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Heritability and longitudinal outcomes of spelling skills in individuals with histories of early speech and language disorders

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Abstract

This study examined the spelling skills in middle childhood and adolescence in individuals with histories of early childhood speech sound disorders (SSD) with and without language impairment (LI). Youth without such histories were also included (No SSD/LI group). The heritability of spelling skills at each age level was estimated. Children with SSD were classified as SSD-only, SSD with LI but without childhood apraxia of speech (SSD + LI/ No CAS), and CAS and LI (CAS + LI). The SSD-only group did not differ in spelling from the No SSD/LI group, suggesting that SSD-only did not increase risk for poor spelling. The SSD + LI/No CAS and CAS + LI groups had poorer spelling skills than the SSD-only and No SSD/LI groups. Spelling was associated with phonological awareness in the middle childhood and adolescent samples and with rapid automatized naming in the adolescent sample. Heritability of spelling skills was stronger in adolescence than in middle childhood. Differences in the correlates of spelling and in heritability at the two ages suggest developmental changes in the factors contributing to spelling.

1. Introduction

Over the past two decades, links between spoken language and literacy skills have been identified and a body of literature has emerged that indicates that children with early spoken language deficits are at risk for later literacy difficulties. Although much of the research has focused on the relationship between language impairment and reading skills (Bishop & Snowling, 2004; Catts, 1996; Catts, Adlof, Hogan, & Weismer, 2005; Catts, Bridges, Little, & Tomblin, 2008; Conti-Ramsden, Durkin, Simkin, & Knox, 2009; Kamhi & Catts, 2012), children with speech sound disorders (SSD), with accompanying language impairment (LI), may be at risk for spelling difficulties (Lewis, Freebairn, & Taylor, 2002). SSD, defined as a

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delay in acquiring speech sounds, are common communication disorders experienced by young children with an estimated 3.8% of 6-year-olds demonstrating SSD (Shriberg, Tomblin, & McSweeny, 1999). Although most SSD resolve by 8 years of age, more than half of these children continue to present with spelling difficulties (Bird, Bishop, & Freeman, 1995; Lewis et al., 2002).

Studies examining spelling problems in children with SSD have focused on primarily on early spelling difficulties and have not followed students into adolescence to determine if spelling difficulties persist (Bird et al., 1995; Hayiou-Thomas, Carroll, Leavett, Hulme, & Snowling, 2017; Lewis et al., 2002; Overby, Masterson, & Preston, 2015). Evidence from genetic studies indicates that spelling skills have greater heritability at older ages, and that environmental factors, such as exposure to print and instruction in spelling, are more strongly related to spelling skills at younger ages (DeFries, Stevenson, Gillis, & Wadsworth, 1991; Friend, DeFries, Wadsworth, & Olson, 2007; Stevenson, Graham, Fredman, & McLoughlin, 1987). However, these studies have primarily included typical children or children with reading disabilities and have not focused on individuals with histories of SSD or identified the cognitive-linguistic abilities associated with spelling at younger and older ages. The present study examined spelling skills in middle childhood (7–11.9 years) and adolescence (12–17.9 years) in a large cohort of students with prior histories of SSD. Our primary aims were to determine if spelling difficulties were evident at both ages, to explore potential age differences in the skills associated with spelling, and to identify differences in spelling outcomes among clinical subtypes of SSD. The second aim was to determine if spelling skills had greater heritability in adolescence than in middle childhood, as reported in studies of typically developing students (Friend et al., 2007; Stevenson et al., 1987).

1.1. SSD and risk for spelling difficulties

Previous studies demonstrated that children with SSD are at risk for spelling difficulties (Bird et al., 1995; Hayiou-Thomas et al., 2017; Lewis et al., 2002; Overby et al., 2015; Peterson, Pennington, Shriberg, & Boada, 2009). These difficulties may result from weaknesses in mapping phonemes to graphemes (Overby et al., 2015), reduced phonological memory and/or perception (Fowler, 1991), and inaccurate or “fuzzy” phonological representations (Lewis et al., 2002). Any of these basic skill deficits in children with SSD may undermine proficiency in spelling and reading (Gillon, 2005; Gillon & Moriarty, 2007).

Our previous studies have indicated that children with histories of SSD with co-morbid LI are more at risk for literacy difficulties than children with histories of SSD-only (Lewis et al., 2002; Lewis et al., 2015; Lewis, O'Donnell, Freebairn, & Taylor, 1998). Children with SSD-only were found to demonstrate a weakness in spelling relative to their language and reading scores (Lewis et al., 2002). Paired *t*-test comparisons revealed that children with SSD-only performed more poorly on the spelling measure than the language or reading decoding measure. One explanation for this finding may be that although word recognition requires only partial identification of orthographic and phonological patterns, spelling requires more complete orthographic and phonological representations (Friend et al., 2007). Another study that employed factor analysis demonstrated that articulation and phonology skills in early childhood are related to later spelling abilities, partially independent of

language skills (Lewis et al., 2006). Spelling impairment at school-age was also predicted by early childhood use of phonological processes, word discrimination, and syntax (Lewis et al., 2002). Taken together these findings confirm that children with early childhood SSD and LI are at greater risk for spelling difficulty than children with SSD-only. An SSD may impact spelling skills to a lesser extent than LI possibly due to phonological processing deficits.

Past research reveals an association of speech errors with spelling difficulties at early school age (Gillon, 2002; Hayiou-Thomas et al., 2017; Overby et al., 2015; Raitano, Pennington, Tunick, Boada, & Shriberg, 2004). Children with different clinical subtypes of SSD may also be at differential risk for spelling difficulties. For example, spelling disabilities in children with SSD and comorbid LI, one subtype of SSD, likely reflect problems in phonological representations and other domains, such as semantics, syntax and morphology. Hayiou-Thomas et al. (2017) reported that literacy difficulties at 8 years of age were related to the persistence of the SSD, disordered speech versus delayed speech, co-morbid LI, and a family history of dyslexia, but that these difficulties were not associated with the severity of the SSD at 3 ½ years of age.

