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Speech-Sound Disorders and Attention-Deficit/Hyperactivity Disorder Symptoms

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Abstract

Purpose—The purpose of this study was to examine the association of speech-sound disorders (SSD) with symptoms of attention-deficit/hyperactivity disorder (ADHD) by the severity of the SSD and the mode of transmission of SSD within the pedigrees of children with SSD.

Participants and Methods—The participants were 412 children who were enrolled in a longitudinal family study of SSD. Children were grouped on the basis of the severity of their SSD as determined by their scores on the Goldman–Fristoe Test of Articulation and history of an SSD. Five severity groups were compared: no SSD, resolved SSD, mild SSD, mild–moderate SSD, and moderate–severe SSD. Participants were also coded for comorbid language impairment (LI), based on scores on a standardized language test. Pedigrees of children were considered to represent bilineal inheritance of disorders if there was a history for SSD on both the maternal and paternal sides of the family. Parents completed the ADHD rating scale and a developmental questionnaire for each of their children.

Results and Conclusions—Children with moderate–severe SSD had higher ratings on the inattention and hyperactive/impulsivity scales than children with no SSD. Children whose family pedigrees demonstrated bilineal inheritance had higher ratings of inattention than children without bilineal inheritance. To determine the best predictors of ADHD ratings, multiple linear regression analyses were conducted. LI was more predictive of ADHD symptoms than SSD severity, bilineal inheritance of SSD, age, or gender. Findings support that LI rather than SSD is associated with ADHD.

Keywords

ADHD; attention; genetics; language impairment; reading disorder; speech sound disorder

Speech sound disorders(SSD), language impairment (LI), and attention-deficit/hyperactivity disorder (ADHD) are common developmental disorders that can significantly impact academic success. The comorbidity of these disorders may substantially add to the difficulties experienced by the child and require a combined treatment approach. SSD and LI are often comorbid, with approximately 40%–60% of children with SSD at preschool (Shriberg & Kwiatkowski, 1994) and 11%–15% of 6-year-old children with SSD presenting with LI (Shriberg, Tomblin, & McSweeney, 1999). LI and ADHD are often comorbid as

well, with rates of comorbidity reported to be 30%–50% in children seen at ADHD clinics (Tannock & Schachar, 1996). Although there are only a few reports on the comorbidity of SSD and ADHD, these studies have suggested that LI, rather than SSD, is associated with ADHD (McGrath, Hutaft-Lee, Scott, Boada, Shriberg, & Pennington, 2007; Short, Freebairn, Hansen, & Lewis, 2008).

Children with SSD have varied academic outcomes depending on the constellation of deficits that they exhibit. In our previous studies, we found that children with SSD in isolation had better school-age outcomes on literacy measures than children with comorbid LI (Lewis, Freebairn, & Taylor, 2002). We also demonstrated that children with SSD and comorbid conditions of LI or reading disorder (RD) or both perform more poorly than children with isolated SSD on measures of endophenotypes, including phonological memory, phonological awareness, and speeded naming (Lewis, Avrich, Freebairn, Taylor, Iyengar, & Stein, 2011). Furthermore, we have reported that children with SSD and comorbid LI are rated more poorly by their parents on an ADHD rating scale (ARS) than children with isolated SSD (Short et al., 2008). In that study, we examined three groups of participants: children with SSD alone ($n = 71$), children with SSD and comorbid LI ($n = 77$), and typical children ($n = 72$). Children with SSD + LI were more likely to have been diagnosed with ADHD by a health care provider, and to have reading problems and spelling problems, than children with SSD alone or typical children. Although the ARS was the best predictor of ADHD, the combination of the ADHD rating score and a score on the Clinical Evaluation of Language Fundamentals 3 (CELF-3) improved the prediction of ADHD. This suggests that language abilities are predictive of ADHD over and above the ARS. However, in these studies the severity of SSD and the family history for disorders were not considered. This article extends these findings by examining differences in ADHD ratings in children with resolved, mild, mild-moderate, or moderate-severe SSD with and without comorbid LI. Furthermore, we consider the impact that family history for SSD reported for both parents (i.e., *bilineal* transmission of SSD) has on the presence of comorbid ADHD.

THE RELATIONSHIP OF ADHD TO SSD AND LI

The onset and prevalence of developmental disorders vary widely. SSD is highly prevalent in young children, with rates as high as 15.6% of 3-year-old children (Campbell et al., 2003) declining to 3.8% of 6-year-old children (Shriberg et al., 1999). LI has reported prevalence estimates of 7.4% in kindergarten (males = 8%, females = 6%; Tomblin et al., 1997). Prevalence estimates of ADHD vary from 1.9% to 17.8% of school-age children, with estimates differing depending on the nature of the sample recruited (Greenhill, Benton, & Tirmizi, 2003).

