

**Prevalence and predictors of persistent speech sound disorder at eight-years-old:
Findings from a population cohort study**

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Abstract

Purpose: To determine prevalence and predictors of persistent speech sound disorder (SSD) in children aged 8.

Method: Data from the Avon Longitudinal Study of Parents and Children (ALSPAC) were used. Children were classified as having persistent SSD based on Percentage of Consonants Correct measures from connected speech samples. Multivariable logistic regression analyses were performed to identify predictors.

Results: The estimated prevalence of persistent SSD was 3.6%. Children with persistent SSD were more likely to be male and from families who were not home-owners. Early childhood predictors identified as important were: weak sucking at 4 weeks; not often combining words at 24 months; limited use of word morphology at 38 months; and being unintelligible to strangers at age 38 months. School-age predictors identified as important were: maternal report of difficulty pronouncing certain sounds and hearing impairment at 7 years; tympanostomy tube insertion at any age up to 8 years; and a history of suspected coordination problems. The contribution of these findings to our understanding of risk factors for persistent SSD and the nature of the condition is considered.

Conclusion: Variables identified as predictive of persistent SSD suggest that factors across motor, cognitive and linguistic processes may place a child at risk.

Keywords: Persistent, speech sound disorder, child, epidemiology, prevalence, speech, ALSPAC

Introduction

Despite variation in the rate of speech development, most children who are native speakers of English master accurate production of all vowels and consonants by age 8 (Dodd, Hulm, Hua, & Crosbie, 2003; James, 2001; Smit, 1993a, Smit 1993b; Templin, 1957). However, some individuals experience difficulties with speech production beyond this age and even into adulthood (Bralley & Stoudt, 1977; Felsenfeld, Broen, & McGue, 1992). These children with persistent speech sound disorder (SSD) constitute a substantial proportion (8.8%) of clinical caseloads (Broomfield & Dodd, 2004). This paper focuses on those children with clinically significant and persistent SSD that goes beyond the /s/ and /r/ distortions defined by Shriberg (1993) as common clinical distortions. Using data from a large longitudinal population study, prevalence at age 8 and associated risk factors are identified to aid our understanding of persistent SSD in the clinical setting.

Previous studies of prevalence of SSD

Studies of the prevalence of SSD have reported rates ranging from 2.3% to 24.6% (Jessup, Ward, Cahill, & Heating, 2008; Keating, Turrell, & Ozanne, 2001; Law, Boyle, Harris, Harkness, & Nye, 2000; McKinnon, McLeod, & Reilly, 2007; Shriberg, Austin, Lewis, McSweeney, & Wilson, 1997b; Shriberg, Tomblin, & McSweeney, 1999). This variation is most likely explained by two methodological issues. First, differences in the sampling process used. For example, decreasing prevalence rates have been associated with increasing age (McKinnon et al., 2007; Shriberg et al., 1997b) while differences in inclusion criteria relating to speech only versus speech and language impairment (Jessup et al., 2008) and variations in the definition of SSD in terms of

which types of errors constitute the disorder (Shriberg et al., 1999) may all exert an impact on the final estimated figure. Second, studies have used a variety of methods to identify SSD including parent/ teacher identification (Keating et al., 2001; McKinnon et al., 2007); formal assessments (Jessup et al., 2008) and speech sampling (Shriberg et al., 1999). The variability in methodology and dearth of age-specific prevalence figures make it difficult to draw firm conclusions about the prevalence of persistent SSD and there is a need therefore for an estimate to be determined from population based data using a robust means of case identification.

Factors associated with persistent SSD

Understanding the risk factors associated with persistent SSD may provide important clues regarding the nature of the disorder. In order to develop a model of risk factors that might form the basis of a new investigation, studies which investigated factors associated with SSD in early childhood and at school-age were examined to identify putative factors. Risk factors that occur early in a child's life do not necessarily play a causative role, however they may enable us to predict which children are likely to go on to have the more resistant and persistent disorders and thus facilitate early identification and prioritisation for intervention. Furthermore, the identification of early risk factors may indicate causative mechanisms that are in themselves amenable to interventions. Factors identified during school-age are associated with a concurrent diagnosis of SSD and therefore cannot be considered as risk factors. Nevertheless, they may suggest candidate variables that could be investigated at earlier ages.

Tables 1 and 2 summarise those studies which have focused on factors in early childhood and school-age which are associated specifically with SSD. Examination of the factors studied shows no consistent modelling of risk for SSD across studies and thus the factors investigated vary in each study. The different research designs and sampling

processes further undermine the comparability of findings and thus the possibility of drawing firm conclusions about which factors are predictive of SSD.

[Tables 1 and 2 about here]

An additional category of studies which have used a broad classification of speech language impairment were considered to see if this achieved greater clarity. This produced a number of additional candidate variables, which are summarised in table 3 and can be considered alongside the findings of the early childhood and school-age risk factor studies. When all the literature is considered together, a pattern of putative risk factors begins to emerge in terms of the child's demography, family/environmental context and developmental progression in speech and language, literacy and learning and other general development.

