Critical Review:
Are children with an early history of isolated speech sound disorders, in the absence of a comorbid language impairment, at risk for poor literacy outcomes?

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This critical review examined if children with an early history of isolated speech sound disorders, in the absence of a language impairment, are at risk for poor literacy outcomes. A literature search yielded nine articles (six mixed design studies, two between groups designs, and one single group design) that met inclusion criteria. Overall, the studies provided suggestive evidence that the risk of poor literacy outcomes in children with isolated speech sound disorders is relatively low, unless there are co-occurring language difficulties. Some studies suggested that children with isolated speech sound disorders are most at risk for phonological awareness deficits and spelling difficulties. Additionally, disordered speech errors and speech difficulties that persist to the age of school entry may increase risk of poor outcomes. Further research is necessary to provide more definitive conclusions. The clinical significance and limitations of the current research are discussed.

Introduction

Children with Speech Sound Disorders have difficulty producing developmentally appropriate speech sounds, leading to reduced intelligibility. The term 'speech sound disorder' (SSD) is a heading that encompasses many sub-categories including articulation disorder, phonological disorder, and childhood apraxia of speech (Bowen, 2011). A large sub-category of children with SSD also have co-existing language impairments (Shriberg, Tomlin, & McSweeny, 1999; Tyler, Lewis, Haskill, & Tolbert, 2003; Shriberg & Kwiatkowski, 1994). In fact, these two disorders have been predicted to co-occur at a rate of 25-30% (Gallagher, Frith, & Snowling, 2000; Lewis, 1996; Pennington & Lefly, 2001).

Research has shown that children with SSD are at an increased risk for poor literacy outcomes (Bird, Bishop, & Freeman, 1995; Raitano et al., 2004). Many factors including speech error patterns, socioeconomic status (SES), nonverbal intelligence (NIQ), the age at which speech errors persist to, and the presence or absence of a language impairment (LI) have been found to influence these outcomes (Bishop & Adams, 1990; Catts, 1993; Hesketh, 2004). There is evidence to suggest that children with SSD and comorbid LI consistently demonstrate poor literacy outcomes. However, when evaluating the literacy development of children with isolated SSD, in the absence of LI, the literature is mixed. (Bishop, Price, Dale, & Robert, 2003; Hesketh, 2004).

Evaluating current evidence to identify which factors place children with SSD at risk for poor literacy outcomes is needed to inform clinical practice. In order for clinicians to deliver appropriate treatment, it is important for them to understand which children with SSD are predisposed to poor literacy outcomes. In being able to identify children at risk early, these individuals can receive the support they need to achieve positive literacy outcomes as soon as possible.

Objectives

The primary objective of this paper was to critically review the existing literature examining if children with an early history of isolated SSD, in the absence of LI, are at risk for poor literacy outcomes.

Methods

Search Strategy

Online databases including PubMed, PsychInfo, and Google Scholar were searched using the following terms: [(speech sound disorder) OR (phonolog* disorder) OR (articulation disorder) AND (language impairment) OR (language disorder) AND (literacy) OR (literacy outcome)]. Reference lists of relevant articles were also searched for articles.

Selection Criteria

Papers were selected if they assessed the early literacy or literacy outcomes of children with SSD, in the presence and absence of comorbid LI. Articles were also included if participants presented with SSD and age appropriate language skills. Lastly, participants had to be diagnosed with SSD in the absence of co-occurring sensory, neurological, physical, or intellectual disabilities.

Data Collection

Results of the literature search yielded nine articles fitting the selection criteria explained above. These articles consisted of six mixed design studies, two between groups designs, and one single group design.

Results

Between Groups Designs

Between groups designs are conducted to compare the outcomes of two or more groups simultaneously. Large sample sizes and the ability to control for environmental factors and confounding variables can reinforce the strength of these studies.

Sices, Taylor, Freebairn, Hansen and Lewis (2007) used a between groups design to examine the early reading and writing outcomes of 125 preschool children (ages 3-6 years) with moderate to severe SSD. Individual associations between SSD severity and comorbid LI on literacy outcomes were examined.