Children with combined SSD and LI comprise approximately 7% of the general population (Johnson et al., 1999) and appear to be at increased risk for ADHD. Reilly, Cunningham, Richards, Elbard, and Mahoney (1999) reported that more than 30% of children diagnosed with SSD + LI met criteria for ADHD. Estimates of comorbid ADHD may vary depending on whether the sample is clinically ascertained or population based. Rates of ADHD were 5 times higher in the language-disabled population obtained from a speech and language clinic than in a sample of children from the general population without a diagnosis of LI. Similarly, rates were quite high in a prospective sample of Canadian kindergarten children (Beitchman, Nair, Clegg, Ferguson, & Patel, 1986), with 16% of children diagnosed with both SSD and LI simultaneously diagnosed with ADHD, as compared with only 6% of typical kindergarten children diagnosed with ADHD. In a later study, Beitchman et al. (1989) reported that 59% of children with both SSD and LI had comorbid attention deficits, whereas only 5% of children with SSD alone demonstrated these attentional problems. Many children diagnosed with ADHD have both articulation and language problems (Baker

& Cantwell, 1992), with expressive language difficulties noted more frequently in children with ADHD than receptive language difficulties (Barkeley, DuPaul, & McMurray, 1990).

In addition, a study by McGrath et al. (2007) examined ADHD symptoms as rated by parents and teachers in 108 children aged 4–7 years with SSD who were also diagnosed with or without LI. Children with combined SSD and LI had higher rates of inattention than children with SSD alone. It should be noted, however, that no differences were found in the children with and without LI on the hyperactivity/impulsivity ratings. An unexpected finding was that children with SSD and LI whose speech disorder had normalized tended to have higher rates of inattention than children with persistent SSD. This suggests heterogeneity within the SSD population. One hypothesis proposed by McGrath et al. to account for their findings is that children with comorbid SSD and LI whose SSD resolves may exhibit more general language learning disabilities that are accompanied by inattention disorders, whereas children with persistent SSD may demonstrate a more circumscribed phonological deficit.

Comorbidity rates of SSD, LI, and ADHD are also high when the sample is ascertained primarily by an ADHD diagnosis. In ADHD clinics, rates of language problems range from 30% to 50% (Tannock & Schachar, 1996). Levy, Hay, McLaughlin, Wood, and Waldman (1996) surveyed a nonselected sample of 1,938 families for both ADHD and speech and language disorders, and found a strong association of ADHD with speech and reading problems.

SUBGROUPING CHILDREN WITH SSD BY SEVERITY

Despite the high prevalence rates of ADHD and LI, few researchers have considered SSD separately from LI, and they have not considered subtypes within the SSD population. One method of subgrouping children with SSD has been to group children on the basis of the severity of SSD (Lewis et al., 2011). Traditionally, clinical severity ratings of SSD can be identified as mild, moderate, or severe depending upon the speech–language pathologist’s (SLP’s) judgment of the child’s intelligibility of conversational speech or the number of speech sound errors that the child exhibits on a standardized articulation test. Another measure of severity, which is more objective, is the Percentage of Consonants Correct (PCC) developed by Shriberg and Kwiatkowski (1982). The PCC includes the number of consonants produced correctly in a continuous speech sample divided by the total number of consonants in the sample. Severity of SSD is classified as mild (85%–100% correct), mild–moderate (65%–85% correct), moderate–severe (50%–65% correct), or severe (<50% correct). Despite the widespread usage of severity ratings to make treatment decisions, however, clinicians do not always agree in their perceptual ratings of a child’s speech (Flipsen, Hammer, & Yost, 2005).

An issue that is not yet resolved is whether children with more severe forms of SSD are qualitatively different than children with milder forms of SSD, and as such, present with different etiologies, speech sound characteristics, and developmental trajectories. Most clinicians and researchers assume that children with severe language disorders are more biologically compromised than those with milder disorders. Geneticists have long used a variety of severity measures in their selection of families and populations (e.g., recurrent clustering in families, earlier age at onset, and the number of manifest conditions) hoping that severity might be a useful tool for isolating genes specifically tied to the condition or disease. As severity is currently defined, children with the most severe forms of the disorder require the most extensive treatments and present with lifelong problems that eventually manifest as lost productivity and poorer perceived quality of life (Ruben, 2000).

GENETIC LOAD AND COMORBIDITY OF SSD AND ADHD

A substantial number of researchers have argued that susceptibility to SSD is genetic (Hurst, Baraitser, Auger, Graham, & Norell, 1990; Spitz, Tallal, Flax, & Benasich, 1997; Whitehurst et al., 1991). Family studies have demonstrated familial aggregation for SSD, with 20%–40% of first-degree relatives affected (Felsenfeld, McGue, & Broen, 1995; Lewis, Cox, & Byard, 1993; Lewis, Ekelman, & Aram, 1989; Tallal, Ross, & Curtiss, 1989). Segregation analyses also have confirmed familial aggregation of SSD and have supported both a major gene and polygenic modes of transmission of the disorder (Lewis et al., 1993). Twin studies (Bishop, North, & Donlan, 1995) also have supported a genetic basis for SSD. Bishop and Haiyou-Thomas (2008) found that LI was more heritable when comorbid with SSD than when occurring in isolation. Rates of heritability for SSD have been estimated as high as .97 whereas the heritability of LI has been estimated to range from 0.38 to 0.76, depending on the severity of the disorder (Viding et al., 2004). Several studies have estimated the heritability of component skills of SSD (Bishop, Adams, & Norbury, 2006). Reported heritabilities for these skills are as follows: 0.37 for articulation, 0.37 for oral motor skills, 0.43 for language, 0.60 for phonological awareness, 0.52 for semantics, 0.30 for syntax, 0.63 for phonological memory, and 0.64 for rapid naming. A study by Bishop et al., (2006) reported the heritability of phonological memory, based on nonword repetition, to be 0.61, a sentence imitation task to be 0.36, and a syntax task to be 0.82.