[Table 3 about here]

Demographic factors

Demographic factors considered in the studies included the child's gender, ethnicity, socio-economic status, and parental marital status. An association between male gender and SSD was found in some studies (Broomfield and Dodd, 2004; Campbell et al., 2003) but not others (Fox, Dodd, & Howard, 2002; Felsenfeld & Plomin, 1997). Similarly, socio-economic status was associated with SSD in one study (Campbell et al., 2003; Law, Rush, Schoon, & Parsons, 2009) but not observed in others (Broomfield & Dodd, 2004) and even shown to be protective in one study (Delgado, Vagi, & Scott, 2005). Variations in this factor can be influenced by how it is measured and a range of methods were used in these studies including maternal education, health insurance category, and parental literacy levels.

Family/Environmental factors

Family/environment factors covered a wide range of areas including family history of SSD; birth order and family size; multiple births; bilingualism in the home; overcrowding; and pre-school education. With regards to family history, Campbell et al., (2003), Fox et al., (2002), Felsenfeld and Plomin (1997), and Lewis and Freebairn (1997) all showed a positive association with SSD though this was not replicated by Broomfield and Dodd (2004).

Associations have been shown between lower language levels and birth order/family size (Choudhury & Benasich, 2003; Harrison & McLeod, 2010; Reilly et al., 2007; Stanton-Chapman, Chapman, Bainbridge & Scott, 2002; Tomblin, Hardy & Hein, 1991; Zubrick, Taylor, Rice & Seglers, 2007); overcrowding in homes (Law et al., 2009); absence of pre-school education and parental literacy levels (Law et al., 2009). With regards to languages spoken in the child's environment, some have found that children are more likely to be identified as speech or language impaired when the language spoken at home is different to that spoken out of the home (Reilly et al., 2009) while others have found the reverse (Broomfield & Dodd, 2004; Harrison & McLeod, 2010; Stanton-Chapman et al., 2002).

Family and environmental factors extend to the pre-and peri-natal factors studied by Delgado et al., (2005), Fox et al., (2002) and Wolke and Meyer (1999) as these factors relate to the medical status into which the child is born. Their studies produced mixed findings with some pre- and peri-natal factors showing a positive association with SSD.

Developmental progression in speech and language

Although one study did not observe a relationship between early language skills and later speech (Broomfield & Dodd, 2004), generally, delay in early language development has been positive associated with SSD (Highman, Hennessey, Sherman, &

Laitao, 1997) and with speech and language impairment generally (Bishop & Edmundson, 1987; Dale, Price, Bishop & Plomin, 2003; Glogowska, Roulstone, Peters & Enderby, 2006; Rescorla, 2002; Rice, Taylor & Zubrick, 2008; Roulstone, Miller, Wren & Peters, 2009). Moreover, the relationship between language development and SSD appears to remain relatively constant overtime with studies of school-age factors showing a similar pattern (Bishop & Adams, 1990; Broomfield & Dodd, 2004; Lewis et al., 2006; Raitano, Pennington, Tunick, Boada & Shriberg, 2004; Shriberg et al., 1999).

Developmental progression in literacy and learning

Studies which have focused on school-age factors have often considered the relationship between SSD and literacy skills. Indeed, given the association observed, there has been much debate about whether literacy skill should be regarded as an outcome of SSD or whether the two are part of the same underlying condition (Bird, Bishop & Freeman, 1995; Felsenfeld, Broen & McGue, 1994; Gillon & Moriarty, 2007; Hesketh, 2004; Larrivee & Catts, 1999; Leitao, Fletcher & Hogben, 2000; Nathan, Stackhouse, Goulandris & Snowling, 2004; Peterson, Pennington, Shriberg & Boada, 2009; Raitano et al., 2004; Rvachew, 2007; Sutherland & Gillon, 2007).

Other developmental factors

Beyond speech/language and literacy/learning development, other areas of development which have shown associations with SSD and/or language skills include use of pacifiers (Fox et al., 2002), delay in motor skills including feeding and dribbling (Highman et al., 2008; Hill, 2001; Hill & Bishop, 1998; Visscher, Houwen, Moolenaar, Lyons, Scherder, & Hartman, 2010; Visscher, Houwen, Scherder, Moolenaar, & Hartman, 2007; Robinson, 1991; Webster, Majnemer, Platt, & Shevell, 2005); general delays and medical conditions (Broomfield & Dodd, 2004; Delgado et al., 2005); and low birthweight (Stanton-Chapman et al., Yliherva, Olsen, Maki-Torkko, Koiranen, &

Jarvelin, 2001). With regards to hearing and ENT status, mixed findings have emerged with some studies showing a relationship with SSD and others suggesting none exists (Browning, Rovers, Williamson, Lous, & Burton, 2010; Campbell et al., 2003; Fox et al, 2002; Pagel Paden, 1994). Indeed, the findings of Paradise et al., (2005, 2007) from a large scale longitudinal study suggest that, in otherwise healthy individuals, OME and associated hearing loss are not associated with SSD.

In conclusion, the information from these studies provides a challenging picture for the clinician to interpret. None of the studies provides a comprehensive analysis of a range of potential variables and their relative importance in relation to predicting persistent SSD. However, the examination of the literature has generated putative factors that may be associated with persistent SSD. These have been used to establish a comprehensive model of risk encompassing demographic, environmental and developmental components of the child's history and characteristics. Data from large scale population-based studies offer the opportunity to study associations between a variety of potential predictor variables and later speech outcomes whilst controlling for other confounding developmental and social factors (Roulstone et al., 2011). The study reported in this paper uses data from the Avon Longitudinal Study of Parents and Children (ALSPAC), a prospective population study taking place in the southwest of England. This large study has collected detailed data on children's speech and language at several time points through direct assessment, along with a wide range of developmental, environmental and social data on the children and their families. This unique data set enables relatively comprehensive consideration of confounding effects in developing the risk model, through taking account of the relationships between such a wide variety of variables.

