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Early Speech-Language Impairment and Risk for Written Language Disorder: A Population-Based Study

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Abstract

OBJECTIVE—Compare risk of written-language disorder (WLD) in children with and without speech-language impairment (S/LI) from a population-based cohort.

METHODS—Subjects included all children born 1976–1982 in Rochester, Minnesota, who remained in the community after age 5 years ($n = 5718$). Records from public and private schools, medical agencies, and tutoring services were abstracted. S/LI was determined based on eligibility criteria for an individualized education plan. Incident cases of WLD were identified by research criteria using regression-based discrepancy, non-regression-based discrepancy, and low achievement formulas applied to cognitive and academic achievement tests. Incidence of WLD (*with* or *without* Reading Disorder [RD]) was compared between children with and without S/LI. Associations were summarized using hazard ratios.

RESULTS—Cumulative incidence of WLD by age 19 years was significantly higher in children with S/LI than without S/LI. The magnitude of association between S/LI and WLD *with* RD was significantly higher for girls than boys. This was not true for the association between S/LI and WLD *without* RD.

CONCLUSION—Risk for WLD is significantly increased among children with S/LI compared to children without S/LI based on this population-based cohort. Early identification and intervention for children at risk for WLD could potentially influence academic outcomes.

Speech-language problems are common among children. The prevalence of speech sound impairment in children ranges from 1.3% to 12.6% depending on age and criteria used, and estimates of prevalence of specific language impairment (SLI) range from 2% to 19%.[1] Written language disorder (WLD) and reading disorder (RD) are subtypes of language-based learning disability (LLD) that are also common in childhood. In two population-based studies, the cumulative incidence of RD ranged from 5.3% to 11.8% and the cumulative incidence for WLD ranged from 6.9% to 14.7% depending on criteria used. [2,3]

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The relationship between speech-language impairment (S/LI) and RD has been well established, with long term implications beyond difficulty learning to read.[4–7] Tomblin, et al. not only found a strong correlation between spoken language skills and reading, but their data suggested an association that leads from oral language difficulties to academic difficulties, which then play a role in the development of behavioral outcomes in some children. In a systematic review of the literature, McCormack et al. found that early history of speech impairment (which included some children with both speech and language impairment) was associated with a broad range of later difficulties including academic skills in reading, writing, calculating, focused attention and thinking, and social outcomes including social and family relationships and job-related skills.

The relationship between S/LI and WLD has been less well studied, but there is a growing body of research exploring the relationship between speech-language skills and written language impairment. [8–11]

Puranik & Lonigan found that as early as preschool, children with oral language impairment were significantly delayed relative to peers with typical development in their early writing-related skills. Several researchers found that children with S/LI produced written narratives that were less complex than either age-matched or language-matched peers.[8–9] Additional studies suggest that speech problems and/or oral language impairment can have an adverse impact on later development of written language skills and an association with more general academic and social struggles.[12–14] This study adds to the current base of knowledge by examining co-occurrence of S/LI and WLD in an unbiased, population-based, non-referred sample.

There is evidence of familial transmission of S/LI and RD, and it seems likely that genetic factors influence vulnerability for WLD as well. However, distinct loci have yet to be identified for specific disorders, in part because these difficulties are understood to be multifactorial in nature.[15–16]

In this paper, we will use the designation "S/LI" to refer to children presenting with speech and/or language impairment, that is, speech impairment, specific language impairment, or both together. Because S/LI may be one early indicator of WLD, understanding the relationship between these disorders can help to determine to what degree S/LI should be considered a risk factor in children who are not yet reading and writing. Early identification of children with S/LI and early intervention for children at risk for WLD could potentially moderate the severity of these problems.

The availability of records from a population-based birth cohort with research-identified WLD[2] provided an opportunity to study the co-occurrence of S/LI and WLD. The primary purposes of this study were to: (1) compare risk for WLD among children with S/LI and children without S/LI; and (2) compare the risk for WLD associated with S/LI between boys and girls. Finally, the association between WLD and SLI was examined separately for children who had WLD *without* RD versus WLD *with* RD.