Participants were well-described using specific inclusion and exclusion criteria. Appropriate methods of evaluation were used to classify participants into two comparison groups: isolated SSD and SSD with comorbid LI. Performance was compared to 68 typically developing (TD) siblings. Comparison groups were similar at baseline; however, the isolated SSD group had significantly higher performance IQ (PIQ) than the SSD and LI group. The TD siblings were claimed to be 'comparable in age', although, no data was provided to confirm this. Literacy outcomes were assessed using valid and reliable standardized tests of pre-academic reading and writing readiness. Performance on various elements of speech and language measures were also examined, but these results will not be discussed for the purposes of this review. Methods were described in adequate detail for replication.

Detailed and appropriate statistical analyses revealed that participants with comorbid LI had significantly lower scores on measures of early reading and writing compared to those with SSD only. Only language skills were related to early literacy outcomes. SSD severity and articulation skills were not associated with early literacy outcomes after comorbid LI was accounted for in the final regression model.

Strengths of this study include the high reliability and validity of both literacy outcome measures. It was limited by a somewhat lenient alpha criteria in statistical comparisons and minimal information was provided regarding TD sibling scores. Differences in PIQ were not statistically controlled for, despite baseline differences. The use of siblings as a control group and a sample that was mostly middle to high SES may limit generalization.

Overall, this study provides suggestive evidence that language skills, not speech skills, are predictive of early reading and writing abilities.

Raitano, Pennington, Tunick, Boada and Shriberg (2004) used a between groups design to compare the pre-literacy performance of 101 children with histories of SSD to 41 TD controls (ages 5-6 years). Specifically, researchers sought to examine if SSD persistence and comorbid LI were independently related to pre-literacy outcomes.

Participants were well described and inclusion and exclusion criteria were mostly appropriate. However, a lenient 30th percentile cut-off was used to determine the presence or absence of SSD. The SSD group had significantly lower NIQ and SES than the TD control group at baseline. Gold standard assessments were used to classify participants into 4 subgroups for comparison according to persistent or resolved SSD and the presence or absence of LI. Appropriate standardized and non-standardized assessments were used to assess pre-literacy outcomes which included measures of phonological awareness (PA), letter knowledge, and rapid serial naming (RSN). Methods and procedures were described in great detail, making replication possible.

Detailed and appropriate statistical analysis revealed that both SSD persistence and comorbid LI were independently associated with poorer outcomes on PA tasks, even when NIQ was statistically controlled for. Comorbid LI was significantly associated with lower letter knowledge scores. No main effects of SSD persistence were found on letter knowledge or RSN after controlling for NIQ. Even children with normalized SSD and no LI had significantly poorer PA outcomes than the TD control group, but did not differ on RSN or letter knowledge tasks.

A significant limitation of this study was the use of a lenient 30th percentile cut-off to determine the presence or absence of SSD, which may have falsely classified children as having SSD. Although NIQ was statistically controlled for, adjustments were not made for differences in SES.

Overall, this study provides suggestive evidence that a history of SSD is independently associated with deficits in some pre-literacy skills, specifically PA.

Mixed Designs

Mixed design studies are conducted to compare the results of two or more groups on repeated measures. These studies provide a similar level of evidence as between groups designs; however, an added limitation is the potential for participant withdrawal over time.

Peterson, Pennington and Shriberg (2009) extended the findings of Raitano et al (2004) (Time 1) and completed a mixed design follow-up study to examine the literacy outcomes of these children when they were 7-9 years of age (Time 2). They also investigated whether language skills were a more powerful predictor of literacy outcomes than SSD persistence.