Numerous papers on a range of health and developmental factors have been reported on the ALSPAC data to date including five on findings relating to children's speech and language development (Roulstone, Law, Rush, Clegg & Peters, 2011; Roulstone, Miller, Wren, & Peters, 2009; Wren, Roulstone, & Miller, 2012; Wren, McLeod, White, Miller, & Roulstone, 2013; Wren, 2015). With regard to speech development and disorder, results from an analysis of the longitudinal data on a subset of the children (N=741) at ages 2,5 and 8 show a relationship between the child's speech error rates at age 2 and 5 and expressive language. SSD at age 8 was predicted by presence of speech errors at age 5 but not at age 2 (Roulstone et al., 2009). Further analysis has been reported on the characteristics of the sample in terms of speech production (Wren et al., 2013) and features which distinguish the groups identified through the process of case identification described in this paper (Wren et al., 2012).

The purpose of the study reported in this paper was to use the data available from this large scale population cohort to investigate persistent SSD and factors associated with it which could be used to estimate prevalence and identify predictor variables which could assist clinicians in identifying young children at risk of persistent SSD and aid our understanding of the nature of persistent SSD.

Aim

The aim of this study was to use direct assessment to identify children with persistent SSD at age 8 years. Following identification, the objectives were as follows:

1. To determine the prevalence of persistent SSD in children aged 8 years.
2. To identify early childhood and later school-age social, cognitive and linguistic predictors that are associated with a classification of persistent SSD at age 8.

Method

Avon Longitudinal Study of Parents and Children (ALSPAC)

This study used prospective cohort data from the Avon Longitudinal Study of Parents and Children (ALSPAC) - a transgenerational observational population study of health and development across the life span. Multiple measures of genetic, epigenetic, biological, psychological, social and other environmental factors have been collected in relation to outcomes. A description of the cohort profile is available (Boyd et al., 2012). During 1991 and 1992, 14,541 mothers enrolled in ALSPAC as they registered their pregnancy within the geographical area then known as Avon in the south west of the UK. Out of the initial 14,541 pregnancies, 14,062 live babies were born and 13,988 children were alive at 1 year.

The main data collection technique for the study has been postal surveys: the mothers completed four questionnaires before their babies were born and approximately annually thereafter, with 16 completed by the time the child was aged 13 years. In addition, since the children were aged 7 years, the entire cohort was invited to attend for direct assessment of varying aspects of development at regular intervals (known as the 'Focus' clinics). The second of these 'Focus' clinics was the 'Focus at 8' clinic in which speech and language was assessed.

The study website contains details of all the data that are available through a fully searchable data dictionary at <http://www.bris.ac.uk/alspac/researchers/data-access/data-dictionary/>. Ethical approval for the study was obtained from the ALSPAC Law and Ethics Committee and the Local Research Ethics Committees.

Participants

Participants in this study were those children who completed the speech and language session at the 'Focus at 8' clinic. All 13,314 children from the cohort who were still alive, consenting and with known addresses were invited to attend this clinic, with appointments arranged for when the children were aged 8 years, 6 months. A total of 7391 children (56%) attended, though records for one child were incomplete and their data were therefore excluded from any further analysis. The sample of children who attended was biased in that it contained a significantly greater proportion of higher educated and older mothers who were more likely to be living in owner-occupied housing. A slightly smaller proportion of boys and non-white children attended compared to non-attendees. Children who attended also had a slightly higher mean birthweight but there was no difference in mean gestation. It is worth noting however that with the size of the sample there are still many people in each category of the categorical variables and across the spectrum of the continuous variables.

The sample was heterogeneous in that it included all children who completed the speech and language session during the 'Focus at 8' clinic. Children were not excluded if they had co-morbid conditions such as cerebral palsy, hearing impairment, cleft palate, learning difficulties or any other condition which could have impacted on or caused their speech development. Data on the numbers of children in the sample who presented with co-morbid conditions are variable and incomplete and therefore unreliable. However, as a population sample, it could be assumed that prevalence for co-morbid conditions within the sample would be likely to match that for the UK population as a whole.

Similarly, attempts were not made to classify the sample into subgroups based on surface level speech errors or into speech only versus speech and language impaired children.

Rather, this paper reports on the group as a whole. It is anticipated that further research

will be carried out to consider the impact of speech only versus speech and language impairment in the future.

Speech sampling

At the 'Focus at 8' clinic, connected speech samples were collected during an expressive language task based on the Weschler Objective Language Dimensions (WOLD: Rust, 1996). In this activity, three tasks were performed: picture description; giving directions using a map; and explaining the steps involved in changing the batteries in a flashlight. All responses in this task were recorded digitally.

