in the phrase speech group (PS group) received an ADOS module 2 or higher at time of testing (n = 372; 70%), whereas a subsample of this group (n = 253) were considered as the fluent speech group (FS group) because they received an ADOS module 3 or 4. Demographic characteristics are presented in Table 1.

Measures

ADI-R

The ADI-R is a standardized parent interview developed to distinguish children with ASD from non-ASD populations. The

TABLE 1 Demographic, Psychiatric, Cognitive, and Autism Symptom Severity Differences Between Those With and Without Phrase Speech

	No Phrase Speech	Phrase Speech
n	163	372
Mean age at enrollment, y	11.5	11.7
Age 8–12 y, %	63	61
Age 13–17 y, %	37	39
Gender (male), %	83	86
Income, %		
≤$50 000	16	17
$51 000–$100 000	39	41
≥$101 000	45	42
Maternal education, %		
No college	14	13
College	67	65
Graduate	19	22
Race (white versus nonwhite), %	63	69
Child Behavior Checklist T score		
Externalizing	55.5	54.2
Internalizing	57.1	58.5
IQ		
Verbal	23.1**	68.3**
Nonverbal	40.4**	80.6**
ADI: autism symptoms		
Social impairment	26.4**	23.3**
Repetitive interests	6.5*	7.1*
Sensory interests	4.1	4.3

* $P < .05$, ** $P < .001$.

ADI-R item 10 was used to identify the age at which the child attained phrase speech. This item distinguishes between those using meaningful 2- to 3-word phrases containing a verb from those who were not communicating using phrases. In addition, the Social Development and Play Subscale; the Total Restrictive, Repetitive, and Stereotyped Patterns of Behavior Subscale; and the unusual sensory interest item (no 71, current) were used as independent variables.

ADOS

The ADOS is a clinician-administered observational assessment that directly assesses social, communicative, and stereotyped behaviors diagnostic of autism. Children in this study were administered 1 of 4 modules on the basis of their language presentation: no words or single words (module 1), phrase speech (module 2), and fluent speech (module 3 or 4). The ADOS was used to classify current level of language attainment (ie, no phrase speech, phrase speech, fluent speech).

Intellectual Functioning

IQ was assessed with various standardized measures, including the Differential Abilities Scale–Second Edition,[14] Mullen Scales of Early Learning,[15] Wechsler Intelligence Scale for Children–Fourth Edition,[16] and the Wechsler Abbreviated Intelligence Scale.[17] Deviation verbal and nonverbal IQ scores were calculated when possible; otherwise, ratio IQ scores were computed. Nonverbal IQ was used as an independent variable.

Child Behavior Checklist

The Child Behavior Checklist[18] is a standardized parent-report questionnaire assessing emotional and behavioral functioning. Internalizing and externalizing subscales were used as independent variables.

Data Analysis

Independent t- and χ^2 tests were used to examine differences in demographic characteristics, autism symptomatology (ADI social, sensory, and repetitive behaviors), and IQ between those with

and without phrase speech (NPS versus PS groups) and between the subset of children within the PS group who attained fluent speech (n = 253) versus those remaining children in the PS group who did not attain fluent speech (n = 119). Significant variables from those bivariate analyses were examined in multivariate regression models to better understand the independent and interaction effects between the predictor or predictors and outcome. The first series of analyses separately examined the likelihood of phrase speech (compared with no phrase speech) and fluent speech (compared with those with phrase speech but without fluent speech) by using logistic regression models. The next set of analyses examined the association between predictors and time at which phrase speech was attained by using Cox proportionate hazards models.[19] Model building procedures involved including a single variable at a time, beginning with IQ, followed by autism symptomatology and the remaining significant variables in the bivariate analyses. To better understand IQ and autism symptoms as categorical variables, a Kaplan-Meier procedure was used to estimate the proportion of children who achieved phrase speech across those children with intellectual disability (IQ <70), borderline to low-average intellectual functioning (IQ of 70–85), and at least typical intelligence (IQ ≥86). An identical procedure was used for those with mild (<22), moderate (22–25), and severe (>25) social deficits as determined by tertiles via the ADI-R.