Participants included 86 children with early histories of SSD and 37 TD controls. Inclusion and exclusion criteria mimicked that of Raitano et al (2004). Appropriate statistical analyses were conducted to ensure that participants who completed measures at Time 2 were generally representative of the sample who completed the study at Time 1. The final SSD and control groups were similar in age, gender, and ethnicity, but differed on NIQ and SES status. Literacy outcomes were assessed using gold-standard measures of single-word reading accuracy, single-word spelling accuracy, reading comprehension, PA skills, and reading fluency. Methods were outlined clearly and with adequate detail for replication.

Appropriate statistical analyses revealed that as a whole, the SSD group performed more poorly than the TD controls on literacy outcome measures. Both LI and persistent SSD were associated with poor PA outcomes, but only language skills predicted literacy outcomes. SSD persistence was not uniquely associated with literacy outcomes once comorbid LI was accounted for.

This study was limited by its use of a lenient 30th percentile cut-off for determining SSD and a control group that was noted to perform somewhat above national norms on literacy outcome measures.

Overall, this study provides suggestive evidence that SSD persistence is associated with PA deficits, but not poor literacy outcomes.

Hayiou-Thomas, Carroll, Leavett, Hulme and Snowling (2017) implemented a mixed design study to examine the influence of SSD on literacy outcomes while considering additional risk factors including co-morbid LI, family risk of dyslexia (FR), and SSD profile (i.e. error type, SSD persistence, SSD severity). For the purposes of this review, results related to family risk of dyslexia will not be discussed.

Participants were recruited from an overarching longitudinal study (Nash, Hulme, Gooch, & Snowling, 2013) and evaluated over three time points (T1 = age 3 ½, T3 = age 5 ½, and T5 = age 8). Children were initially assessed and identified with SSD at T1 (n = 68) and further classified into 4 subgroups for comparison: SSD only, SSD and LI, SSD and FR, and SSD, LI and FR. Results were compared to TD controls (n = 68). Adequate and well-specified criteria were used to categorize participants into the four SSD subgroups. Although the SSD and control groups did not differ in age, the control group had significantly higher SES and PIQ. Literacy outcomes were assessed with credible measures of phoneme awareness, word-level reading, and spelling at T3 and again for word-level reading, reading comprehension, and spelling at T5. The majority of methods and procedures were described in detail, however, specific information regarding the location, time-length, and order of testing was not provided.

Appropriate statistical analyses revealed no significant differences in literacy outcomes between each of the four SSD subgroups. However, language was found to be a stronger predictor of literacy outcomes than SSD alone. At T3, isolated SSD predicted a small but significant risk for difficulties in phoneme awareness and spelling, but these difficulties appeared to be mostly temporary and did not persist to T5. At T5, isolated SSD was associated with poor word reading only, and the unique variance was minimal. The predictive relationship between SSD and literacy did not change when differences in PIQ were statistically controlled for. Persistent SSD to the point of school entry and disordered speech errors were associated with poorer outcomes at T3. SSD severity did not predict literacy outcomes.

This study was limited by the small number of participants in each subgroup, which reduced statistical power to detect significant differences. Additionally, SES was not employed as a covariate in statistical analysis and may serve as a confounding variable.

Overall, this paper provides highly suggestive evidence that children with isolated SSD exhibit minor early literacy difficulties, that are mostly temporary. Further, it indicates that children with persistent SSD and disordered speech errors may be at an increased risk for poor outcomes.

Lewis et al. (2015) used a mixed design study to examine the literacy outcomes of adolescents (ages 11-18 years) with early histories of SSD with and without co-morbid LI. The influence of additional factors including SES and NIQ were also examined, but will not be discussed for the purposes of this paper.

Participants included 170 children with moderate to severe SSD who were recruited in early childhood (ages 4-6) using well-defined and specific criteria. Gold standard measures for assessing articulation and language skills were administered to classify participants into three comparison groups: no SSD, SSD only, and SSD and LI. Siblings without LI or SSD were used as the TD control (no SSD) group (n = 146). Demographic characteristics for the SSD only and no SSD group were similar at baseline; however, the SSD and LI group had significantly lower PIQ and SES than the no SSD and SSD only groups. Literacy outcome measures at adolescent follow-up included credible standardized assessments of word-level reading, reading comprehension, and spelling. Parent reports were also used to determine which participants were enrolled in special services for reading. Additional speech and language skills were examined as outcome measures, but will not be discussed for the purposes of this review. Methods were outlined clearly and with sufficient detail for replication.