A subtype of SSD is childhood apraxia of speech (CAS). CAS is a severe form of SSD with impairment in the planning and/or programming of speech movement sequences (ASHA, 2007). There is disagreement as to the core diagnostic features of CAS. Some view CAS as solely a motor programming deficit that may also present with difficulties in the suprasegmental aspects of speech (Shriberg et al., 2017). Others consider CAS to be a symptom complex that includes linguistic deficits, especially with phonological representations (Aram, 1984; Marion, Sussman, & Marquardt, 1993; Marquardt, Sussman, Snow, & Jacks, 2002; Velleman & Strand, 1994). Froud and Khamis-Dakwar (2012) provided evidence that children with CAS demonstrate phonological over-specification. They concluded that CAS has a core phonological component in addition to a motor planning component. Other studies have examined auditory perceptual deficits in children with CAS (Nijland, 2009) and found children with CAS to show auditory perceptual deficits for both higher and lower order linguistic input. Deficient phonological representations and impaired auditory perceptual deficits may place children with CAS at greater risk for spelling difficulties than children with other types of SSD. Stackhouse and Wells (1997) hypothesized that accurate phonological representations needed for spelling are not formed as the child with CAS is unable to produce complex words, words are not correctly encoded, and auditory feedback is disrupted. The child with CAS may also have cognitive and phonological deficits, or difficulties in morphology and syntax (Gillon & Moriarty, 2007; Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004) that impair spelling skills. In our previous work we demonstrated that weakness in phoneme segmentation and phoneme sequencing in children with CAS were associated with poorer speech production than in children with other subtypes of SSD and spelling problems (Lewis et al., 2004). In the current study we focused on spelling skills in a sample of children with early childhood SSD to examine potential differences related to the type of SSD, underlying cognitive skills, and age at assessment.

1.2. Endophenotypes for spelling

Endophenotypes are conceptualized as the basic cognitive-linguistic competencies that contribute to acquisition of more complex traits such as spelling with potential utility in detecting genetic influences (Gottesman & Gould, 2003). Phonological awareness (PA) is an endophenotype hypothesized to underlie speech, language, reading and spelling (Nathan, Stackhouse, Gouldandris, & Snowling, 2004; Raitano et al., 2004; Lewis et al., 2011). Poor PA may result in inaccurate mappings of phonemes onto graphemes. PA is dependent on strong phonological representations that may be deficient in children with SSD, as well as some children with LI (Bird et al., 1995; Preston & Edwards, 2010; Rvachew & Grawberg, 2006) and may also contribute to difficulties in encoding phonemes onto graphemes (Ball & Blachman, 1991; Catts, Kamhi, & Adlof, 2012).

Another endophenotype in common with reading and spelling abilities is speed of name retrieval as assessed by rapid automatized naming (RAN), a skill that has been hypothesized to tap lexical access and phonological processing (Denckla & Rudel, 1976; Norton & Wolf, 2012). RAN requires naming of a small set of randomly presented colors, objects, numbers or letters as quickly as possible. Although most studies have examined the association of RAN performance to reading abilities, cognitive skills that are associated with RAN may also underlie spelling skills (Stainthorp, Powell, & Stuart, 2013). RAN predicts spelling disability independent of PA in typical children and children with reading disabilities, suggesting separate spelling-related processes (Furnes & Samuelssen, 2010; Norton & Wolf, 2012). These spelling-related processes include the graphotactics, orthography, and phonological representations, rapid access and retrieval of phonological information, and oral motor skills—all skills that may be impaired in children with SSD (Norton & Wolf, 2012; Treiman, 2017). Stainthorp et al. (2013) demonstrated that children with good PA but poor RAN skills were poorer in spelling skills than matched controls, especially in spelling irregular words such as “yacht”. They concluded that RAN was associated with the establishment of fully specified orthographic representations. PA skills, on the other hand, may be more associated with phonological decoding. The present study examined the contribution of PA and RAN to spelling skills at middle childhood and adolescence in individuals with histories of SSD.

1.3. Genetics of spelling disorders

In our previous work, we noted familial aggregation of poor spelling skills in family pedigrees of children with SSD (Lewis et al., 2002). Although school-age children with combined SSD and LI performed more poorly than children with SSD-only on spelling measures, children with SSD-only exhibited a weakness in spelling skills relative to their reading skills, language abilities and performance IQ. These findings suggest that spelling in particular may be difficult for children with a history of SSD and that these spelling difficulties aggregate within families.

Compared to the many family and genetic studies of reading, few studies have examined spelling skills independently of a reading phenotype. Rarely have studies included individuals with histories of SSD. The few studies that have examined spelling skill independent of reading indicate that this skill is highly heritable, with estimates the

heritability (h^2) from twin studies ranging from 0.62 to 0.73 when controlling for intelligence (DeFries et al., 1991). There is also evidence that the relative contributions of genes and environment to spelling skills varies with age. An early study (Stevenson et al., 1987) suggested that spelling skills may be more genetically mediated than reading with genetic influences on spelling skills increasing with age. A study that tested heritability of reading and spelling skills in younger (8–10.5 years) and older twins (10.6–20 years) reported heritabilities for reading as measured by word recognition of 0.64 for younger twins and 0.58 for older twins (Friend et al., 2007). The heritability of spelling skills was 0.51 for the younger group and 0.67 for the older group. However, these group differences did not reach statistical significance.

The hypothesis of increasing genetic influences on spelling skills with age is consistent with increasing genetic influences with age reported for other cognitive traits such as IQ (Bouchard, 2013; Plomin & Deary, 2015; Tucker-Drob, Briley, & Harden, 2013). Genes are turned on and off during development. The genes at work at adolescence and adulthood may not be as influenced (modified) by the environment as are genes active early in development. Another explanation for increasing heritability with age is that at an early age the child does not control his or her environment. As the individual ages the child selects, modifies, and creates environments that are compatible with their genetic predisposition (Plomin & Deary, 2015). However, as these previous studies focused on twin pairs only, they may have overestimated heritability, as it is difficult to separate the variance due to genes and shared environment in twins (Visscher, Hill, & Wray, 2008).