Identification of cases of persistent SSD

The process of case identification for persistent SSD within the cohort consisted of three phases:

1. **Listener judgement:** Assessors noted children whose speech sounded atypical for their age and whose errors were inconsistent with the local accent during the speech and language assessment. Children were assessed by qualified speech-language pathologists (85.9%) or psychologists trained in the delivery of the assessments by a speech-language pathologist (14.1%). Those children whose errors, as observed by assessors, were limited solely to common clinical distortions as defined by Shriberg, (1993) were identified. Typically in the UK, children with these types of errors are not seen for intervention at this age, and for this reason they were excluded from the definition of persistent SSD. The remaining children –those showing a range of substitution, omission, addition and atypical distortion errors, with or without the common clinical distortions, were considered potential cases.
2. **Transcription:** All sounds within the connected speech samples of the potential case group were transcribed and analysed using Computerized Profiling (CP:

Long, Fay & Channell, 2006). Broad transcription was used for sounds which were perceptually correct and for whole sound substitutions, omissions and additions while atypical distortion errors were narrowly transcribed.

Simultaneously a further 50 speech samples were transcribed from children who were randomly selected from the rest of the cohort (25 male, 25 female) to act as controls for the purpose of calculating prevalence. Transcribers were blind to the status of the sample being transcribed and were all qualified speech-language pathologists.

3. Comparison with controls: Means and standard deviations (SD) for the Percentage Consonants Correct (PCC)-late 8 and PCC-adjusted measures (PCC-A) (Shriberg et al., 1997a) were calculated for the 50 control children. PCC is a measure of speech accuracy in which the number of correctly produced consonants is counted and calculated as a percentage of the total target number of consonants within the sample. Given the age of the children, the PCC-late 8 (/s,z,ʃ,ʒ,θ,ð,ɹ,l/) was considered to be more sensitive for this age group than total PCC. The PCC-A was selected because this measure accepts common clinical distortions as correct but not atypical distortions, thus matching the criteria with which the children were selected at phase 1.

Means and standard deviations (*SDs*) were calculated separately for girls and boys and used to identify cases. Using the control group as a reference, potential cases were classified as persistent SSD if they scored less than 1.2 standard deviations below the mean on both the PCC late 8 and PCC-A. This cut off was selected for consistency with Records and Tomblin's (1994) observations that clinicians' decisions regarding diagnosis was associated with a cut-off composite score of approximately $-1.2SD$.

Thus the criteria for categorisation of persistent SSD in this study was a score of less than 1.2 *SD* below the mean of the control group on both the PCC-late 8 and the PCC-A on connected speech samples taken during picture description tasks. The data for these ‘case’ children were used in comparison with the rest of the cohort (N=6399) in subsequent analyses to identify early childhood and school-age predictors. The two groups of children identified exclusively with common clinical distortions and the group of potential cases who did not reach criteria for case status (that is, scored at or above 1.2SD below the means for either of the PCC-late 8 and PCC-A) were excluded from this analysis. A separate analysis revealed that these latter two groups showed distinct features in terms of demographic factors, IQ, nonword repetition and diadochokinetic tasks when compared to the case children and those in the rest of the cohort (Wren, Roulstone & Miller, 2012). Inclusion of their data could therefore have contaminated findings in the analyses carried out in this study.

A randomly selected sample of 48 children was transcribed by a second member of the original transcription team to check reliability. Point to point inter-judge agreement was 92.3%. As reliability was completed post-hoc it was not possible to resolve discrepancies and the first transcription was used in the analysis.

Identification of candidate predictor variables for persistent SSD

The ALSPAC data source was investigated to identify predictors potentially associated with persistent SSD based on the literature summarised in the introduction. Potential predictors were grouped into early childhood and school-age predictor variables and analysed separately. Early childhood predictors were those collected between the prenatal and immediate post birth period up to the age of the school entry

assessments¹. The one exception to this was the data relating to the range of languages spoken in the home, which was included in a questionnaire to the mothers when the children were aged 6 years and 9 months. However, the data relating to this question were included in the early childhood group because the impact would occur from birth. School-age predictor variables were those which were collected between the ages of 5 years, 9 months and 8 years, 7 months. The exception to this was the demographic variables which were included in the analysis of both early childhood and school-age predictors as potential confounding variables.

Tables 4 and 5 list the variables included in the categories of early childhood and school-age predictors respectively together with the timing and method of data collection. They were grouped conceptually for later analysis within each of the two categories. Further details on all the variables included in the analysis are available in the supplementary material files one and two online.

[Tables 4 and 5 about here]

Statistical Analysis

Following identification of the case group, the prevalence of persistent SSD was calculated using all children who had attended the speech assessment in the ‘Focus at 8’ clinic as the denominator minus those with missing samples. Following appropriate descriptive statistics (means, *SDs* and proportions), univariable and multivariable logistic regression analyses (Peters, 2008) were used to obtain odds ratios (ORs), their 95% confidence intervals and likelihood ratio *P* values for the associations between persistent SSD and various early childhood and school-age predictor variables. Both continuous and categorical explanatory variables were used in the analysis. The first stage of analysis tested all variables for their association with the outcome variable – that is, the

¹ Children in the UK start school in the September after their 4th birthday

child's case status at 8 years. Variables with a p-value of <0.1 in univariable analyses were retained for use in the multivariable analyses. A deliberately tolerant level was used so as not to miss any potentially influential variables at this point whereas $p<0.05$ was used in all the multivariable analyses. In addition, maternal age was retained in all regression models owing to evidence of its possible contribution in a related study using the same data set (Roulstone et al., 2009).