METHODS

Study Setting and Resources

Characteristics unique to Rochester, Minnesota provide the opportunity for population-based epidemiologic research on WLD and S/LI. Rochester is a relatively isolated center in southeastern Minnesota, 90 miles southeast of the nearest major urban center of Minneapolis-St Paul. Virtually all medical care in the area is provided by Mayo Clinic, Olmsted Medical Center, and 3 affiliated hospitals. A medical record-linkage system of

Rochester and Olmsted County residents is a part of the Rochester Epidemiology Project, which provides indexing of all diagnoses and surgical procedures for automated retrieval. The medical records include details about medical, mental health, social services, and home visits, as well as psychiatry and psychology reports and test results.

Additionally, there is a research agreement with the Minnesota Independent School District (ISD) #535, which serves the city of Rochester, for permission to access their well-documented school records for all birth cohort members registered at any of the 41 public, private, or parochial schools, including individuals who graduated, who were home-schooled, who moved from the district, or were deceased. This local school district has a long history of excellent management of children with special needs, including S/LI and WLD. The cumulative school record includes: all school assessments and reassessments (including any special education testing); dates and details of individualized education plans (IEPs); all individually administered academic, achievement and cognitive ability test results; and observations of any type of learning, behavior, or performance issues made by teachers, parents, special education teachers, school psychologists, school social workers, counselors, or physicians. Under an additional research agreement, permission was obtained to view the records of a privately owned tutoring center that was in existence in the community during the school years relevant to the birth cohort members. The study was approved by the Institutional Review Boards of Mayo Clinic and Olmsted Medical Center.

Birth Cohort

The birth cohort consisted of all children born between January 1, 1976 and December 31, 1982 to mothers residing in the townships comprising Minnesota ISD #535 ($n = 8458$). The target population consisted of 5718 children (2956 boys and 2762 girls) who still lived in Rochester at or after age 5 years and who were followed retrospectively from birth until the occurrence of death, emigration, or graduation. The steps and resources used for identification and follow-up of this birth cohort, and analysis of potential influence of migration bias, have been reported previously.[17]

S/LI, WLD, and RD, Incident Cases-Identification and Case Definition

Identification of RD and WLD cases involved several steps and multiple sources of information that included individual test results, evidence of educational intervention, and documentation of parent and teacher observations. Details of these steps and sources were previously described.[2,3] Briefly, several steps were used to identify the number of possible S/LI, WLD, and RD incidence cases, starting with the cumulative school records of each child in the birth cohort ($n = 5718$). School records were searched for any indication of concerns about learning and behavior (e.g., referral forms, reports of periodic IEP review, IEP assessment/reassessment report forms, medical reports, private evaluation reports, individually administered tests, any notations made by teachers, etc.). Based on these documented concerns by school psychologists, physicians, social workers, school nurses teachers, and parents, 1961 children were designated as *children with learning/behavioral concerns*. Detailed data for these 1961 children were abstracted from school and medical records, and records from a local private tutoring agency. Abstracted data from all of the above sources included: all individually administered academic achievement and cognitive ability test results, information related to behavioral problems, and speech-language test results. Standardized tests were consistent with those commonly in use by schools and other providers during that time frame, such as the Woodcock-Johnson, Stanford-Binet, etc. Nineteen children clinically diagnosed with severe intellectual disability or with a full scale IQ score <50 were excluded from the study. Two children with ataxia and 3 children with special education services for hearing loss were also excluded.

Identification of *S/LI incidence cases* involved identifying children with an IEP designating speech-language as the primary impairment, excluding children under this category who had goals only for voice or dysfluency. Children for whom speech-language was the primary impairment on an IEP in Minnesota were required to meet stringent criteria, that is, scores on standardized tests ≥2 SD below the mean for age. The standardized tests varied according to age of child and primary area(s) of concern, but included typical measures used in clinical practice to identify children with speech or language impairment, such as the Peabody Picture Vocabulary Test, Goldman-Fristoe Test of Articulation, and Test of Language Development.