Only a few molecular genetic studies of SSD have been conducted. Two genes specific to speech and voice disorders (i.e., *FOXP2* and *TOSPEAK*) have been isolated, whereas other genes appear to primarily be associated with RD and LI (i.e., *KIAA0319*, *DCDC2*, *CNTNAP2*, *DYX1C1*, and *ROBO1*).

Discovery of the *FOXP2* gene resulted from studies of a large three-generation family with an SSD, designated as the KE family, whose members demonstrated orofacial apraxia and associated speech–language disorders. The *FOXP2* gene is located on chromosome 7. Individuals who carried the mutant *FOXP2* allele evidenced a variety of deficits, including SSD, as well as impairments in IQ, LI, and RD (Lai, Fisher, Hurst, Vargha-Khadem, & Monaco, 2001). Findings from neuroimaging studies of the KE family suggested that the *FOXP2* gene has pleiotropic (i.e., multiple) effects on many aspects of brain development, accounting for the co-occurrence of SSD and LI (Liégeois, Morgan, Connelly, & Vargha-Khadem, 2011). Although the *FOXP2* mutation has been found in only a few families and does not account for the majority of SSD, these findings confirm the existence of genes that influence SSD (Shriberg et al., 2006). This gene remains of interest because mouse pups with a knockout (i.e., deactivation) for *FOXP2* showed disruptions in ultrasonic vocalization that interfered with the communication between pup and mother (Shu et al., 2005). Thus, it is feasible that unidentified variants in this molecule cause some forms of SSD.

The genetics of ADHD are also complex (Pauls, 2005; Poelmans, Pauls, Buitelaar, & Franke, 2011). Genetic studies of ADHD including family, adoption, and twin studies (Faraone & Doyle, 2000; Smalley, 1997; Tannock, 1998) and molecular genetic studies (Fisher & DeFries, 2002) have suggested a genetic component to ADHD, with estimates of heritability exceeding 0.70. A recent review of five genome-wide association studies of ADHD revealed 85 candidate genes, with 45 of these influencing a neurodevelopmental network (Poelmans et al., 2011). Candidate genes for ADHD influence processes such as neuronal migration, cell adhesion, and cell division (Banaschewski, Becker, Scherag, Franke, & Coghill, 2010). Endophenotypes examined in genetic studies of ADHD include those relating to executive function, motor deficits, reaction time, speed of processing, set shifting, and cognitive impulsiveness (Kebir & Joober, 2011; Rommelse et al., 2009).

Given the comorbidity of disorders that often occur in SSD, LI, and ADHD, researchers have been assuming common underlying genetic bases for these disorders. Pennington and Bishop (2009) have contended that the comorbidity observed in these diagnostic groups may result from shared deficits in endophenotypes or cognitive skills, including memory, processing speed, or attention (McGrath et al., 2007; Pennington & Bishop, 2009; Peterson, Pennington, Shriberg, & Boada, 2009). Quite likely, these endophenotypes may be influenced by genes that have broad effects on neural development and processing (Lewis et al., 2011). For example, Castellanos and Tannock (2002) have suggested that endophenotypes, such as a delay in temporal processing or working memory deficits, appear to underlie ADHD, with the same endophenotypes contributing to SSD and LI. A child with ADHD may not attend to features of speech and language necessary for mastery. Similarly, the multiple deficit model suggests that shared deficits in processing speed may account for the comorbidities of these disorders (Pennington, 2006).

Not surprisingly, genetic overlap between these disorders has been observed. Genetic linkage studies have demonstrated that SSD and ADHD share a common locus on chromosome 6p22 (Smith, Pennington, Boada, & Shriberg, 2005; Wilcutt et al., 2002). However, studies of children with SLI have not found linkage to this locus (Bartlett et al., 2002; The SLI Consortium, 2002, 2004). Although SSD, LI, RD, and ADHD often may be comorbid, there is evidence of both shared and independent genetic effects for these disorders that is consistent with the pleiotropic effects of some genes, that is, with one gene influencing multiple traits or phenotypes (Gayan et al., 2005; Hart et al., 2010).

To date, only a few studies have specifically examined the comorbidity of SSD and ADHD. Although these studies have considered comorbid LI, they have not examined the severity of SSD, nor have they considered family history for SSD. The purpose of this study was to examine rates of comorbid ADHD in a clinical sample of children diagnosed with early SSD with and without comorbid LI. Children with SSD are a heterogeneous group, and factors such as severity of impairment and family history for SSD may influence the developmental trajectory of the disorder. In addition, high genetic load, which is defined as bilineal inheritance in families that report affected individuals for SSD in both maternal and paternal pedigrees, may be associated with higher rates of comorbidities. This study addressed two research questions:

1. Is there an association between severity of early SSD (none, resolved, mild, mild–moderate or moderate–severe) and later ADHD symptoms; and, if so, which variables (i.e., severity of SSD, comorbid LI, gender, or age) best predict ADHD ratings?
2. Is there an association between early SSD and later ADHD symptoms if the family demonstrates bilineal inheritance of SSD; and, if so, which variables (i.e., bilineal inheritance of SSD, comorbid LI, gender, or age) best predict ADHD ratings?