A 'staged' multivariable regression approach (Patel, Peters & Murphy, 2005) was then employed – first within the groups of conceptual variables, as listed in tables 4 and 5, and then across groups, resulting in a final model of demographic and early childhood and school-age predictors independently associated with case status.

At each step in this process, only one variable was dropped from or added to the model at any one point so as to ensure that all independent influences on the outcome were retained. In the final stage of analysis, variables from the above within-group multivariable analyses that were associated with case status ($p<0.05$) were combined into two final models of predictors associated with case status (early childhood and school-age predictors). This between-groups model was adjusted for the child's gender and social class and maternal age.

Given the nature of the study and the number of variables collected, there were missing data at various points in the analysis. In each analysis, we worked with the maximum data available for the variables under investigation. All analyses were conducted in Stata version 13.

Results

Prevalence of persistent SSD

Figure 1 summarises the process of case identification. Of the 7390 children who had data from the Focus at 8 speech and language assessment, 991 children had speech which sounded immature or unusual for their age and errors which were inconsistent with the local accent during the listener judgement phase. From the remaining 6399 children whose speech sounded typical for their age and accent, 50 had been selected at random as a control group. The data for three of the control children were markedly outside the range of the remaining 47 controls – specifically, PCC-A scores of 71.9, 74.0 and 77.4 compared with a range of 94.7-100 for the remaining 47 controls. Inclusion of these children’s data would have markedly altered the SD cut-offs for the identification of the case group, so their data were not used to calculate means and SDs for the control sample but were used, along with the rest of the cohort, in the regression analyses for identification of predictor variables. Table 6 provides the means and SDs for both the PCC late 8 and the PCC-A for the 47 controls plus the calculation of the -1.2SD cut-off for each measure.

[Table 6 about here]

[Figure 1 about here]

Of the 991 children who were identified through listener judgement as showing speech which was atypical for their age and accent, 580 made common clinical distortions exclusively (Shriberg, 1993), while the remaining 411 showed a range of whole sound substitutions, omissions, atypical distortions and additions, with or without common clinical distortions as described above.

Within the sample of 411 potential cases, five cases were removed from all analyses due to missing speech samples. The rest of their data was removed from the study for all further analyses. For the remaining 406 children, PCC-late 8 and PCC-A scores from the transcribed connected speech samples were compared with those obtained from the 47 controls and used to confirm cases of persistent SSD.

Two children within the potential case group had a PCC-A score of 100% and a PCC score of less than 100%. This would suggest that all their errors were distortions of sibilants and rhotics (because PCC-A scores all speech errors, including common clinical distortions as correct whereas PCC scores them as incorrect) and they were therefore added to the group of children previously identified as showing only common clinical distortions, taking the total in this group to 582. Therefore 582/7385 (total cohort of 7390 minus 5 with missing data), or 7.9% (95% CI: 7.3%, 8.5%) of the cohort presented with common clinical distortions (Figure 1).

Of the 404 remaining children identified through listener judgement as potential cases, 263 (169 boys and 94 girls) were confirmed as cases based on cut-off values derived from PCC-late 8 and PCC-A scores obtained from the 47 control children. From a total sample size of 7385, 263 cases yields an estimated prevalence of 3.6% (95% CI: 3.1, 4.0) overall. In terms of gender, this equates to a prevalence of 4.6% for boys (based on a total sample of 3687 males) and 2.5% for girls (total sample of 3698 females), giving a ratio of 1.8:1.

Table 7 shows the descriptive statistics for the PCC-late 8 and PCC-A scores across the three groups (controls, confirmed cases of persistent SSD and potential cases who did not reach the criteria for case status). This confirms that children with persistent SSD had a lower mean and larger SD for each measure, although there is some overlap in the ranges for both PCC-late 8 and PCC-A for all three groups.

[Table 7 about here]

Predictors associated with persistent SSD

The regression analyses were conducted using maximum numbers of 263 children with confirmed persistent SSD and the 6399 children comprising the rest of the

cohort (including all 50 of those who had been randomly selected as controls and whose samples had been transcribed). For the final across groups analysis, a total sample size of 5066 children (out of a possible 6662) was available for the early childhood predictor variables and 4303 children for the school-age predictor variables.

In univariable analysis, compared with the rest of the cohort, case children were more likely to be boys, to have mothers who were less well educated and in manual professions, and to live in rented homes. These socio-demographic factors were then considered separately for the early childhood and school-age predictor variables alongside the other grouped variables in a staged process of within-group and between-group multivariable regression models which were reduced using a manual forward and backward stepwise process. The results of the univariable analysis are available online in supplementary material three and four.

Early childhood predictors

Factors with p values greater than 0.1 following univariable analysis were ethnicity, maternal age, attendance at preschool provision up to age 33 months and at 54 months, reading to the child at age 42 months, preterm delivery, pregnancy complications (except vaginal bleeding), method of labour, breastfeeding, smoking in early pregnancy or pre-pregnancy, teenage motherhood, maternal depression and anxiety, stuttering at 38 months, communication, social and gross motor scores at 6 months, low birth weight, laterality at 42 months, and various feeding factors at 4 weeks. These factors were excluded from further analysis.