Identification of *WLD incidence cases,* as described in previously published reports, consisted of applying 3 psychometric criteria. Specifically, for each child designated with learning/behavioral concerns, all writing achievement and IQ test scores were used to form pairs of cognitive ability and writing performance measures within a calendar year. In each of the following formulas, x represents the study subject's IQ score, and y represents the standard score from the writing achievement test. Children who had standard scores in writing achievement >1.75 SD below their predicted standard score on an individually administered measure of cognitive ability (IQ) [3] were identified as having WLD by the Regression Formula-Minnesota ($y < 17.40 + 0.62\ x$). Children who had differences between age-based standard scores on individually administered intelligence measures and writing achievement that varied by grade were identified through the Discrepancy Formula. That is, $x - y \geq 15$, 19, or 23 points, for kindergarten–3rd, 4th–6th, and 7th–12th grade, respectively. This formula was used in ISD #535 before 1989, when members of our birth cohort attended school. The final method was identify children who had a discrepancy between performance and IQ were identified by the Low-Achievement Formula: $x \geq 80$ and $y \leq 90$, an alternative method that has been used to identify learning disabilities.[18–20] The earliest date among these pairs of scores for which the scores met at least 1 of these psychometric criteria, was designated the date of WLD research diagnosis. *Incident cases of RD* were identified by applying the same 3 psychometric criteria used for identification of WLD cases.

Non-S/LI members of the birth cohort still living in Rochester at ≥5 years of age, not identified as S/LI and without severe intellectual disability, ataxia, or hearing loss, were included as controls.

Statistical Analysis

Analyses were performed separately for the 3 events of interest: any WLD, WLD *with* RD, and WLD *without* RD. The cumulative incidence of WLD was calculated according to the Kaplan and Meier method.[21] Because WLD *with* RD and WLD *without* RD are competing risks (ie, an event whose occurrence either precludes the occurrence of the other even or alters the probability of occurrence of this other event), the cumulative incidence of WLD *with* RD and WLD *without* RD, respectively, were calculated by taking into account this competing risk.[22]

The Cox proportional hazards model was applied to obtain hazard ratios (HR) and corresponding 95% confidence intervals (CIs) separately for each of the 3 events (WLD, WLD *with* RD, and WLD *without* RD). In each model, the incidence of the event was regarded as the outcome variable whereas S/LI case status (incident S/LI cases versus non-cases) was regarded as an explanatory variable. Unadjusted and adjusted HRs were calculated. In the latter case, the child's sex, child's race (white vs non-white), child's birth weight, maternal years of education, and maternal age at birth of the child were included in the model. [22] Given the overall sample size, the prevalence of S/LI, and the number of events for the 2 outcomes of interest, the study has 80% power to detect an HR of 1.7 for the overall association between S/LI and WLD *with* RD and an HR of 2.4 for the association between S/LI and WLD *without* RD. *P*-values (2-sided) <0.05 were considered statistically significant. Analyses were performed using the SAS® version 9.2 software package (SAS® Institute, Inc.; Cary, NC).

RESULTS

Among the 5694 remaining subjects, 294 were identified as having S/LI based on IEPs indicating speech-language as the primary impairment. The cumulative incidence of S/LI overall at age 19 years was 5.7% (95% CI: 5.0–6.3); for girls, 3.9% (95% CI: 3.2–4.7) and for boys, 7.3% (95% CI: 6.3–8.3). Of the 294 children with S/LI, 163 (55%) were identified as also having WLD based on meeting at least one of the three psychometric criteria for WLD.[2] Of these 163 children, 139 children had S/LI and WLD *with* RD (47%), and 24 children had S/LI and WLD *without* RD (8%). The co-occurrence of WLD among children with S/LI is graphically represented in Figure 1, depicting percentages of children having WLD *with* RD and WLD *without* RD among children with S/LI.

Table 1 shows that for both genders, the cumulative incidence of WLD by age 19 years is significantly greater among children with S/LI than among children without S/LI (boys: 61.4% vs 18.5%; girls: 55.1% vs 9.4%; $P < .01$). In addition, the risk of WLD associated with S/LI is greater among girls than among boys (adjusted HR: 7.60 vs 3.89; $P < .001$).