1.4. Goals and hypotheses of the current study

The current study includes two aims. The first aim was to examine spelling skills in a sample of children with early childhood histories of SSD followed longitudinally to middle childhood and adolescence. The children were originally recruited as part of a longitudinal study that aimed to identify predictors of outcomes for children with different subtypes of early SSD. Children were divided into three groups including SSD-only, SSD with LI but not CAS (SSD + LI/No CAS), and CAS with LI (CAS + LI). A group without histories of SSD or LI (No SSD/LI) was also assessed. Group differences in spelling and associations of spelling with language and other cognitive abilities including phonological awareness (PA) and rapid automatized naming (RAN), were examined at two age levels in a longitudinal cohort (middle childhood, 7–11.9 years; adolescence, 12–17.9 years). The second aim was to estimate heritability of spelling skills at each age level. Specific research questions addressed were:

1. Do children with histories of SSD differ from children without histories of SSD (No SSD/LI group) in spelling skills in middle childhood and in adolescence? Do children with different subtypes of SSD (SSD-only, SSD + LI/no CAS, and CAS + LI) differ from one another in spelling skills in middle childhood and in adolescence? Based on previous research findings, we hypothesized that individuals with a history of early SSD would have weaker spelling skills compared to the No SSD/LI group at both age levels. We further hypothesized that children with histories of SSD + LI/no CAS would perform more poorly than those with SSD-only due to combined phonology and language deficits and

that children with CAS + LI would have the greatest deficits in spelling as they had the most severe SSD and LI in our cohort.

2. Are spelling skills heritable and does heritability differ for younger compared to older youth? Based on previous research findings, we hypothesized the spelling skills in middle childhood would be more strongly associated with environmental factors whereas spelling in adolescence would be more heritable. Heritability of spelling skills may become more pronounced at older ages as the individual is required to spell more complex words. Differences in heritability among younger students may not be as pronounced as the spelling of less complex words may be more related to instruction and exposure to words (environment). Different genes may be influencing spelling skills at different ages. Genetic influences at adolescence may be less modified by the environment than genetic influences at younger ages.

2. Method

2.1. Participants

This project was approved by the Institutional Review Board of University Hospitals Case Medical Center. The research was conducted in accordance with human subject guidelines and principles. Informed consent was obtained from the parent or guardian and assent was obtained from the child prior to enrollment in the study.

The data for this study were obtained as part of a larger longitudinal family study of speech and language disorders (Lewis et al., 2011). The sample included 408 children evaluated in middle childhood (7–11.9 years) and 271 evaluated in adolescence (12–17.9 years). The middle childhood sample comprised 155 probands with histories of SSD, 103 siblings of the probands with SSD, and 146 siblings without SSD. The adolescent sample comprised 84 probands with histories of SSD, 68 siblings of these probands with SSD, and 109 siblings without SSD. Families were originally recruited into the study on the basis of having a child with SSD who was 4–6 years of age at the time of enrollment (probands). Recruitment was from the clinical caseloads of speech-language pathologists working at community speech and hearing centers, schools, or in private practice in Northeast Ohio.

Probands were screened to ensure that they met the following criteria: 1) moderate to severe expressive SSD as defined by a score of 1 SD or greater below the mean on the Goldman-Fristoe Test of Articulation Sounds-in-Words subtest (Goldman & Fristoe, 1986; Goldman & Fristoe, 2000) and by commission of at least 3 phonological error types as identified by the Khan-Lewis Phonological Analysis (KLPA; Khan & Lewis, 1986, 2002), with speech sound differences due to dialect being scored as correct; 2) normal hearing acuity as defined by passing a pure tone audiometric screening test at 25 dBHL ISO for 500, 1000, 2000, and 4000 Hz bilaterally and < 6 episodes of otitis media prior to 3 years as reported by the parent; 3) normal peripheral speech mechanism as documented by the Total Structure Score on the Oral and Speech Motor Control Protocol (Robbins & Klee, 1987) or, for children older than 5 years, by intact oral structures as measured using the Oral Speech Mechanism Screening Exam—3rd Edition (St. Louis & Ruscello, 2000); 4) absence of a history of

neurological disorders or developmental delays other than speech and language as reported by the parent; and 5) normal intelligence defined as a Performance IQ ≥ 80 on the Wechsler Preschool and Primary Scale of Intelligence—Revised (Wechsler, 1989) or Wechsler Intelligence Scale for Children, 3rd Ed. (WISC-III, Wechsler, 1991). Family socioeconomic status (SES) was determined at the initial assessment based on parent education level and occupation using the Hollingshead Four Factor Index of Social Class (Hollingshead, 1975).

All available siblings of the probands were also invited to participate in the testing. Siblings were categorized as having a SSD if they met the same criteria described above for probands and/or had a history of speech language therapy for a SSD. All probands and siblings were invited to participate in follow-up assessments in middle childhood (7 to 11.9 years, $M = 9.4$; $SD = 1.4$) and/or adolescence (12 to 17.9 years; $M = 14.5$; $SD = 1.7$).

Because some children were not followed to adolescence and some adolescents were not assessed in middle childhood, the samples were not identical. However, 133 (33%) of the 404 children in the middle childhood sample were also seen in adolescence. When comparing youth seen only in middle childhood to those seen only in adolescence, the subset of children seen only at middle childhood had a higher prevalence of SSD (62% versus 24%; $p = 0.01$) and LI (36% versus 27%; $p = 0.01$). In our previous study of adolescents (2015), we observed a similarly lower prevalence of communication disorders in children assessed at later compared to earlier ages. Likely reasons for age differences in sample make-up are differential loss to follow-up of children with these disorders and inclusion of more unaffected siblings at the older age level. Among the middle childhood youth, there was no significant difference in the prevalence of SSD, LI, or CAS in the children that were followed to adolescence versus those that were not followed ($p > 0.05$, data not shown).

In early childhood (4–6 years of age), probands and siblings who met criteria for a SSD were further classified into SSD-only, SSD + LI/ No CAS, and CAS + LI groups. The SSD-only group and the SSD + LI group did not present with CAS. Children with SSD-only obtained scores > 1.25 standard deviations below the mean on all subtests of the Test of Language Development—Primary:2 (TOLD-P:2, Newcomer & Hammill, 1988) or the Test of Language Development—Primary:3 (TOLD-P:3, Newcomer & Hammill, 1997). Children with SSD + LI met the criteria for language impairment adapted from Tomblin and Buckwalter (1998), which included a score equal to or < 1.25 SD below the mean on at least two of the composite scores of the TOLD-P:2 or TOLD-P:3. The diagnosis of CAS was based initially on a provisional diagnosis by the child's speech language pathologist. The children were then screened by members of the research team for a speech production disorder involving deficits in motor programming. A final diagnosis of CAS required that the child demonstrate at least four commonly reported characteristics of CAS suggestive of an oral-motor programming deficit. These characteristics included difficulties sequencing phonemes and syllables, trial and error groping behaviors, prosodic disturbances, metathetic errors, decreased diadochokinetic rates, consonant deletions, increased errors on polysyllabic words, and inconsistency in articulation with unusual error forms on consonants and vowels (Hall, Jordan, & Robin, 1993; Stackhouse, 1982; Velleman & Strand, 1994). All children with CAS also met the criteria for LI. See Lewis et al. (2015) for a detailed description of the cohort.