Detailed and appropriate statistical analyses revealed that the SSD and LI group performed significantly lower on all literacy outcome measures compared to the no SSD and SSD only groups. The literacy performance of children with SSD only did not differ significantly from children without SSD. However, effect sizes for all group comparisons were small.

Strengths of this study include its large sample size and use of stringent alpha criteria to account for multiple comparisons. Limitations include the lack of adjustments for SES and PIQ despite baseline differences in the SSD and LI group. As well, the use of siblings as a control group may limit generalization.

Overall, this study provides highly suggestive evidence that an early history of isolated SSD is not a significant risk factor for poor literacy outcomes, unless accompanied by a co-existing LI.

Nathan, Stackhouse, Goulandris and Snowling (2004) used a mixed design study to compare the

literacy outcomes between three groups: 19 children with isolated SSD, 19 children with SSD and co-morbid LI, and 19 TD controls. All three groups were individually matched on age, SES, and NIQ. An additional aim of this study was to investigate the critical age hypothesis; however, these results will not be discussed for the purposes of this review.

Participants were recruited in early childhood (ages 4-5 years) and assessed over three time points during preschool, kindergarten, and first grade. Participants were well described and inclusion and exclusion criteria were explained with a clear rationale. Gold standard measures of SSD and LI were used to classify children into subgroups. Methods were outlined clearly and thoroughly. Literacy outcomes included credible measures of letter name knowledge, single-word reading, prose reading, non-word reading, spelling, spelling from pictures, and PA skills.

Detailed and appropriate statistical analyses revealed that the literacy performance of children with isolated SSD did not differ significantly from the TD control group. Children in the SSD and LI group performed significantly worse on measures of phoneme awareness at T3. Children with persistent SSD at age seven had significantly poorer literacy outcomes than those whose speech difficulties had resolved. However, this group also involved children with LI, making these results equivocal for the purpose of this review.

Matching participants on age, SES, and NIQ is a significant strength of this study, as many other studies fail to do so and do not control for these variables in statistical analyses. It was limited by its small sample size, which may have impacted statistical power to detect significant differences between subgroups.

Overall, this study provides suggestive evidence that children with isolated SSD do not differ significantly from TD children on literacy outcome measures.

Lewis, Freebairn, and Taylor (2000) used a mixed design study to compare the literacy outcomes of 28 children with isolated SSD and 24 children with SSD and comorbid LI. This study also examined the rates of speech and language disorders among nuclear family members, but these results will not be discussed for the purposes of this review.

Participants were recruited in early childhood (ages 4-6) and classified into the two comparison groups based on well-defined and adequate criteria. Literacy skills were measured at follow-up when children were in the third and fourth grades. The two groups differed in age, SES, and PIQ at baseline; however, these differences were statistically controlled for at follow-up. Outcome measures included appropriate standardized tests of phoneme awareness, word decoding, reading comprehension, and spelling of both predictable and unpredictable words. Methods and procedures were clearly described with sufficient detail for replication.

Appropriate statistical analyses revealed that children with isolated SSD achieved mean standardized scores within the normal range on all literacy outcome measures. In the isolated SSD group, only one child scored below average on measures of reading and eight children had below average spelling scores. The spelling scores of children with isolated SSD were significantly lower than their reading scores. Children with both SSD and LI performed significantly poorer on measures of reading, spelling, and phoneme awareness compared to individuals with SSD only.

Strengths of this study include statistically controlling for differences in baseline demographic characteristics and its high inter-rater reliability for standardized articulation test scores. It was limited by its lack of a TD control group. Although the literacy outcomes of children with isolated SSD fell within normal limits on standard measures, a TD control group is needed to determine if children with isolated SSD are performing at or below the level of their same-aged peers.