The risk for WLD *with* RD among boys and girls with and without S/LI is shown in Table 2. Children with S/LI had significantly increased risk of WLD *with* RD, compared to children without S/LI. There was a significant gender × S/LI interaction in the incidence of WLD *with* RD, with greater risk for girls than for boys (adjusted HR: 9.48 for girls and 4.36 for boys; $P < .001$ in adjusted model).

Table 3 shows that there was also a significant increase in the risk of WLD *without* RD for both boys and girls with S/LI compared to children without S/LI (adjusted HR: 3.81 for girls and 2.30 for boys). However, the risk of WLD *without* RD associated with S/LI was not significantly different between boys and girls (gender × S/LI interaction $P = .25$ for adjusted model).

DISCUSSION

This research is unique in using a population-based birth cohort to examine the comorbidity of S/LI and WLD based on carefully defined research criteria. Our study of S/LI and WLD in a non-referred sample of boys and girls contributes significantly to understanding the natural history of the comorbidity between S/LI and WLD. Our major finding is a strong association between S/LI and WLD. Among children with S/LI, the cumulative incidence of WLD by age 19 years was 61.4% for boys and 55.1% for girls. In contrast, among children without S/LI, the cumulative incidence of WLD by age 19 years was 18.5% for boys and 9.4% for girls. This strong relationship between S/LI and WLD is consistent with results across a number of studies[8–9,23–25] reporting that the written narratives produced by children with S/LI contained patterns of errors in grammar and syntax different from the chronologic-age matched and language-age matched comparison groups. Not only were children with S/LI more likely to have difficulty with written language, but their errors reflected what may be fundamental differences in the development of spoken and written language in children with S/LI compared to peers without S/LI. Mackie and Dockrell[23] noted that there was *not* a direct transformation of errors from the spoken modality to written expression. In addition, factors such as a) adverse effects of a limited vocabulary contributing to reduced complexity of both spoken and written language, b) different allocation of cognitive resources needed for translating thoughts to written words, and c) environmental influences in the form of both direct and indirect instruction related to written language, along with other influences, may contribute to the association between S/LI and WLD.[23] Our data also cannot be interpreted to suggest a developmental progression from impairment in speech and language skills to impairment in written language but rather that there is a strong association among skills in both domains.

Prior studies have established a strong association between S/LI and RD.[4–6] Speaking, reading, and writing can be seen as highly overlapping, but distinct areas of linguistic capability.

The strength of the association between S/LI and WLD has not been previously explored in a population-based birth cohort. Our study extends knowledge about the relationships among aspects of spoken and written language by demonstrating an association between S/LI and WLD *with* RD as well as an association between S/LI and WLD *without* RD. Researchers have suggested that one underlying factor in S/LI and RD is an impairment in phonological representations.[11] There is a need to consider whether there may be a different underlying association between language and WLD if it is possible to have S/LI and WLD *without* RD. Puranik and Lonigan found that nonverbal cognitive abilities appeared to influence early writing abilities of children, but that the relationship was affected by level of language skill.[11] In addition, there may be genetic factors that we do not yet understand that influence children's vulnerability to these problems.[15]

We found both boys and girls with S/LI in this cohort were more at risk for WLD than children with no S/LI. The rate of occurrence of S/LI, WLD, and RD has previously been shown to be higher in boys than in girls.[26] Interestingly, although boys were at greater risk than girls for RD and WLD[14,16], the risk of WLD associated with S/LI was higher among girls than boys (HR: 7.60 vs 3.89). We suggest the possibility that girls with S/LI may be inherently more severely affected, from a neurodevelopmental perspective, and therefore more likely to also have other problems like WLD.

There are three limitations to this study to consider. First, this was a retrospective study, with the possibility of under-ascertainment of S/LI or WLD incident cases. Our S/LI cases were identified solely by the presence of an IEP with speech or language as the primary service. Many more children were served under other primary disabilities (eg, Emotional/Behavior Disorder, Learning Disability) while receiving assistance for a language-related impairment such as vocabulary development. Additionally, children served under an IEP for speech-language services were eligible based on scores that were lower than −2 SD for age, while a large-scale study[6] has suggested that children performing at −1.25 SD should be considered language impaired. Therefore, our identification of children with S/LI may under-represent children in the cohort with less severe S/LI. Our use of IEP data potentially restricted our sample size. This precluded separation into types of S/LI (e.g., "speech-only", "language-only" and "speech and language") for analysis.