2.2. Measures

2.2.1. Phonological awareness (PA)—Elision was originally assessed with the pre-publication version of the Comprehensive Test of Phonological Processing and later with the published version (CTOPP, Wagner, Torgesen, & Rashotte, 1999). This test requires the child to say how an orally presented word would sound with a given part of the word deleted. Scaled scores were used in data analysis. Reliability for the Elision Subtest for individuals with speech-language disabilities is 0.93 according to the test manual.

2.2.2. Rapid automatized naming (RAN)—Rapid naming of colors was administered to assess speed of lexical access, or RAN (Denckla & Rudel, 1976). In this test the child names several rows of different colored squares as quickly as possible on a large stimulus sheet. *Z*-scores for the time taken to name all of the colored squares were computed based on the age-related means and standard deviations provided by Denckla and Rudel (1976). Although the reliability of the RAN was not provided in the original article (Denckla & Rudel, 1976), the rapid naming of colors for individuals with speech and language disorders on a similar test has been reported as 0.83 (CTOPP; Wagner et al., 1999).

2.3. Reading decoding

Reading decoding was assessed by administering the Word Identification (WI) and Word Attack (WA) subtests of the Woodcock Reading Mastery Tests—Revised (Woodcock, 1987). WI assesses the child's skill in reading real single words. WA assesses the child's skill in decoding non-words. Performance was defined in terms of age-adjusted standard scores. Split-half reliability coefficient for the WI subtest is 0.98 and the WA is 0.87 according to the test manual.

2.4. Spelling

Spelling was assessed on the Test of Written Spelling—Third Edition (TWS-3, Larsen & Hammill, 1994). The TWS-3 is composed of predictable words that evaluate the child's ability to spell dictated words that are spelled like they sound such as “stop” and “ambiguous” and unpredictable words that measures the ability to spell words that are exceptions to common spelling patterns such as “two,” “eight,” and “feign.” Age-adjusted standard scores were used in the analyses. Inter-scorer reliability for the TWS-3 is 0.98 according to the test manual.

2.5. Data analysis

Analysis of variance (ANOVA) and chi square analyses were conducted at each age level to compare the early childhood classifications of No SSD/LI, SSD-only, SSD + LI/No CAS, and CAS + LI on age, sex, diagnosis of a reading disorder and SES. ANOVAs were also employed to compare these groups on the TWS-3, PA, RAN, WI and WA. Tukey's tests were used in post-hoc group comparisons. To account for family relationship, ANOVA and Tukey's tests were performed using a linear mixed model (lmer) in the R package, using family membership as the random effect. To identify predictors of spelling scores at each age level, regression analyses were carried out co-varying for SES and using the mixed models package (lme4) in the R package to control for sibling clusters. Although our

primary interest was in associations of spelling with PA and RAN, reading decoding (WI) was controlled in these analyses to account for the well-established association of spelling and word recognition and to allow examination of the associations with PA and RAN that were unique to spelling. To account for the association of SSD classifications with spelling, models additionally adjusted for the presence/absence of SSD and/or LI based on early childhood assessments. The variance inflation factor failed to reveal evidence for multicollinearity.

Heritability may be estimated using a variety of study designs, including twins, parents and offspring, and sibling pairs (Rice & Borecki, 2001). Historically, the twin design was most commonly used for heritability estimation, but recent studies have noted the statistical limitations of that design for precise estimation (Benchek & Morris, 2013, Visscher, Hill, & Wray, 2008). An alternative approach, estimating the total polygenic variance from relative pair data, reduces the bias seen in the twin design and incorporates gene-gene interaction effects (Morris, Elston, & Stein, 2010). Polygenic variance is estimated using the kinship coefficient matrix, using the same mathematical principles as recently developed heritability estimation methods for unrelated subjects (Yang et al., 2010). A further description of these methods can be found in Kim et al. (2015). Thus, we used structural equation modelling (SEM) to partition spelling at each age level into heritable components, random environmental components, and the influence of SES. SEM analyses utilized the “strum” package (Song, Stein, & Morris, 2015), which implements a generalized framework for SEM for family data (Morris et al., 2010). As this analysis only included participants that had complete data at both time points, it was a longitudinal model.

3. Results

3.1. Group differences

Table 1 presents demographics for the middle childhood and adolescent samples by group (No SSD/LI, SSD-only, SSD + LI/No CAS, and CAS + LI). Although the groups did not differ significantly in age for either sample, there were group differences in SES at both age levels. Gender differences were also evident for the middle childhood sample, with a more equal gender distribution for the No SSD/LI group (54% females) and proportionally fewer females in the SSD groups (range 25%–43%).

At follow-up, participants were assessed to determine if speech sound errors were resolved or persistent. Children’s speech sound errors were considered resolved if they made no errors on the Goldman-Fristoe Test of Articulation. Sixty-seven percent of the SSD only group, 66% of the SSD + LI, no CAS group, and 36.7% of the CAS group resolved their speech sound errors. Also at follow-up, participants were assessed to determine if spoken language impairments were resolved. Children’s language impairment was considered resolved if they scored at or > 1 SD below the mean of the Clinical Evaluation of Language Fundamentals (CELF) or the Test of Language Development (TOLD). Ninety-eight percent of the SSD only group, 56% of the SSD + LI, no CAS group and 37.2% of the CAS group resolved their language disorder.