Overall, this study provides suggestive evidence that generally, children with isolated SSD have age-appropriate literacy skills, but may exhibit some spelling difficulties. It further confirms that children with SSD and comorbid LI are at greatest risk for literacy difficulties.

Leitao and Fletcher (2004) conducted a mixed design study to examine the literacy performance of children with histories of SSD who were classified into two groups based on their speech error patterns: developmental speech errors and non-developmental speech errors. Developmental speech errors were defined as those that resemble normal developmental processes and are seen in younger TD children (e.g. fronting, stopping). Non-developmental speech errors were described as errors that do not regularly occur in TD speech (e.g. backing, initial consonant deletion).

A total of 36 children were recruited in early childhood (ages 5-6 years) and evaluated over two time points (T1 = beginning of primary school, T2 = end of primary school). Participants included 14 of these children who were available and reassessed at T2. Gold standard measures were used to ensure participants presented with moderate to severe SSD, in the absence of LI. Appropriate criteria were used to classify participants into subgroups based on developmental or non-developmental speech error patterns. Intra- and interrater reliability for these classifications were determined to be high.

Appropriate statistical analyses were conducted to ensure the children assessed at follow-up were representative of the original cohort who were not reassessed. Outcome measures included standardized tests of text reading accuracy, reading comprehension, spelling ability, word-reading efficiency, and phonological processing (i.e. PA skills, phonological memory, rapid naming). These measures were deemed to be only somewhat appropriate, as two of the four standardized tests did not have standardized scores

available and researchers chose to convert raw scores to age-equivalent scores.

A series of t-tests revealed that children in the non-developmental group performed significantly more poorly than the developmental group on measures of PA and reading comprehension. Both groups had literacy outcomes below the expected chronological age for measures of reading and spelling ability, however these results should be interpreted with caution, as age-equivalent scores are not a valid measure for making diagnostic decisions (Reynolds, 1981). The use of non-parametric tests in statistical analyses would have been more suitable considering the small sample size of both subgroups.

The use of age equivalents is a limitation of this study, which may be an inaccurate representation of performance. Additionally, its small sample size and lack of TD control group limits generalization.

Overall, this study provides somewhat suggestive evidence that children with isolated SSD who make non-developmental speech errors are at an increased risk for poor literacy outcomes.

Single Group Designs

Single group designs evaluate the results of one group in the absence of a comparison measure or control group, which inherently reduces the strength of these studies. Further limitations include potential participant selection bias and lack of generalization to the broader population.

Hesketh (2004) used a single group design to evaluate the literacy outcomes of 35 children (ages 6;6-7;9) with early histories of SSD, and age appropriate language skills. Researchers also investigated which factors in their earlier profiles were most predictive of literacy achievement.

Participants were originally recruited between the ages of 3;6 to 5;0 as part of an earlier intervention study. Criteria for participant inclusion was well described, however, a lenient inclusion criteria for determining LI was used. Literacy outcome measures included credible standardized assessments of word reading, spelling, and a complete phonological assessment battery (e.g. PA skills, non-word reading, digit-naming speed, fluency tests). Methods were outlined clearly and thoroughly.

Results of standardized testing revealed that participants' mean standard scores were within normal limits on all outcome measures. Of the 35 children that were tested, only four children had scores that fell more than one standard deviation (SD) below the mean, and just one child's score fell more than two SDs below the mean. Appropriate multiple regression analyses revealed that SSD severity did not predict literacy outcomes and that PA skills at age 3;6-5;0 were the best predictor of literacy achievement.

Criteria for identifying LI was a weakness in this study. Lenient criteria were used which may not have ensured the language skills of all participants were age appropriate.

Overall, this study provides suggestive evidence that children with early histories of isolated SSD are not significantly at risk for literacy delays.