Second, emigration from the entire birth cohort of 8548 children is a potential limitation. Detailed comparison of children who left the community before age 5 years and those who stayed, however, indicates that the 5718 children included in the study are representative of the entire birth cohort.[17] Third, during the time in which these children attended school in Rochester, MN, it was primarily a white, middle-class community, which may limit generalization of these findings to other populations. Nevertheless, our data provide important and unique information about the relationship between S/LI and WLD for comparison with other epidemiologic studies.

CONCLUSIONS

This is the first epidemiologic study using a population-based birth cohort to examine the association between S/LI and WLD. Our data clearly show a strong association between early impairment in spoken language and later WLD. There is a significantly increased risk of WLD among children with S/LI, and the relationship is even stronger when RD is present. There is also a significant association of S/LI and WLD *without* RD. Consistent with previous studies indicating that boys are at higher risk for learning problems,[26] boys had a higher cumulative incidence of WLD than girls. However, hazard ratios showed that the risk of WLD associated with SLI appears to be higher for girls, which is a new finding.

Another interesting finding in our data was that a few children in our study were identified with WLD *prior* to identification with S/LI. Minnesota's relatively restrictive criteria of performance greater than 2 standard deviations below average for defining eligibility for services could be a factor. There was not sufficient information in most of these records to draw conclusions, however, we speculate that some or all of these children had earlier speech-language issues that were not severe enough to warrant formal speech-language testing or services at a younger age, and later, identification occurred when the discrepancy between performance and age expectations increased. Further study of the value of early intervention for children with less severe impairment is certainly needed. Whether proactive intervention for any early S/LI can diminish risk for emergence of later WLD during primary grades clearly requires further investigation. Given the strong association between S/LI and WLD, children who present with early S/LI should be closely monitored for reading and written language problems so that appropriate educational services can be provided before problems become severe. At minimum, our findings suggest a need to monitor emerging writing skills analogously to current means of monitoring reading skills, which are being assessed and taught as early as the preschool years. It does not seem unreasonable to suggest that encouraging early writing skills might facilitate written language development, particularly for children with weak oral language skills. Finally, additional long-term follow-up is needed for evaluating adult outcomes of children identified with S/LI in their early years.

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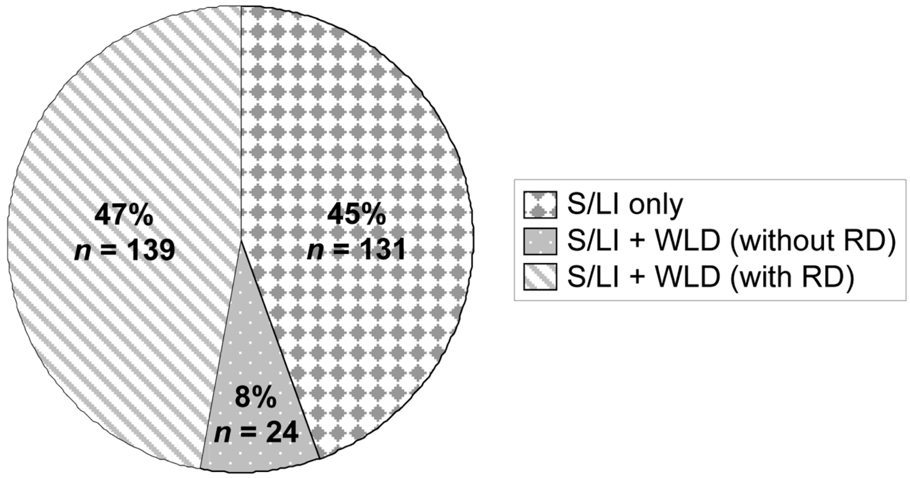

FIGURE 1.
Distribution of WLD (with and without RD) among 294 individuals with S/LI.