Significant group differences were observed on the TWS-3, WI, WA, PA, and RAN tests at both age levels. Fig. 1A and B reveal similar mean TWS-3 scores for No SSD/LI and SSD-only groups. For the middle childhood sample, the SSD + LI/no CAS and CAS + LI groups had significantly lower spelling scores than the No SSD/LI and SSD-only groups and the CAS + LI had significantly lower scores than the SSD + LI/No CAS group (Fig. 1A and Table 2). For the adolescent sample, the SSD + LI/No CAS group did not differ from the CAS + LI group, but the SSD + LI/No CAS and CAS + LI groups had lower scores than the No SSD/LI and SSD-only groups (Fig. 1B and Table 2).

Similarly, for the middle childhood sample, the SSD + LI/No CAS and CAS + LI groups had significantly lower scores on PA, RAN, WI, and WA than the No SSD/LI and SSD-only groups. For the adolescent sample, the SSD + LI/No CAS and CAS + LI groups had significantly lower scores on PA, WI, and WA than the No SSD/LI and SSD-only groups, but only the CAS + LI group had significantly lower scores on the RAN than the No SSD/LI and SSD-only groups. Table 2 presents group comparisons of children with speech sound disorders on measures at middle childhood and adolescence. As seen in Table 2, effect sizes were small at both ages. See Appendix A for Cohen's d effect sizes of subtype comparisons on each measure.

3.2. Correlates of spelling

Table 3 shows predictors of total spelling scores at middle childhood and adolescence. After adjustments for SES, single word reading, and SSD classification, spelling was associated with PA ($p = 0.003$) in middle childhood, and with PA ($p = 0.011$) and RAN ($p = 0.015$) in adolescence. Results were similar in models that were adjusted for the effects of SSD classification. As expected, single word reading as assessed by WI was significantly associated with spelling at both age levels (p 's < 0.001).

3.3. Heritability estimation

To estimate heritability at middle childhood and adolescence a subset of children who were seen at both time points were employed ($n = 133$). In this sample, there were 2 twin pairs and no half siblings. We randomly removed one child from each of the twin pairs for this analysis, leaving 131 participants included in the model. Participant demographics for this sub-sample are presented in Table 4.

As illustrated in Fig. 2, findings from the SEM model revealed that polygenic variance contributed significantly to spelling scores for the adolescent sample ($h^2 = 91\%$, 95% confidence interval 50%–100%, $p < 0.001$) but not for the middle childhood sample ($h^2 = 27\%$, 95% confidence interval 0%–88%, $p = 0.2$). Conversely, estimated unique environmental (error) variance contributed significantly to spelling scores for the middle childhood sample (69%, 95% confidence interval 12–100%, $p = 0.01$) and not for the adolescent sample (5%, 95% confidence interval 0–50%, $p = 0.4$). Although the genetic influence on spelling at adolescence and environmental influence at middle childhood were significant, the covariances due to genetic and environmental influences between the two time points were nonsignificant.

4. Discussion

This project examined differences in spelling skills in middle childhood and adolescence in clinical subtypes of individuals with histories of SSD and identified cognitive-linguistic and genetic factors associated with these skills at each age level. Previous studies indicated that students with SSD were at risk for spelling difficulties but these studies did not examine skills at adolescence nor did they report heritability estimates of spelling skills for students with SSD. The findings from our study revealed that individuals with histories of early childhood SSD have poorer spelling skills than children without such histories when the SSD was accompanied by LI in middle childhood and adolescence. As reviewed below, the results also document subtypes differences in spelling and age-related differences in the cognitive-linguistic correlates and genetic basis of spelling.

4.1. Spelling skills of clinical subtypes of SSD

Children with SSD are a heterogeneous group. We hypothesized that clinical subtypes of SSD-only, SSD + LI/No CAS, and CAS + LI differ in the severity of their spelling deficits. Children with SSD-only and children with no history of SSD (No SSD/LI/CAS) did not differ in spelling skills. As anticipated, youth with more severe disorders such as CAS + LI scored more poorly on a spelling measure than other youth in the sample, including those with other forms of SSD (SSD-only and SSD + LI/No CAS) as well as those without these disorders (No SSD/LI). Individuals in both of the LI groups (i.e. SSD + LI/No CAS or CAS + LI) also performed more poorly on spelling measures than individuals with SSD-only or No SSD/LI. The SSD-only group did not differ from the No SSD/LI, suggesting that SSD alone does not increase risk for spelling difficulties. These differences in spelling skills among clinical subtypes persisted into adolescence, with the exception that the CAS + LI group performed more poorly than the SSD + LI/No CAS group only in middle childhood. See Appendix A for effect sizes of subtype comparisons on each measure.

The differences in spelling skills observed among the clinical subtypes may be related to differences in underlying endophenotypes such as PA or RAN, or to differences in reading skills. At both age levels, comparisons of subtypes revealed significant differences in PA, RAN, and single word reading, with the SSD + LI/No CAS and CAS + LI groups performing more poorly on all measures than the SSD-only and No SSD/LI groups. The lack of differences between the SSD + LI/No CAS and CAS + LI groups at adolescence may reflect the fact that both groups presented with LI. Although the speech sound difficulties may resolve by adolescence in both groups, LI in domains such as vocabulary and morphology may persist and continue to impact spelling skills. The findings raise the possibility that spelling interventions for children with histories of early SSD may be more effective if targeted to the deficits underlying children's spelling difficulties.

4.2. Endophenotypes associated with spelling skills

We further explored the role of these endophenotypes in spelling by determining predictors of spelling skills. After accounting for the effects of SSD and LI, PA was associated with spelling skills at both age levels. In contrast the RAN was associated with spelling skills only in adolescence. A substantial number of studies have associated RAN with dyslexia

(Wolf, Bowers, & Biddle, 2000), however few have examined the relationship between RAN and spelling skills. Studies have demonstrated PA and RAN to be early predictors of later reading difficulties (Pennington & Lefly, 2001; Schatschneider et al., 2004). Although both RAN and PA require retrieval of phonological codes, they are not strongly correlated ($r = 0.38$; Swanson, Trainin, Necochea, & Hammill, 2003). The RAN and PA account for unique variance in decoding skills. The contribution of RAN to reading skills changes with age. Scores on the RAN in second grade were predictive of reading skills in 8th grade (Byrne et al., 2005; Petrill et al., 2006).