METHOD

Participants

All participants were part of an ongoing family study of speech and language disorders (Lewis et al., 2002; Lewis, Freebairn, & Taylor, 2000a, 2000b). Families were recruited from the clinical caseloads of SLPs working in the greater Cleveland area. All children in each family were tested regardless of whether they were enrolled in speech–language therapy. Proband children with SSD ($n = 163$) met the following criteria at the time of their initial assessment: (a) scored below the 10th percentile on the Goldman–Fristoe Test of Articulation (GFTA)–Sounds in Words Subtest (Goldman & Fristoe, 1986); (b) exhibited at least three phonological process error types on the Khan–Lewis Phonological Analysis

(Khan & Lewis, 1986); (c) passed a pure tone hearing screening bilaterally suggesting normal hearing and had less than six episodes of otitis media prior to the age of 3 years; (d) exhibited a normal oral peripheral speech mechanism as determined by a licensed SLP; (e) showed no signs of other neurological or developmental delays per parent report; (f) demonstrated normal cognitive skills on the performance subscale of the Wechsler Preschool and Primary Scale of Intelligence Revised (WPPSI-R) (performance IQ [PIQ] > 80; Wechsler, 1989) or Wechsler Intelligence Scale for Children-3rd Edition (WISC-III) (PIQ > 80; Wechsler, 1991). Siblings of the proband children ($n = 248$) also were enrolled in the study. Siblings were classified as having SSD if they met the aforementioned criteria or if they had a history of SSD that required speech–language therapy. Siblings who had never had SSD served as the typical control group (no SSD group).

The severity of SSD in the participants was defined by scores, based on the GFTA (Goldman & Fristoe, 1986). A mean GFTA percentile score of 75 ($SD = 31$) was obtained for children who had never received speech–language therapy and had not reported a history of speech or language disorders. On the basis of this mean score, five groups were created: moderate–severe SSD ($< -2 SD$ from the mean; $n = 109$), mild–moderate SSD ($-2 SD$ but $< -1 SD$, $n = 119$), mild SSD ($-1 SD$ but below the mean; $n = 54$), resolved SSD (at or above the mean but a reported history of therapy for SSD; $n = 39$), and no history of SSD (at or above the mean with no history of SSD; $n = 91$).

Children were further diagnosed as SSD with comorbid LI, if they earned a scaled score of 8 or less on two or more subtests of CELF-3 (Semel, Wiig, & Secord, 1995) or the Test of Language Development–Primary 3rd Edition (TOLD-P:3; Newcomer & Hammill, 1997) prior to their initiation of therapy. This definition was based on the criteria for LI of Tomblin et al. (1997), which defined LI as a scaled score of less than 8 on two or more subtests of the Test of Language Development–Primary 2nd Edition. The normal speech and language group ($n = 91$) did not meet criteria for either SSD or LI.

Affection status of parents and extended family members was determined by a semistructured interview, described in the following text. Family members were considered affected if the parents reported that they had a history of SSD. Families were coded as demonstrating bilineal inheritance if both the paternal and maternal family pedigree had at least one member other than the proband and his or her siblings with an SSD.

Measures and procedures

Speech–sound development—The GFTA was administered to determine the presence of speech–sound errors. It uses 35 picture plates and 44 target words to elicit consonants in the initial, medial, and final position of words, as well as 12 consonant clusters (blends) in the initial position. Responses were audio taped, phonetically transcribed, and plotted on the consonant error matrix. Age-adjusted percentile scores were employed in the severity classification.

Language—The Clinical Evaluation of Language Fundamentals–Preschool (CELF-P; Wiig, Secord, & Semel, 1992), CELF-3, or TOLD-P:3 was administered to assess expressive and receptive language skills. Age-adjusted standard scores were employed to classify children with LI.

Performance IQ—Participants were required to demonstrate normal intelligence as defined by a prorated PIQ of at least 80 on the WPPSI-R or WISC-III.

Semistructured interview—All comorbidity data were obtained by parent report in a semistructured interview. Parents were asked to indicate whether their child was ever diagnosed with the following disorders: learning disabilities, LI, ADHD, or RD. Family pedigree data were collected for both the paternal and maternal families through the Family History Interview (Lewis & Freebairn, 1993), a pedigree was drawn, and families were coded as demonstrating bilineal inheritance if both the maternal and paternal sides of the family reported affected members for SSD.

ADHD rating scale—In this study, comorbid ADHD was documented either by parent report of a previous diagnosis of ADHD by a health care provider or by ratings within the clinical range on the ADHD Rating Scale (ARS) (*Diagnostic and Statistical Manual of Mental Disorders* [Third Edition Revised] and *Diagnostic and Statistical Manual of Mental Disorders* [Fourth Edition] versions; DuPaul, 1991; DuPaul et al., 1996). Symptoms of ADHD (but not necessarily a clinical diagnosis of ADHD) were documented through the ARS. The ARS assesses the commonly noted symptoms of ADHD, including hyperactivity, impulsivity, and inattention. The home report version of the ARS was employed in the present investigation. The parent is asked to evaluate the child's behavior over the last 6 months by rating 18 different home behaviors on severity rating scale of "0" (never or rarely) to "3" (very often). Three global percentile scores were obtained from the ARS: an inattention index, a hyperactivity/impulsivity index, and a total ADHD index. The ARS has been shown to discriminate children diagnosed with ADHD from those children who have learning disabilities, to differentiate children with hyperactivity from children without hyperactivity, and to be sensitive to stimulant drug effects (Barkeley et al., 1990). Behavior ratings scales are widely used to assess ADHD because they are norm based and reliable (Merrell, 1999, Mueller & Tomblin, 2012). Although the use of parent rating scales may result in the overidentification of behavior problems in children with speech and language impairments (Redmond, 2002), to our knowledge there are no data to suggest that the ratings of parents of children with both SSD and LI would be more inflated than the ratings of parents of children with SSD alone. Children meeting rating scale criteria for ADHD, based on the ARS, were those with percentiles greater than 85 on any of the three global scores.