[Table 8 about here]

Table 8 lists the variables where the p-value following univariable regression was less than 0.1. These variables were taken forward to the within-group multivariable

analysis. From these analyses, 13 variables (gender, maternal occupation², home ownership, mother reads to child at 18 months, overcrowding at 8 weeks, family history of referral to speech and language therapy, intelligibility to others, combining words at 24 months, use of irregular grammar at 24 months, range of word morphology at 38 months, fine motor skills at 42 months, gross motor skills at 42 months, weak sucking at 4 weeks and dribbling at four weeks) showed some evidence of association ($p < 0.05$) at this stage (Table 9). All of these, except maternal social class ($p = 0.014$), were retained in the between-group multivariable analysis stage to end with the variables listed as the ‘best’ model.

Maternal social class was excluded because of its association with home ownership ($p < 0.001$). Although both were to some extent independently associated with the outcome in the relevant within-groups model, they are likely to confound each other in later models. Hence only the measure with the stronger evidence was retained in the models presented here.

[Table 9 about here]

Table 9 also shows the results of the final between-groups multivariable regression analyses, retaining gender as an important covariate, given the higher prevalence rating for boys, even though its association was no longer significant ($p = 0.17$). Five variables were independently associated with case status. Case children were more likely to come from families who did not own their own homes ($p = 0.036$), to be less intelligible to others at 38 months ($p < 0.001$), to use single words rather than two or three word phrases at 24 months ($p = 0.006$), to use incorrect word morphology at 38 months ($p = 0.001$) and to have had a weak suck as a baby ($p = 0.05$).

² Where information on maternal occupation was unavailable, paternal occupation was used instead

Of these variables, the strongest association was low intelligibility to strangers at 38months (OR=2.38). Children who used single words rather than combining words at 24 months were nearly twice as likely to be case children (OR=1.81) while those with higher scores on the word morphology task at 38months (OR=0.91) were less likely to be case children. Being part of a family who did not own their own home and having a weak suck at age 4 weeks was associated with odds ratios of 1.5 and 1.45 respectively.

School-age predictors

Factors with p values greater than 0.1 following univariable analysis included ethnicity, maternal age and the diadochokinetic (DDK) tasks requiring repetition of /pə/ and /kə/. These factors were excluded from further analysis. Table 10 provides descriptive statistics and univariable regression models comparing children with persistent SSD against the rest of the cohort for those variables with the designated strength of evidence ($p < 0.1$) of an initial association. Thirteen variables showed some evidence of association ($p < 0.05$) with case status following the within-group multivariable analyses (Table 11). These 13 variables were taken forward to the final stage of modelling across all groups of variables.

[Table 10 about here] [Table 11 about here]

Table 11 shows that, in the final model, four variables remained strongly associated with case status (reported difficulty pronouncing certain sounds and nonword repetition $p < 0.001$; gender $p = 0.003$; tube insertion $p = 0.005$). There was weaker evidence for three further variables (home ownership $p = 0.028$; suspected coordination problem $p = 0.011$; hearing impairment $p = 0.017$). The strongest association was for reported difficulty pronouncing certain sounds (OR=5.6). Children who had tube insertion and/or hearing impairment, and those for whom coordination problems were suspected were roughly twice as likely to be within the persistent SSD case group (ORs around 2) while

higher scores on the nonword repetition task were associated with a decreased risk of being in the case group (OR 0.82). In terms of demographic factors, case children were more likely to be male and in families who did not own their own homes (ORs of approximately 1.6).

Discussion

Using prospectively collected data from a large population-based cohort, we obtained a prevalence estimate for persistent SSD at 8 years of 3.6%. Children with persistent SSD in this study were more likely to be boys and to be from families who do not own their own homes. Early childhood predictors associated with persistent SSD were lower socio-economic status, low intelligibility to strangers at 38 months, early speech and language delay and weak sucking as a baby. School-age predictors associated with persistent SSD were hearing impairment (above 20db loss) on assessment at age 7 years, a history of tympanostomy tube insertion, parental report of difficulty pronouncing sounds at age 7, poor performance on nonword repetition tasks and reports of suspected motor coordination problems.

Limitations

As with any study of this size which takes place over an extended period of time, retention of participants and missing data are a problem and bias in the samples appears. Children attending the 8 year clinic had higher educated and older mothers and were more likely to be living in owner-occupied housing than those children who did not attend though good coverage across all levels of education and socio-economic status was maintained in the sample.

The control group was limited to just 50 participants, of whom 3 were identified as outliers based on their PCC-A and PCC-late 8 scores, relative to the rest of the control

group. Time and funding considerations prevented the transcription of a greater number of control samples but without doubt, this would have added weight to the analysis. The three outliers constitute 6% of the control sample which is nearly twice the size of the prevalence estimate obtained from this dataset. It is impossible to know whether the three identified outliers represent exceptional data or whether the rest of the cohort, which functioned as a control group for the identification of predictor variables, are in fact more varied than has been assumed. For the purposes of this paper, it has been assumed that three outliers do indeed constitute exceptional data but without the benefit of further transcribed samples from the rest of the cohort, this cannot be confirmed. The findings from this paper must therefore be interpreted bearing this in mind.