Performing the RAN task requires the coordination and integration of a range of cognitive processes. Wolf and Denckla (2005) list attentional and visual processes and the integration of graphotactic information with phonological representations and the access and retrieval of phonological information that may be related to spelling performance (Norton & Wolf, 2012; Wolf & Denckla, 2005). In the current study we employed the RAN colors (rather than letters) to eliminate orthography from this measure. Students with a weakness in orthography were not penalized in this rapid naming task. We were able to examine speed of visual processing and semantic retrieval. Phonological encoding of the color names for speech production is also required by this task. Individuals with SSD may have deficient phonological representations and/or a weakness in accessing these phonological representations (Ramus, Marshall, Rosen, and Lely, 2013). However, Norton and Wolf (2012) note that RAN and PA are distinct tasks that tap unique cognitive skills with distinct neurological and genetic bases. Genetic studies of RAN, PA and reading have demonstrated that RAN and PA have both shared and unique genetic components (Byrne et al., 2005; Compton, Davis, DeFries, Gayan, & Olson, 2001; Petrill, Deater-Deckard, Thompson, DeThorne, & Schatschneider, 2006).

4.3. Heritability of spelling skills

Spelling skills in middle childhood and adolescence are partially heritable but, as demonstrated by our findings (see Fig. 2) and as found in studies of typical children (Friend et al., 2007), are also associated with SES. The associations with SES were especially robust in the middle childhood sample, suggesting that environment and instruction are more critical to the early development of spelling skills. We included SES in our model, but there may be other unknown environmental factors contributing to spelling such as family environment, neighborhood school, print exposure, environmental toxins, nutrition, number of siblings, experiences such as visits to the library, and number of books in the home.

Strong heritability was observed at adolescence ($h^2 = 0.91$) but not at middle childhood ($h^2 = 0.27$), suggesting that environmental factors may contribute more to the early phases of spelling acquisition whereas genetic factors are more important determinants of higher level spelling skills. These results are generally consistent with previous studies of typical children that estimated heritability of spelling skills to range from 0.62 to 0.73 when controlling for intelligence (DeFries et al., 1991). Although our estimate for the adolescent sample was above this range, we did not control for intelligence and our sample comprised individuals with comorbid speech and language disorders who had poorer than average spelling skills. Although selective inclusion in this study of children at both higher and lower

risk for spelling difficulties may be responsible for the higher heritability estimate, previous research indicated similar genetic influences in good and poor readers (Boada et al., 2002; DeFries et al., 1991). Our findings also confirm that genetics contribute more to variability in spelling skills with advancing age and thus are consistent with findings by Friend et al. (2007). Greater heritability of spelling in adolescence may reflect the increasing complexity of spelling demands.

Other cognitive skills, such as intelligence, also show increased heritability with age (Plomin & Deary, 2015). An explanation for this increase in heritability offered by Plomin and Deary is not that the genes for intelligence differ by age, but rather that genetic-environment interactions magnify the effects of genes as youth modify their own environments to better suit their genetic predispositions. If spelling is environmentally mediated at a young age, it is critical to monitor the spelling abilities of children with SSD and provide intervention as warranted.

The heritability estimation method used in this study may be based on different assumptions than methods applied in previous studies. Our method estimates polygenic variance (Morris et al., 2010; Song et al., 2015), whereas twin studies estimate additive genetic variance and sometimes dominance genetic variance. Estimates of polygenic variance using the kinship coefficient matrix may be higher because they include both additive and dominance genetic variance as well as gene-gene interaction effects. In addition, the specific measure of spelling skills examined in this study may have differed from those used in previous research, which could also contribute to different heritability estimates. Furthermore, we included SES in our models, which partially accounts for shared environment. Our estimates are unlikely to be upwardly biased due to gene-environment interaction effects.

Further evidence for a genetic basis for spelling has been provided by molecular genetic studies. Schulte-Körne et al. (1998) examined 7 multiplex families with spelling disability and found linkage to 15q21. Froster, Schulte-Korn, Hebebrand, and Remschmidt (1993) found spelling and reading difficulties associated with an expressive speech disorder related to a balanced translocation on chromosome 1 (1p22; 2q31). More generally spelling disorders have been linked to chromosomes 1p, 2p, 2q, 3p, 4p, 6q, 7q, 9p, 15q, 17p, 18p, and Xq (Bates et al., 2006; Rubenstein, Matsushita, Berninger, Raskind, & Wijsman, 2011; Schulte-Körne, 2001; Schulte-Körne et al., 1998). However, these studies recruited probands with reading disorders who may not have had SSD. If spelling disorders are related to heritable deficits in phonological representations, individuals with histories of early childhood SSD are at genetic risk for spelling deficits. Endophenotypes of PA and RAN skills are highly heritable ($h^2 = 0.63$ for PA; $h^2 = 0.64$ for RAN; Byrne et al., 2005) and may underlie co-morbid disorders of SSD, reading disorder, and spelling difficulties.

4.4. Limitations

Several limitations of this study should be noted:

1. The TWS-3 employs a limited set of words that may not capture all dimensions of spelling. Responses are scored as correct or incorrect and not analyzed in relation to the type of spelling error. Moreover, only two endophenotypes (PA

and RAN) were included as predictors of spelling. Future studies may examine vocabulary, morphological awareness, orthography, and other cognitive skills associated with spelling such as verbal/phonological memory.

2. The confidence intervals for the estimation of genetic and environmental variance were wide. A larger sample size may provide a more accurate estimate of these parameters. The inclusion of siblings in the No SSD/LI group does not provide an independent comparison group as siblings share approximately 50% of genes. Because siblings limit variability in group comparisons, their inclusion provides a more conservative approach to study of group differences. Although sibling relationships were controlled for in these analyses, we would expect to find even greater differences between individuals with SSD and unrelated controls.
3. We undertook cross-sectional rather than longitudinal analysis for the group difference and regression analyses because many children in the original sample were not assessed in both middle childhood and adolescence. This approach increased power for examining subtypes differences and correlates but precluded examination of individual change across the two ages. A related limitation was the difference between the middle childhood and adolescent samples in rates of SSD. However, many children were assessed at both ages and the age difference in rates of SSD most likely resulted from attrition and the greater numbers of unaffected siblings in the adolescent sample rather than from sample differences in SSD. Yet the SEM analyses were truly longitudinal; subjects included in this analysis had complete data at both time points.
4. Although we controlled for SES, other environmental variables may contribute to spelling skills. Some of the estimated heritability may be due to other common environmental effects. As siblings were employed and there were no identical twin pairs included in this study, we were unable to explicitly estimate the common environmental contribution to variation in spelling.
5. The characteristics of our ascertained sample, including heritability and phenotype associations, may not generalize to an un-ascertained population. Our previous work has shown that the unaffected siblings in our sample tend to obtain somewhat lower scores compared to normative means on many of these measures (Lewis et al., 2011) and adjustment for ascertainment in this SEM model is complex (Morris et al., 2010). However, this ascertained sample is of particular clinical interest given that it is enriched for individuals with SSD.