Procedures

The participants were tested individually in two sessions. A licensed and certified speech–language pathologist administered all of the measures. Testing was carried out in a speech research laboratory at Case Western Reserve University or, at the parent's request, in a quiet and adequately lit room in the family's home. The speech productions from the GFTA were recorded using a Sony Professional Tape Recorder (WM-DC6; Walkman Professional, Sony Corporation, New York, NY) and an Audio-Technica Omnidirectional Microphone (AT-804; Audio-Technica U.S., Inc., Stow, Ohio). Responses were recorded initially online using phonetic transcription. In addition, children were administered the standardized language tests according to the instruction manuals. A semistructured interview, the Family History Interview, and the ADHD questionnaire were completed by the examiner with parents. This study was approved by the Human Subjects Committee of University Hospitals Case Medical Center of Cleveland, OH. Informed consent and assent were obtained from participants before testing.

Design and analysis

Chi-square analyses and one-way analysis of variance (ANOVA) were conducted to compare severity groups on demographic variables, articulation and language measures, and the ARS scores. ANOVAs were employed to compare bilineal inheritance and lack of bilineal inheritance groups on the proband's ARS scores. Significant differences were followed by Tukey post hoc analyses.

To examine the influence of predictor variables of SSD severity group, bilineal inheritance, gender and age on the ARS ratings of inattention, hyperactivity/impulsivity, and total rating score, we conducted linear regression analyses. Regressions were conducted in a backward-stepwise fashion, such that all variables were in the starting model, and variables were taken out one at a time starting with the highest p values. At each iteration, we checked how closely the model fit the data by evaluating R^2 values to make sure the highest possible R^2 value was used with the lowest p values for each variable in the model. The final models included only significant variables ($p < 0.05$).

RESULTS

As can be seen in Table 1, the groups (no SSD, resolved SSD, mild SSD, mild-moderate SSD, and moderate-severe SSD) differed significantly in gender, $\chi^2(4, 411) = 14.9, p = .005$, and age, $F(4, 411) = 12.32, p < .0001$. There were proportionally more males than females. The mean age at the time of the initial assessment with the moderate-severe SSD group ($M = 5.69; SD = 2.34$ years) was less than that for the other groups and the no SSD ($M = 8.54; SD = 2.26$) and the resolved SSD ($M = 8.58; SD = 1.66$) were the oldest groups. The groups also differed on the composite language scores of the TOLD-P:3, CELF-P, or CELF-3, with the moderate-severe group performing more poorly ($M = 89.93; SD = 15.18$) than the no SSD, mild SSD, and mild-moderate SSD groups. The no SSD group also scored higher than the resolved and mild-moderate SSD groups. In addition, the no SSD group scored higher on the PIQ measure ($M = 109.18; SD = 15.34$) than the mild-moderate ($M = 102.13; SD = 16.07$) and moderate-severe ($M = 95.99; SD = 14.49$) SSD groups.

Comorbid conditions reported by the parents for participants in each group are shown in Table 2. Language impairment ($n = 166$) was the most frequently reported comorbid condition; the next most commonly reported conditions were RD ($n = 92$) and ADHD ($n = 65$). With the exception of RD, the moderate-severe group had the highest rates of comorbid disorders (LI = 74%; RD = 21%; ADHD = 21%). Although the resolved group had higher rates of RD (46%), this observation is consistent with the older age of this group and the fact that RD is typically diagnosed at an older age.

The severity groups were also compared on the ARS scores of inattention, hyperactivity/impulsivity, and total score. As shown in Table 3, significant differences across groups were observed for the inattention scale, $F(4, 411) = 3.22, p = .013$. In addition, significant differences across groups were observed on the hyperactivity/impulsivity score, $F(4, 411) = 3.20, p = .013$. Finally, significant differences across groups were observed on the total scores, $F(4, 411) = 3.86; p = .004$. Post hoc testing revealed significant group differences between the no SSD and the moderate-severe SSD groups for all three scores at the $p < .05$ level. Effect sizes were small for these comparisons ($\eta^2 = 3.06$ for inattention score; $\eta^2 = 3.05$ for the hyperactivity/impulsivity score; and $\eta^2 = 3.66$ for total score; Cohen, 1973). This indicated that severity of SSD made a small contribution to the differences in inattention, hyperactivity/impulsivity, and total score ratings among the groups.

Comparisons of the bilineal and nonbilineal inheritance groups were made for probands only, because siblings and probands share identical pedigree information. As shown in Table 4, probands with bilineal inheritance demonstrated higher ratings of inattention than probands whose pedigrees were not bilineal, $F(1, 162) = 5.08, p = .026$. No significant differences were observed for the hyperactivity/impulsivity scale or total score. The effect size was small for this comparison ($\eta^2 = 3.06$), indicating that bilineal inheritance made a small contribution to variations in ratings of inattention on the ARS. As shown in Table 5, chi-square analyses revealed no significant differences between the bilineal and not bilineal

groups in severity of SSD or comorbidity of LI; hence, these factors fail to account for the effects of bilineal inheritance on symptoms of ADHD.