TABLE 1

Risk of WLD[a] Associated with S/LI According to Gender

	n	Number of WLD Cases by Age 19 Years	Cumulative Incidence of WLD by Age 19 Years, % (95% CI)	Unadjusted HR[b] (95% CI), *P*	Adjusted HR[c] (95% CI), *P*
Both genders					
S/LI	294	163	59.2% (52.9–64.7)	5.76 (4.85–6.85), <.01	4.71 (3.92–5.66), <.01
Non-S/LI	5400	639	14.0% (13.0–15.0)	1.00	1.00
Boys					
S/LI	196	112	61.4% (54.4–67.9)	4.40 (3.57–5.43), <.01	3.89 (3.12–4.86), <.01
Non-S/LI	2751	429	18.5% (16.9–20.0)	1.00	1.00
Girls					
S/LI	98	51	55.1% (43.5–64.3)	8.22 (6.04–11.18), <.01	7.60 (5.51–10.47), <.01
Non-S/LI	2649	210	9.4% (8.2–10.6)	1.00	1.00

[a]Written Learning Disability assessed by any of the 3 formulas (Regression Formula Minnesota, Discrepancy Formula, Low Achievement Formula).

[b]$P = .001$ for gender*S/LI interaction.

[c]Models for boys and girls separately have been adjusted for child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. Model with both genders combined has been adjusted for sex, child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. $P < .001$ for gender*S/LI interaction.

TABLE 2

Risk of WLD (*with* RD)[a] Associated with S/LI According to Gender

	n	Number of WLD (*with* RD) Cases by Age 19 Years	Cumulative Incidence of WLD (with RD) by Age 19 Years, %	Unadjusted HR[b] (95% CI), *P*	Adjusted HR[c] (95% CI), *P*
Both genders					
S/LI	294	139	50.1%	6.55 (5.41–7.92), <.01	5.36 (4.38–6.57), <.01
Non-S/LI	5400	460	9.9%	1.00	1.00
Boys					
S/LI	196	96	52.1%	4.85 (3.86–6.09), <.01	4.36 (3.42–5.56), <.01
Non-S/LI	2751	321	13.6%	1.00	1.00
Girls					
S/LI	98	43	46.1%	10.08 (7.15–14.22), <.01	9.48 (6.61–13.59), <.01
Non-S/LI	2649	139	6.1%	1.00	1.00

[a]Written Learning Disability and Reading Disability were assessed by any of the 3 formulas (Regression Formula Minnesota, Discrepancy Formula, Low Achievement Formula).

[b]$P < .001$ for gender*S/LI interaction.

[c]Models for boys and girls separately have been adjusted for child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. Model with both genders combined has been adjusted for sex, child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. $P < .001$ for gender*S/LI interaction.

TABLE 3

Risk of WLD (*without* RD)[a] Associated with S/LI According to Gender

	n	Number of WLD (*without* RD) Cases by Age 19 Years	Cumulative Incidence of WLD (without RD) by Age 19 Years, %	Unadjusted HR[b] (95% CI), *P*	Adjusted HR[c] (95% CI), *P*
Both genders					
S/LI	294	24	9.1%	3.44 (2.24–5.27), <.01	2.74 (1.74–4.32), <.01
Non-S/LI	5400	179	4.1%	1.00	1.00
Boys					
S/LI	196	16	9.2%	2.88 (1.70–4.87), <.01	2.30 (1.30–4.08), <.01
Non-S/LI	2751	108	4.8%	1.00	1.00
Girls					
S/LI	98	8	8.9%	4.17 (2.00–8.68), <.01	3.81 (1.81–8.02), <.01
Non-S/LI	2649	71	3.3%	1.00	1.00

[a]Written Learning Disability and Reading Disability assessed by any of the 3 formulas (Regression Formula Minnesota, Discrepancy Formula, Low Achievement Formula).

[b]$P = .385$ for gender*S/LI interaction.

[c]Models for boys and girls separately have been adjusted for child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. Model with both genders combined has been adjusted for sex, child's race (white vs non-white), child's birth weight, mother's education level, and mother's age. $P = .253$ for gender*S/LI interaction.