4.5. Summary and conclusions

This large prospective longitudinal study examined spelling skills of individuals with histories of early childhood SSD in middle childhood and adolescence. The findings demonstrated that children with early histories of SSD and LI are at risk for spelling difficulties that persist into adolescence relative to children without SSD and SSD-only. Children with SSD-only did not differ significantly from children without a history of SSD or LI. A test assessing PA was associated with spelling at both time points whereas a test of

RAN, a measure of lexical access, was related to spelling only in adolescence. As reported in previous studies spelling skills were more heritable in adolescence than in middle childhood, with evidence for larger environmental influences on spelling skills in the younger children.

Several clinical implications may be drawn from these findings. First, children with combined SSD and LI would benefit from monitoring of spelling problems whether or not they have reading difficulties. Many times LI is not diagnosed until after the SSD has begun to resolve, especially if early speech is limited to single words or syllables and is highly unintelligible, as is the case with children with CAS. As CAS has been defined primarily as a motor speech disorder, expressive language difficulties are often overlooked. Speech difficulties may persist into early elementary school, a crucial time for the acquisition of literacy skills. Continued assessment of language and early literacy skills of children with SSD is warranted until language difficulties are identified. Of particular concern is the persistence of these spelling deficits into adolescence, which may impact later-developing academic competencies and possibly occupational outcomes. Finally, evidence for robust environmental influences on spelling support the utility of early intervention for children's spelling difficulties and of approaches that bolster foundational skills such as PA and establishment of orthographic representations.

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Appendix A

Cohen's d for pairwise comparisons among subtypes of children with speech sound disorders on measures at middle childhood and adolescence.*

	Middle childhood group (N = 404)					Adolescent group (N = 261)				
	Measure					Measure				
Pairwise group comparison	TWS-3	PA	RAN	WI	WA	TWS-3	PA	RAN	WI	WA
No SSD/LI–SSD-only	−0.24	0.17	0.01	0.02	−0.26	0.19	0.17	0.21	−0.08	−0.30
No SSD/LI–SSD + LI/no CAS	1.01**	0.67*	−0.45	1.02**	0.88**	1.00**	0.66*	−0.47	1.19**	0.98**
No SSD/LI–CAS	1.83**	1.31**	−0.60*	1.48**	1.66**	0.98**	0.92**	−1.10**	1.18**	1.12**
SSD-only–SSD + LI/no CAS	0.76*	0.88**	−0.46	1.08**	0.67*	1.09**	0.78*	−0.27	1.12**	0.70*
SSD-only–CAS	1.51**	1.49**	−0.62*	1.54**	1.41**	1.13**	1.02**	−0.89**	1.18**	1.00**
SSD + LI/no CAS–CAS	0.62*	−0.54*	−0.05	0.53*	0.56*	0.25	−0.41	0.60*	0.30	0.47

Cohen's d effect sizes reported for each pairwise comparison.

* Indicates medium effect size.

** Indicates large effect size.

TWS-3 = Test of Written Spelling—3rd Ed.; PA = phonological awareness; RAN = rapid automatized naming; WI = word identification; WA = word attack.

As seen in the table, differences between the No SSD/LI group and the SSD-only group did not reach significance at either age. Large effect sizes were observed for all measures between the No SSD/LI group and the CAS group at both ages. Large effect sizes were also observed for the SSD-only group and the CAS group, with the exception of the RAN at middle childhood which demonstrated a medium effect size. Comparisons of the SSD + LI/no CAS and the CAS groups revealed medium effect sizes for TWS-3, PA, WI, WA, at middle childhood and for RAN at adolescence. Comparisons of the No SSD/LI and SSD + LI/no CAS revealed large effect sizes for TWS-3, WI, and WA and medium effect sizes for PA at both ages. Comparisons of the SSD-only and SSD + LI no CAS at middle childhood revealed large effect sizes for PA and WI and medium effect sizes for TWS-3 and WA. At adolescence, large effect sizes were observed for SSD-only and SSD + LI/no CAS on TWS-3 and WI and medium effect sizes for PA and WA.

Middle childhood (Figure 1-A)

Adolescence (Figure 1-B)

Fig. 1.

Comparison of mean spelling scores for clinical groups.

A and B show spelling score means and ranges for the No SSD/LI group and the clinical subgroups of SSD-only, SSD + LI and CAS + LI (Apraxia) at middle childhood and adolescence. The means for No SSD/LI and SSD-only groups are similar at both ages. The SSD + LI and CAS + LI groups are significantly poorer ($p < 0.05$) than the No SSD/LI or SSD-only groups at middle childhood and adolescence. The dashed lines indicate the minimum and maximum scores for each subgroup (range). The box encompasses the area where 50% of the scores fall (interquartile range).

Fig. 2.

Structural equation model of spelling at middle childhood and adolescence.

As illustrated in Fig. 2 the SEM model show that the TWS scores from individuals during middle childhood and scores from their adolescent years have heritable components, random environmental components, and SES factors contributing to them. It also shows the covariance between the heritable components and between random environmental components contributing to the scores at the two time points. Spelling scores show greater heritability at adolescence (0.91) than at middle childhood (0.27). Environmental effects are stronger at middle childhood (0.69) than at adolescence (0.05).