Information on co-morbidities was patchy and therefore unreliable making it impossible to determine the extent to which a child's presenting SSD was part of a more general learning or developmental disorder or linked to a diagnosis of Childhood Apraxia of Speech or other neurological or structural condition. Given this limitation, the study has focused on reporting the results of the large heterogeneous group of children who could be described as having persistent SSD and has not tried to link findings to aetiology or to identify subgroups in terms of risk factors. Whilst it was possible to build language scores into the regression analysis to allow some consideration of the level of comorbidity with language deficits, an analysis has not been carried out to determine to what extent co-morbid language impairment can explain the findings. Previous work by the authors using a subset of the data presented here combined with longitudinal findings found that expressive language skill at age 2 and 5 was predictive of speech outcome at age 8 (Roulstone et al., 2009) suggesting that many children in the sample described in this paper may have had additional language problems. This is consistent with the findings of Shriberg et al. (1999) who found that almost half of their sample of children

with SSD also had language impairment. Many previous studies (Reilly et al., 2010; Stanton-Chapman et al., 2002; Tomblin, Smith, & Zhang, 1997; Zubrick et al., 2006) have looked at risk factors for language impairment in younger children. A future investigation which considers the relative importance of speech factors compared to language factors in older children in this dataset would be invaluable in understanding which variables explain both speech and language problems and which are exclusive to one or the other.

The benefits of using the large ALSPAC dataset are offset by the limitations involved in collecting and analysing the information within such a large cohort. This impacts on the level of detail available for some variables. For example, data on the family history of speech and language impairment or interventions received rely on single questions requiring parental recall. However, concurrent evidence detailing intervention suggests a very low dosage of therapy was typical (Glogowska, Roulstone, Enderby & Peters, 2000). There were also limitations with the variables relating to hearing. Information was not available on the dates of tube insertion and pure tone audiometry results were only available for the children at age 7 and not at the same time as the speech assessment.

Finally, it was not possible to complete the reliability of the transcribed samples until later in the study, meaning that discrepancies were uncovered after it was possible to resolve these. However, a reliability figure of 93% is comparable to that of other studies of normal and disordered speech (Shriberg et al., 1999). The study started with a large number of variables and hence while the analyses would have attended to a wide variety of potential confounding effects the results of all such models should be considered exploratory, at least until replicated elsewhere.

Case identification

Identification of the case group required making a distinction between what constitutes pathology and what can reasonably be considered ‘typical’ behaviour. The wide range of variation in typical development of speech and language and the continuum from typical to atypical speech makes this process difficult while the range of definitions used in the literature seems to confirm that there is no easy solution to this dilemma. The context of this study, within a large population-based sample, allowed the identification of a case group in comparison to their immediate peers, rather than needing to use normative data from very different samples. However, the overlap between the scores of those children with observed errors and the 50 drawn from the rest of cohort shows that it is still challenging to identify distinct case and non-case groups.

Some might consider that the term persistent SSD should include children with common clinical distortions. However, for the purpose of this study we have opted for a narrower definition of persistent SSD, based on the children who would typically access services in the UK, in which children whose speech errors are restricted to common clinical distortions are excluded..

Nevertheless, it is important to acknowledge that the common clinical distortions group may include children who would have fulfilled criteria for SSD at a younger age and may also share some characteristics with those children defined as persistent SSD in this study. Whilst there is a need to look at trajectories within the ALSPAC sample to determine case status over time, a separate study has considered how the common clinical distortions group compared with the persistent SSD group and those who did not reach criteria for case status in terms of gender, socioeconomic status, IQ, nonword repetition and diadochokinetic (DDK) tasks (Wren et al., 2012). While the case group and those who did not reach criteria for case status shared similar characteristics and were different to the common clinical distortions group on most measures, the common

clinical distortions group were more similar to the persistent SSD group on measures of DDK, suggesting that there may be some overlap in their areas of difficulty in the area of rapid speech movements.

Prevalence

The prevalence of 3.6% for persistent SSD obtained in this study is consistent with findings from other studies carried out in other English speaking countries (Kirkpatrick & Ward, 1984 – 4.6% children aged 5-7 in Australia; Shriberg et al., 1999 – 3.8% children aged 6 in the US). However, there are important differences in how the numbers are derived. Single word naming (Kirkpatrick & Ward, 1984) provides a rapid means of case identification but may miss errors which occur across word boundaries (Howard, 2004, 2007) and which would be observed in the connected speech samples used in this study and that of Shriberg et al.

This study and Shriberg et al share other characteristics (children with concomitant language impairment and motor disorder were included in the sample) but differ in the way that cases were identified. Shriberg et al.'s figures are based on a multiple categorical system from the Speech Disorders Classification System (SDCS) in which a range of different possible classifications of speech status are available. Prevalence was calculated for the specific category of Speech Delay which is based on the presence of substitution or deletion errors for four or more consonants or two or three consonants and vowels (Shriberg et al., 1997b). In contrast, this study used a cut-off point on two measures of PCC compared with a control group of children. Whilst it is anticipated that the two case groups are broadly similar, some differences in the composition of each group are likely to exist.