Regression analyses were conducted to determine whether severity of SSD was a predictor of ADHD ratings. Predictors in the regression model included severity of SSD, gender, age at ADHD assessment, and comorbid LI. As seen in Table 6, comorbid LI predicted all three scores—inattention ($R^2 = .13$), hyperactivity/impulsivity ($R^2 = .06$), and total ($R^2 = .11$).

A second series of regressions were employed to determine whether bilineal inheritance was a predictor of ADHD ratings. Again, only the probands were included in this analysis so as not to include each family pedigree more than once. Predictors in the regression models were the presence/absence of bilineal inheritance, gender, age at ADHD assessment, and comorbid LI. Bilineal inheritance, age, and comorbid LI predicted the inattention score ($R^2 = .24$). The only predictor for the hyperactivity/impulsivity score was LI ($R^2 = .07$). Predictors for the ADHD total score were gender, age, and LI ($R^2 = .18$; see Table 7).

DISCUSSION

This study examined ADHD symptoms in a clinical population of children recruited for SSD. Previous research suggested that children with SSD and comorbid LI were more at risk for ADHD than children with isolated SSD (McGrath et al., 2007; Short et al., 2008). However, these studies did not consider the phenotypic or genetic heterogeneity within their SSD cohorts. Findings by McGrath et al. (2007) suggested that heterogeneity of SSD may account for the difference in ADHD rates for children with resolved SSD as compared with children with persistent SSD. This study extended findings of previous research by examining differences in symptoms of ADHD as a function of the severity of SSD and bilineal inheritance for disorders.

Severity of SSD and ADHD symptoms

Children with more severe forms of SSD had poorer language scores, as measured by the CELF-P, CELF-3, or TOLD-P:3, and more frequent comorbid LI. The moderate–severe SSD group demonstrated more symptoms of inattention and hyperactivity/impulsivity than the children without SSD. These findings are in partial agreement with those of McGrath et al. (2007) who found that speech and language disorders were associated with inattention disorders. However, McGrath et al. (2007) did not find differences in the hyperactivity ratings as we did, perhaps, in part, because of the older age of their participants. However, as the moderate–severe group had the highest rate of comorbid LI at 74%, it may be that LI rather than SSD is related to inattention and hyperactivity/impulsivity symptoms. This was supported by the regression analyses that found that LI rather than severity of SSD was the most predictive of inattention and hyperactivity/impulsivity ratings. Our findings lend further support to a recent meta-analysis by Ebert and Kohnert (2011), in which they reported that children with LI demonstrate subclinical deficits in sustained attention when compared with typically developing peers.

Our resolved SSD group, children who had a history of SSD but no longer presented with speech sound errors, had the highest rates of RD (46%) and a rate of previous diagnosis of ADHD by a health provider (31%) that was higher than that of the moderate–severe SSD group (21%). These findings are consistent with those of McGrath et al. (2007) who reported that their resolved group had higher rates of ADHD than their persistent SSD group. McGrath and colleagues suggested that SSD in the resolved group may have presented early in life due to neurodevelopmental immaturity, with their speech sound errors resolving with development or intervention or both. Further evidence for this hypothesis of neurodevelopmental immaturity can be seen in the work of Raitano, Pennington, Tunick,

Boada, and Shriberg (2004), who found the resolved SSD group with LI was poorer on rapid serial naming, a processing speed measure, and had lower nonverbal IQs than the resolved SSD group without LI. In our sample, all our SSD groups (resolved, mild–moderate, and moderate–severe) had lower PIQs than the no SSD group.

Bilineal inheritance of SSD and ADHD

To examine the influence of family history for disorders on the comorbidity of ADHD with SSD, pedigrees were sorted according to the presence/absence of bilineal mode of inheritance of SSD. The bilineal and nonbilineal groups did not differ in the severity of SSD or the rates of comorbidity of LI. However, proband children from bilineal families had higher ratings of inattention symptoms than children whose pedigrees did not demonstrate bilineal inheritance. These findings support the association between inattention symptoms and SSD but not hyperactivity/impulsivity symptoms. Results also suggest that a higher genetic load for disorders is associated with inattention symptoms. Although the groups did not differ in rates of LI, regression analyses revealed that comorbid LI was predictive of all three scores on the ARS (inattention, hyperactivity/impulsivity, and total). However, comorbid LI was more closely associated with the inattentive rating than with bilineal inheritance.

There may be several explanations for these findings. First, children whose families demonstrate bilineal inheritance may represent a subtype of SSD with a polygenic mode of transmission of disorders, with many genes making small contributions to the phenotype. These genes may have biological functions that influence cognitive processes or endophenotypes affecting multiple developmental disorders. For example, genes related to slow speed of processing may contribute to SSD, LI, and ADHD. A second explanation is that a major gene or multiple genes may be responsible for the developmental problems of children without a bilineal pattern of inheritance of SSD. These genes may be unique to SSD, LI, or ADHD, but do not influence cognitive processes that are shared across multiple disorders. For example, a gene that influenced oral motor skills may be unique to SSD but may not contribute to LI or ADHD.