RESULTS

Demographic Differences

Descriptive demographic, psychiatric, and developmental statistics are presented in Tables 1 and 2. No differences were found between the PS and NPS groups. Comparisons between the PS-only and FS groups indicated older age, male gender, and slightly higher

internalizing symptoms for those in the FS versus only-PS groups.

Autism Symptomatology and IQ

Tables 1 and 2 also show differences between groups with regard to autism symptoms (ADI-R social development, repetitive interests, and sensory behaviors) as well as IQ. Significant differences were found across IQ and autism symptoms, with the PS group showing higher (better) mean verbal and nonverbal IQ scores, less social impairment, and surprisingly more (worse) stereotyped behaviors compared with those with in the NPS group. Children in the FS group had higher mean verbal and nonverbal IQ scores as well as lower social impairment and stereotyped behaviors compared with children in the with PS-only group; no differences were found for unusual sensory interests across groups.

Predictors of Phrase Speech

Results from the multivariate logistic regression analyses revealed that

higher nonverbal IQ (odds ratio [OR]: 1.08; $z = 11.21$; 95% confidence interval [CI]: 1.08, 1.10) and lower social impairment (OR: 0.86; $z = -4.07$; 95% CI: 0.79, 0.92) were associated with an increased likelihood of phrase speech attainment, with IQ being a stronger predictor (both $P < .001$). When nonverbal IQ was entered as a covariate, repetitive interests/stereotyped behavior was not associated with phrase speech attainment.

Predictors of Fluent Speech

Results from the multivariate logistic regression analyses revealed that higher nonverbal IQ (OR: 1.06; $z = 7.51$; 95% CI: 1.05, 1.08), lower social impairment (OR: 0.80; $z = -5.87$; 95% CI: 0.74, 0.86), increased age (OR: 1.34; $z = 5.08$; 95% CI: 1.20, 1.51), and increased internalizing symptoms (OR: 1.05; $z = 3.2$; 95% CI: 1.02, 1.08) were associated with an increased likelihood of fluent speech attainment (all $P < .01$). Notably, repetitive interests/stereotyped behaviors was not significant in the

TABLE 2 Demographic, Psychiatric, Cognitive, and Autism Symptom Severity Differences Between Those With and Without Fluent Speech

	Only Phrase Speech	Fluent Speech
n	119	253
Mean age at enrollment, y	11.0**	12.0**
Age 8–12 y, %	71**	56**
Age 13–17 y, %	29**	44**
Gender (male), %	80*	89*
Income, %		
≤ $50 000	15	17
$51 000–$100 000	38	42
≥$101 000	47	40
Maternal education, %		
No college	13	13
College	68	64
Graduate	19	23
Race (white versus nonwhite), %	64	71
Child Behavior Checklist T score		
Externalizing	55.1	53.8
Internalizing	56.6*	59.4*
IQ		
Verbal	46.1**	78.7**
Nonverbal	65.5**	87.7**
ADI: autism symptoms		
Social impairment	25.5**	22.2**
Repetitive interests	7.5*	6.9*
Sensory interests	4.4	4.2

* $P < .05$, ** $P < .001$.

full model, and although there was a statistically significant difference between groups in internalizing behavior symptoms, neither group reached clinical significance.

Predictors of Age at Speech Attainment

Results from the multivariate Cox proportionate hazards regression analyses revealed that higher nonverbal IQ (hazard ratio: 1.01; $z = 3.38$; 95% CI: 1.01, 1.02) as well as lower social impairment (hazard ratio: 0.94; $z = -5.14$; 95% CI: 0.92, 0.96) scores were independently associated with earlier phrase speech attainment. Notably, the effect of social impairment was slightly stronger than IQ. Again, repetitive interests/stereotypical behavior was not associated with age at speech attainment after adjustment for nonverbal IQ.