Discussion

The current literature review examined the literacy outcomes of children with early histories of isolated SSD, in the absence of comorbid LI. Overall, results from the selected studies were reasonably consistent in providing suggestive evidence to support that the risk of poor literacy outcomes for children with isolated SSD, is relatively low. However, inconsistency in the evidence emerged throughout this review, warranting further investigation before definitive conclusions can be made.

Seven out of nine studies indicated that the risk of poor literacy outcomes for children with isolated SSD was minimal or non-existent (Hayiou et al., 2017; Hesketh, 2004; Lewis et al., 2015; Lewis et al., 2000; Nathan et al., 2004; Peterson et al., 2009; Sices et al., 2007). Two of these studies found that the literacy outcomes of children with isolated SSD did not differ from controls (Lewis et al., 2015; Nathan et al., 2004), two demonstrated that speech skills did not predict early literacy or literacy outcomes (Peterson et al., 2009; Sices et al., 2007), one found that speech skills predicted minor PA and spelling deficits that were mostly temporary (Hayiou et al., 2017), and two indicated that the mean standardized scores of literacy outcomes of children with isolated SSD were within normal limits (Hesketh, 2004; Lewis et al., 2000).

Comorbid Language Impairment

All studies that compared outcomes of children with isolated SSD to those with SSD and comorbid LI found that children with both disorders consistently demonstrated poorer literacy skills. Additionally, all studies that employed multiple regression analyses found that language skills were a stronger predictor of literacy outcomes than speech skills only. This is consistent with the growing body of evidence to suggest that a notable portion of the overlap between SSD and literacy difficulties may be attributed to the third variable of LI.

Considering the significant variety in the methodologies used in this critical review, it is not surprising that the results were somewhat variable. Most notably, literacy outcome measures and the age at which they were evaluated varied across studies, making them difficult to compare to one another. This resulted in inconsistent findings, as some studies found isolated SSD was associated with literacy difficulties in areas that other studies did not measure.

Phonological Awareness and Spelling

Three studies suggested that isolated SSD was associated with PA deficits and two indicated spelling difficulties. The extent to which PA deficits lead to

literacy difficulties remains unclear, as Peterson et al. (2009) found that isolated SSD predicted PA skills but not literacy outcomes, and Hesketh (2004) found that PA skills were the best predictor of literacy achievement. The predictive relationship between PA skills and literacy outcomes is outside the scope of this review and further research is needed to evaluate this association.

Nature of Speech Sound Disorder
There was evidence to suggest that SSD characteristics also influence literacy outcomes. Some studies indicated that persistent SSD to the age of school entry and the presence of disordered speech errors may increase risk for poorer outcomes. It is speculated that children with disordered or non-developmental speech errors are likely to have deficits in their phonological representations of words, leading to impaired PA skills and reading outcomes (Dodd, 1995). Interestingly, SSD severity was not associated with poorer outcomes.

Confounding Variables
Baseline characteristics for SES and PIQ often differed across participants and these variables were not always controlled for in statistical analyses. As well, many studies reported the inability to control for the effects of concurrent speech and language therapy on the extent of speech, language or literacy difficulties. Inability to control for each of these confounding variables may have influenced results.

Clinical Implications

The importance of the development of literacy skills in achieving academic and life success cannot be overstated. Speech-Language Pathologists (SLPs) have a pivotal role in supporting children with SSD. Based on the suggestive evidence presented in this review, SLPs working with children with SSD can be moderately confident that unless there are co-occurring language difficulties, the risk of poor literacy outcomes is relatively low. Inconsistencies and limitations in the evidence warrant further research to gather more compelling evidence on this topic before this statement can be used undeniably. Nonetheless, it is recommended that clinicians be mindful of the heterogeneity that exists among these individuals and monitor their literacy development accordingly. Subtyping SSD by the presence or absence of LI may be clinically useful when determining prognosis for literacy outcomes, as there is evidence to suggest that children with SSD and comorbid LI consistently demonstrate poorer literacy skills. Children with SSD and LI may benefit from direct early literacy instruction in addition to articulation and language therapy.

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