Clinical implications

The findings from this study have several clinical implications. First, as reported previously, children with combined SSD and LI are at risk for attention deficits. Longitudinal studies have reported that children with isolated SSD have better academic prognoses than children with comorbid LI (Baker & Cantwell, 1992; Beitchman et al., 1986; King, Jones & Lasky, 1985). Children with SSD and LI are at risk for literacy difficulties (Aram & Hall, 1989; Hall & Tomblin, 1978; Lewis et al., 2000a; Nathan, Stackhouse, Goulondris, & Snowling, 2004). Additional attention difficulties will most likely impede academic success even more. Clinicians should be aware of the comorbidity of these disorders and coordinate remediation strategies for each.

Second, SSDs are heterogeneous and subtypes may differ in their association with comorbid conditions. In this study, we subtyped SSD by severity and found that more severe disorders were associated with attention difficulties. The presence of comorbid attentional impairments complicates the treatment regimen for children with SSD. Although attentional impairments may be treated effectively by psychopharmacological agents, SSD and LI are not. Failure to distinguish attentional problems from language processing difficulties may result in inappropriate treatment of the problem. That is, children may be seen as inattentive when in fact their surface presentation of inattention stems, not from dys-regulation, but rather, from a failure to understand language (Short et al., 2008). Unfortunately, we were unable to separate out the effects of comorbid LI in our SSD sample. Clinicians should be

aware that SSD subtypes may differ in their response to treatment, in part, due to variations in their comorbid conditions. Differential diagnoses, particularly as they pertain to teasing out the presence of LI and attentional problems, are needed to insure that comorbid conditions are identified and treated appropriately.

Finally, family history is important for identifying children who are at risk for comorbid attention deficits. Children with bilineal inheritance should be monitored for attention problems. Although the shared genetic bases for these disorders have not been identified, it is likely that both polygenic and major genes influence SSD and its comorbid conditions.

Limitations and future directions

Several limitations of this study should be noted. First, the ADHD ratings were limited to parent reports, and teacher ratings were not obtained. Teacher ratings of ADHD would likely differ from parent ratings, as teachers observe the child in a more structured setting that requires more linguistic proficiency than the home setting (Redmond, 2002). Redmond and Rice (1998) reported differences between teacher and parent ratings for children with LI, with teachers more stringent in their ratings than parents. Second, the study did not include a group of children with LI only, and, as such, we cannot determine the influences of LI alone on ADHD ratings. Future studies should include this group so that associations of LI and ADHD may be teased out from the influences of SSD. Also, neuropsychological assessments of attention or executive function were not administered. Such measures may help to quantify the deficits in attention and self-regulation and aid in designing remediation programs. Finally, other subgrouping schema besides severity of SSD should be examined, as they might yield different results. One possibility is subgrouping SSD by specific cognitive skill deficits or endophenotypes, including oral motor skills, phonological memory, vocabulary, speeded naming, and phonological awareness skills (Lewis et al., 2011). Another subtyping schema, such as the one proposed by Dodd (2005), which is based on surface speech sound errors, also may be important to consider. Dodd proposed subtypes of articulation disorder, phonological delay, consistent phonological disorder, and inconsistent phonological disorder. Finally, genetic heterogeneity may exist within the bilineal or nonbilineal groups, and, therefore, different phenotypes may be represented within a single group.

The findings from this study highlight the genetic and phenotypic complexity of these developmental disorders. Unique combinations of comorbid conditions may require different treatment strategies, result in diverse outcomes, and represent differing genetic bases. Molecular genetic studies are needed to identify genetic subgroups of SSD with various comorbidities. Ultimately, the identification of pleiotropic and unique genes may result in uncovering neural networks that underlie these comorbidities.

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Table 1
Comparison of SSD severity groups on demographics speech, language, and PIQ measures

	No SSD	Resolved SSD	Mild SSD	Mild-Moderate SSD	Moderate-Severe SSD	Total	η^2	F (df)	p
<i>n</i>	91	39	54	119	109	412			
Male/female	42/49	22/17	33/21	63/56	78/31	238/174		14.94 (4,411) (χ^2)	.0048
Age at SSD classification, mean years (SD)	8.54 (2.26)	8.58 (1.66)	6.26 (2.29)	7.93 (5.41)	5.69 (2.34)	7.31 (3.65)	10.80	12.32 (4,411)	<.0001 ^{a,b,c,d,e,f}
Age at ADHD testing, mean years (SD)	10.15 (4.04)	9.50 (3.26)	8.26 (5.76)	9.12 (6.00)	6.74 (4.05)	8.69 (5.01)	6.36	6.91 (4,411)	<.0001 ^{a,d,f}
GFTA percentile	97.05 (5.93)	95.85 (7.83)	59.87 (8.70)	25.61 (8.84)	5.03 (4.18)	46.99 (37.98)	96.62	2,906.09 (4,411)	<.0001 ^{a,b,c,d,e,f,g,h,i}
PIQ standard score	109.18 (15.34)	102.46 (16.76)	102.85 (18.05)	102.13 (16.07)	95.99 (14.49)	102.31 (16.46)	8.10	8.33 (4,332)	<.0001 ^{d,g}
Language standard score	106.37 (16.59)	94.56 (20.16)	100.48 (17.68)	98.90 (17.04)	89.93 (15.18)	97.96 (17.83)	11.35	12.55 (4,396)	<.0001 ^{d,f,g,i,j}

Note. ADHD, attention-deficit/hyperactivity disorder; GFTA = Goldman-Fristoe Test of Articulation; PIQ = performance IQ; SSD = speech-sound disorders.