Figure 1 shows a Kaplan-Meier procedure that allows for a graphical examination of the effect of nonverbal IQ, as a categorical variable, on time to language attainment. The figure shows that children with "typical" intelligence (nonverbal IQ >85) attained language at the quickest rate; no differences were found between those with borderline to low-average nonverbal IQ (70–85) and children falling in the range of intellectual disability (IQ <70). Figure 2 also shows a Kaplan-Meier graph, except this figure shows the effect of social impairment on age at language attainment. Here, the effect of ASD social impairment on rate of language acquisition appears to be uniform across severity, with those who have less social impairment gaining speech at an earlier age.

DISCUSSION

In our large sample of children with ASD and a history of severe language delays, attainment of phrase and/or fluent speech was achieved by the majority
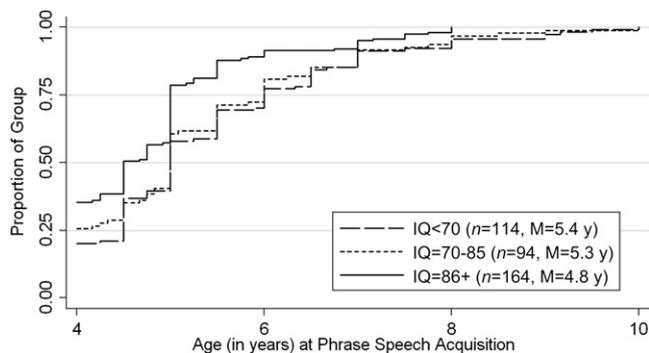
(70%) of participants by age 8 years, with almost half of the sample achieving fluent speech. As such, many children with ASD who present with severe language delay (ie, having only few single words at most) at age 4 years can be expected to make notable gains in the development of language. This proportion of language attainment is comparable to the longitudinal findings reported by Anderson et al[5]; however, given the differences in sample characteristics (ie, present sample included only children with severe language delays), our findings suggest that a greater proportion of children in the general ASD population may be capable of attaining phrase

speech than previously reported. This finding is particularly salient given the strengths of the present sample characteristics (drawn from a large, multisite collaboration). With that stated, because our sample did not consist of exclusively nonverbal children, the rate of speech attainment may be lower if the sample was divided further by level of language to create a more homogenous group.

Results from the current study continue to support the importance of nonverbal intelligence and social engagement as primary predictors of language development for children with ASD,[5,6] even on simultaneous consideration of other defining and interfering

FIGURE 1

Effect of IQ on age at phrase speech. A Kaplan-Meier procedure shows that children with intelligence in the average range (IQ >85) attained language at the quickest rate, although no differences were found between those with borderline to low-average intelligence (IQ of 70–85) and those with intellectual disability (IQ <70) with regard to age at phrase speech attainment. Kaplan-Meier statistics were used because of natural censoring. M = Mean.
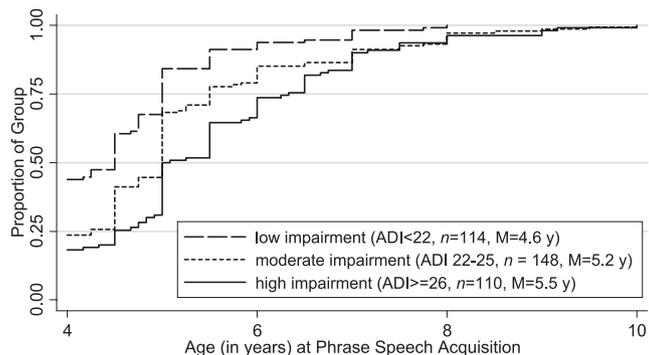

FIGURE 2

Effect of social deficits on age at phrase speech. A Kaplan-Meier graph shows the effect of social impairment (low, moderate, and high) on age at language attainment. Here, the effect of ASD social impairment on the rate of speech acquisition appears to be uniform across severity. Kaplan-Meier statistics were used because of natural censoring. M = Mean.

behaviors (ie, repetitive and abnormal sensory behaviors). Our findings suggest that these core abilities have greater influence on the development of communication for children with ASD than do other associated behaviors (eg, repetitive/stereotyped, sensory) that have been previously associated with early language development in ASD.[7–9] This discrepancy may be partially explained by our focus on older, school-aged children (versus examining young children, <3 years old, who may not necessarily go on to experience a severe language delay).