	Middle childhood group (N = 404)				Adolescent group (N = 261)				p-Value
	No SSD/LI (n = 146)	SSD-only (n = 111)	SSD + LI (n = 107)	CAS (n = 40)	No SSD/LI (n = 109)	SSD-only (n = 66)	SSD + LI (n = 63)	CAS (n = 23)	
Participant characteristics									
Age, mean (SD)	9.4 (1.4)	9.2 (1.4)	9.6 (1.4)	9.4 (1.3)	14.4 (1.8)	14.6 (1.7)	14.4 (1.5)	15.1 (1.8)	0.36
Female sex, n (%)	79 (54%)	48 (43%)	34 (32%)	10 (25%)	53 (49%)	22 (33%)	23 (37%)	6 (26%)	0.06
Family characteristics									
Hollingshead SES									
1	4 (3%)	0 (0%)	9 (9%)	2 (5%)	1 (1%)	0 (0%)	8 (13%)	0 (0%)	< 0.001
2	14 (10%)	6 (6%)	10 (10%)	2 (5%)	8 (8%)	4 (6%)	5 (8%)	1 (4%)	
3	19 (13%)	17 (16%)	25 (25%)	12 (32%)	18 (17%)	7 (11%)	9 (15%)	9 (39%)	
4	53 (37%)	43 (40%)	35 (34%)	15 (39%)	33 (31%)	28 (43%)	24 (40%)	7 (30%)	
5	54 (38%)	41 (38%)	23 (23%)	7 (18%)	45 (43%)	26 (40%)	14 (23%)	6 (26%)	
Not reported	2	4	5	2	4	1	3	0	

Table 2

Comparisons among groups of children with speech sound disorders on measures at middle childhood and adolescence.

Measure, mean (SD)	Middle Childhood Group (N = 404)					Adolescent Group (N = 261)					p-Value	Eta ²
	No SSD/LI (n = 146)	SSD-only (n = 111)	SSD + LI/no CAS (n = 107)	CAS (n = 40)	p-Value	Eta ²	No SSD/LI (n = 109)	SSD-only (n = 66)	SSD + LI (n = 63)	CAS (n = 23)		
TWS-3, mean (SD)	100.6 (15.1)	96.9 (15.8)	84.8 (16.2)	76.3 (12.7)	< 0.001 ^{a,b,c,d,e}	0.22	107.5 (11.8)	109.8 (12.6)	93.5 (17.1)	88.6 (20.6)	< 0.001 ^{a,b,c,d}	0.23
Elision (PA), mean (SD)	0.50 (0.70) n = 116	0.43 (0.88) n = 95	-0.52 (1.14) n = 89	-0.92 (1.00) n = 32	< 0.001 ^{a,b,c,d}	0.27	0.13 (0.56) n = 57	0.23 (0.53) n = 53	-0.44 (1.04) n = 47	-0.86 (1.19) n = 19	< 0.001 ^{a,b,c,d}	0.16
RAN, mean (SD)	-0.18 (1.18) n = 118	-0.17 (0.94) n = 99	0.53 (2.03) n = 87	0.62 (1.37) n = 34	< 0.001 ^{a,b,c,d}	0.06	-0.28 (0.95) n = 63	-0.09 (0.93) n = 56	0.16 (0.92) n = 49	0.70 (0.88) n = 20	< 0.001 ^{b,d}	0.09
Word ID, mean (SD)	106 (13.9) n = 145	106 (12.6) n = 110	90.8 (15.7) n = 106	82.2 (16.5)	< 0.001 ^{b,c,d,e}	0.25	105 (11.1)	104 (9.7)	89 (16.3) n = 61	83 (19.1)	< 0.001 ^{b,c,d}	0.29
Word AT, mean (SD)	105 (14.2) n = 145	104 (14.0) n = 107	88 (19.1) n = 106	80 (19.3) n = 39	< 0.001 ^{b,c,d}	0.23	105 (11.2)	101 (8.8)	93 (15.8) n = 61	85 (16.6)	< 0.001 ^{b,c,d}	0.23

^aNo SSD/LI/CAS differs from SSD + LI.

^bNo SSD/LI/CAS differs from CAS.

^cSSD only differs from SSD + LI.

^dSSD only differs from CAS.

^eSSD + LI differs from CAS.

Table 3

Predictors of total spelling scores at middle childhood and adolescence.

	Unadjusted for SSD and LI			Adjusted for SSD and LI		
	Beta	Std error	p-Value	Beta	Std. error	p-Value
Middle childhood						
SES	0.51	0.62	0.406	0.58	0.63	0.353
Elision (PA)	2.73	0.78	<0.001	2.34	0.78	0.003
RAN	-0.36	0.46	0.431	-0.01	0.45	0.977
Word identification	0.67	0.05	<0.001	0.67	0.05	<0.001
Adolescence						
SES	0.24	0.69	0.725	0.2	0.68	0.765
Elision (PA)	3	1.11	0.008	2.82	1.1	0.011
RAN	-1.71	0.72	0.019	-1.78	0.72	0.015
Word identification	0.82	0.06	<0.001	0.81	0.07	<0.001

Table 4

Demographic characteristics and group differences of study participants included in the SEM model.^a

	No SSD/LI (<i>n</i> = 44)	SSD-only (<i>n</i> = 39)	SSD + LI (<i>n</i> = 35)	CAS (<i>n</i> = 13)	<i>p</i> -value
Participant characteristics					
Middle childhood age, mean (SD)	9.2 (1.1)	9.1 (1.5)	9.2 (1.3)	9.5 (1.4)	0.87
Adolescent age, mean (SD)	15.0 (1.7)	14.8 (1.6)	14.7 (1.5)	16.1 (1.5)	0.07
Female sex, <i>n</i> (%)	24 (55%)	16 (41%)	13 (37%)	2 (15%)	0.07
Reading disorder, <i>n</i> (%)	10 (23%)	7 (18%)	27 (77%)	9 (69%)	<0.001
Family characteristics					
Hollingshead SES					
1	0 (0%)	0 (0%)	3 (9%)	0 (0%)	0.11
2	2 (5%)	3 (8%)	2 (6%)	0 (0%)	
3	4 (9%)	4 (11%)	4 (12%)	4 (31%)	
4	16 (37%)	16 (43%)	16 (48%)	6 (46%)	
5	21 (49%)	14 (38%)	8 (24%)	3 (23%)	
Not reported	1	2	2	0	

^aNote that there were 133 children assessed at both middle childhood and adolescent ages. There were two pairs of twins among the 133 and no half-siblings. We randomly removed one of from each twin pair for our SEM analysis, resulting in *n* = 131 used in the SEM analysis.