^aRecovered SSD differs from moderate SSD.

^bNo SSD differs from mild SSD.

^cMild SSD differs from mild-moderate SSD.

^dNo SSD differs from moderate SSD.

^eRecovered SSD differs from mild SSD.

^fMild-moderate SSD differs from moderate SSD.

^gNo SSD differs from mild-moderate SSD.

^hRecovered SSD differs from mild-moderate SSD.

ⁱMild SSD differs from moderate SSD.

^jNo SSD differs from recovered SSD.

Table 2

Number of participants with comorbid conditions by severity group

	No SSD	Resolved SSD	Mild SSD	Mild-Moderate SSD	Moderate-Severe SSD
Language impairment (<i>n</i> = 166)	11 (12%)	19 (49%)	16 (30%)	39 (33%)	81 (74%)
Reading disorder (<i>n</i> = 92)	21 (23%)	18 (46%)	13 (24%)	17 (14%)	23 (21%)
ADHD (<i>n</i> = 65)	9 (10%)	12 (31%)	9 (17%)	12 (10%)	23 (21%)

Note. ADHD = attention-deficit/hyperactivity disorder; SSD = speech-sound disorders.

Table 3
Comparison of SSD severity groups' performance on ADHD rating scale mean (SD)

	No SSD, <i>M</i> (<i>SD</i>)	Resolved SSD, <i>M</i> (<i>SD</i>)	Mild SSD, <i>M</i> (<i>SD</i>)	Mild-Moderate, <i>M</i> (<i>SD</i>)	Moderate-Severe SSD, <i>M</i> (<i>SD</i>)	Total	η^2	<i>F</i> (<i>df</i>)	<i>p</i>
ADHD inattention scale	38.65 (32.41)	54.08 (35.85)	45.22 (32.90)	43.96 (34.24)	53.83 (33.61)	46.5 (34.02)	3.06	3.22 (4,411)	.0129 ^a
ADHD hyperactivity/impulsivity scale	31.63 (30.32)	45.13 (37.93)	46.36 (34.50)	43.61 (34.18)	47.20 (32.99)	42.4 (33.83)	3.05	3.20 (4,411)	.0132 ^a
ADHD total score	34.81 (29.41)	51.10 (35.62)	44.09(32.51)	45.42 (33.06)	51.84 (32.23)	45.1 (32.69)	3.66	3.86 (4,411)	.0043 ^a

Note. ADHD = attention-deficit/hyperactivity disorder; SSD = speech-sound disorders.

^aNo SSD differs from moderate-severe SSD.

Comparison of probands with bilinear and not bilinear inheritance of SSD on ADHD rating scale

Table 4

	Not Bilinear, <i>M</i> (<i>SD</i>)	Bilinear, <i>M</i> (<i>SD</i>)	Total	η^2	<i>F</i> (<i>df</i>)	<i>p</i>
ADHD inattention scale	47.54 (34.69)	60.35 (33.08)	51.40 (34.54)	3.06	5.08 (1,162)	.0256 ^a
ADHD hyperactivity/impulsivity scale	43.86 (33.58)	51.69 (34.83)	45.89 (34.13)	1.17	1.91 (1,162)	.1687
ADHD total score	47.36 (33.30)	55.76 (32.77)	49.61 (33.23)	1.42	2.32 (1,162)	.1294

Note. ADHD = attention-deficit/hyperactivity disorder.

^aNot bilinear inheritance differs from bilinear inheritance.

Table 5

Comparison of probands with bilineal and not bilineal inheritance on SSD severity and comorbidity of language impairment

	Bilineal	Not Bilineal	χ^2 (df)	<i>p</i>
Resolved SSD	5	4	3.79 (1,162)	.2846
Mild SSD	4	11		
Mild–moderate SSD	13	37		
Moderate–severe SSD	32	57		
SSD alone	18	39	0.0950 (1,162)	.7579
SSD + LI	36	70		

Note. SSD = speech-sound disorders.

Table 6
Predictors of ADHD ratings of inattention, hyperactivity/impulsivity, and total ADHD scores

Variable	Predictor	β	b (SE)	p	R ²
ADHD inattention scale	Language impairment	25.16	3.19	<.0001	.13
ADHD hyperactivity/impulsivity scale	Language impairment	16.72	3.30	<.0001	.06
ADHD total score	Language impairment	22.40	3.10	<.0001	.11

Note. ADHD, attention-deficit/hyperactivity disorder.

Predictors of ADHD ratings of inattention, Hyperactivity/Impulsivity and Total ADHD scores by bilineal inheritance (probands only)

Table 7

Variable	Predictor	β	b (SE)	p	R ²
ADHD inattention scale	Bilineal	11.60	5.06	.0231	.24
	Age	4.39	1.61	.0071	
ADHD hyperactivity/impulsivity scale	Language impairment	29.55	5.00	<.0001	.07
	Language impairment	19.26	5.35	.0004	
ADHD total score	Gender	10.26	5.08	.0450	.18
	Age	3.03	1.46	.0398	
	Language impairment	25.48	4.99	<.0001	

Note. ADHD, attention-deficit/hyperactivity disorder.