With regard to treatment planning, findings further substantiate the importance of considering both nonverbal intelligence and social communication, potentially supporting use of intervention strategies for these children that focus on the development of social cognition strategies (eg, perspective taking/theory of mind). Further research into the impact of these interventions on the development of spoken language is warranted. In addition, results uniquely suggest that the level of intellectual ability is a critical consideration. Specifically, concerning age at phrase speech acquisition, the most meaningful distinction was between children with normal (ie, ≥1 SD of the mean) nonverbal intelligence from those who fall below 1 SD of the mean. Whereas children with both intellectual disability and normal nonverbal intelligence gained phrase speech, those falling below the 1-SD mark were delayed in their acquisition of phrase speech by ∼6 months. Although there was also an independent effect of social impairment on age at phrase speech acquisition, the effect was more uniform across symptom severity. As such, treatment expectations may be adjusted for lower functioning children (ie, children with nonverbal IQ falling in the low-average to borderline-impaired range and with notable social

impairment), such that treatment gains can be expected but at a slower pace than in those with more typical nonverbal intelligence or less social impairment. In addition, given that the relative importance of nonverbal IQ and social impairments depends on outcome (ie, nonverbal IQ was the strongest predictor of phrase speech and social impairment was similar in strength when predicting fluent speech), the focus of treatments may also be adjusted dependent on developmental language progress. Further research examining the relationship among specific social deficits and fluent speech development is clearly warranted and may hold important implications to the design of intervention.

Although attempts were made to confirm current language level (ie, by ADOS module administration), the most significant limitation of the current study was the reliance on retrospective parent report to classify children as language delayed. Research on parent-reported language achievement has shown that parents of older children report later age at language acquisition than do parents of younger children (referred to as "forward telescoping[20]") Therefore, it is possible that there is some bias in our sample, possibly toward overestimating the severity of the language delay of included subjects. Nevertheless, we have assumed that these biases are constant through the sample. In addition, although the current study used a large sample, recruited from multiple sites across the United States (and Canada), the generalizability of findings is limited by inclusion of only simplex families (ie, families with only 1 child with autism). Because autism is highly heterogeneous and heritable, children from simplex families may reflect a subset not reflective of the autism population as a whole. Future longitudinal studies including both simplex and multiplex

families are required to fully capture the prevalence and predictors of language development in severely delayed children with ASD.

CONCLUSIONS

Consistent with previous reports, many severely language-delayed children with ASD attain phrase/fluent speech. Nonverbal cognition was the strongest predictor of phrase speech, whereas social interest and engagement were as robust, if not greater, when predicting age at phrase speech and fluent speech; other ASD-related features such as repetitive and sensory behaviors did not predict language outcome, further emphasizing the overlap between socialization and communication for this population. Upon closer examination, children with nonverbal intelligence within 1 SD of the mean (ie, IQ ≥85) attained language almost 6 months ahead of those with nonverbal intelligence below the 1-SD mark, suggesting that children whose nonverbal intelligence is in the borderline to low-average range are at similar risk to those whose levels are consistent with intellectual disability. Taken together, for children without phrase speech before age 4 years, average nonverbal intelligence and evidence of social interest and engagement reflect more a positive prognosis, with the prognosis of combinatorial language attainment (ie, at least phrase speech) after age 10 years for children with intellectual disability being quite limited.

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Pediatrics 2013;131;e1128

DOI: 10.1542/peds.2012-2221 originally published online March 4, 2013;

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DOI: 10.1542/peds.2012-2221 originally published online March 4, 2013;

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