In other studies that also used direct assessment of children's speech, higher prevalence figures of 16.5% and 8.7% for children aged 6 were obtained (Tuomi &

Ivanov, 1977; Jessup et al., 2008). These studies used a more tolerant definition of case status, including children with milder problems. If children with common clinical distortions only (7.88% of the sample) had been included in the persistent SSD group here, prevalence would have reached 11.4%, a more comparable figure.

Nevertheless, the prevalence figure of 3.6% for persistent SSD alone is a robust estimate of clinical need. It was obtained from a large population study and has been defined with clear parameters. It suggests that in a class of thirty school children aged 8, there is likely to be one child with a clinically significant speech problem.

Predictor variables associated with persistent SSD

Analysis of the predictor variables using a staged multivariable regression approach led to the identification of a small number of important variables within the two broad categories of early childhood and school-age predictors based on the age of the child when the variable was measured. Results are discussed below across the categories of early childhood and school-age predictors but within the subcategories of demographics, family/environment, speech and language performance, literacy and learning skills, and other developmental measures.

Demographics

Low socio-economic status (SES), as measured by home ownership, was an important predictor of persistent SSD for both the early childhood and school-age categories while male gender was an important variable in the school-age predictors only. With regard to SES, reports in the literature have been conflicting with some providing support for a relationship with SSD (Shriberg et al., 1999; Winitz and Darley, 1980) and others not (Keating et al., 2001; McKinnon et al., 2007). Variation in how SES is measured may account for these differences while Law et al. (2000) point to the

possibility that SES could be operating as a proxy variable in some instances and should therefore be treated with caution.

In this study, maternal education and occupation were also included as measures of SES in the analysis but only home ownership remained in the final model. This contrasts with Campbell et al.'s (2003) study of risk factors for SSD in three-year-olds which found that of two measures of SES included in their study, maternal education was the more important when considered with medical insurance category as a second measure. This raises the question for this study of whether low SES was an important factor in accounting for variance in the findings or whether another factor or factors related to home ownership were associated with persistent SSD. Factors commonly linked to living in rented accommodation such as lack of stability and financial security, suitability and size of living area and quality of accommodation could impact on family interactions and thus speech development over and above low SES in isolation.

A greater number of boys than girls were identified with persistent SSD in this sample in common with other prevalence studies (Campbell et al., 2003; Harrison & McLeod, 2010). Whilst this variable was important in the school-age predictor group, the results from the early childhood predictors analysis suggest that when considered alongside other factors, it is not as important as variables relating to the environment and early development. This is consistent with the findings of Fox et al. (2002) who found gender to be less important than family history, pre- and perinatal history and the use of pacifiers.

Family/Environment

None of the variables relating to family and environmental factors remained in the final model. While some showed evidence at the within-groups multivariable stage (mother reading to the child daily at 18 months, overcrowding at 8 weeks prenatally and

family history), they did not remain in the model after adjustment for the other variables in the between-group multivariable analysis. This contrasts with Harrison and McLeod's (2010) study of 4983 children which found that parity (older siblings) was a risk factor while use of other languages by parents was protective. However, case status in the Harrison and McLeod study was determined by parental report of concern rather than direct assessment and analysis of speech, as in this study. This method of classification achieved a positive response from 25.2% of the sample, suggesting a much larger and more diverse case group than the 3.6% identified in this study.

Whilst there is contradictory evidence in the literature for many of these factors, the most surprising finding is arguably family history, albeit that this was only confounded by other factors in the between-groups model. This factor has emerged as an important predictor in a number of studies of SSD specifically and speech and language impairment more generally (Campbell et al., 2003; Felsenfeld & Plomin, 1997; Fox et al., 2002; Lewis et al., 2007; Lewis et al., 2006; Tomblin et al., 1991). However, family history was measured by a single questionnaire item in this study regarding referral to specialist services, relying on parents' ability to recall information from their own early childhood. By contrast, other studies used more comprehensive questionnaires devoted specifically to the issue of family history of speech and language difficulties (Campbell et al., 2003; Felsenfeld & Plomin, 1997; Tomblin et al., 1991), or used interviews and direct testing as part of a genetic linkage study (Lewis et al., 2007; Lewis et al., 2006) or asked whether the parents had experienced problems with speech and language in childhood rather than referral to specialist services (Fox et al., 2002).

Other environmental factors that have been associated with SSD generally but were not identified as independently associated with persistent SSD in this study included factors relating to the birth. In the literature there are mixed findings (Campbell

et al., 2005; Fox et al., 2002) and, while the findings from this study suggest that ‘pregnancy complications’ and ‘smoking during pregnancy’ were not important, the measure used to account for this was crude and it is possible that more sensitive measures may produce associations with specific aspects related to pregnancy and birth. Overcrowding, family size, attendance at playgroup, nursery or with a childminder and reading to your child were not associated with persistent SSD at age 8 once other factors had been taken into account.

Speech and language performance

‘Difficulty pronouncing sounds’, as measured via parent questionnaire when the children were aged 7 years, was the strongest predictor variable. This is not surprising given the method of identification of persistent SSD. The second strongest predictor was difficulty being understood by non-family members at age 38 months. For many children in the persistent SSD group, their presentation at age 8 will reflect a speech sound system which was typically immature when younger so the fact that they presented with unintelligible speech at a younger age is not remarkable. Further research is needed to determine the degree to which this association is consistent over time and to what extent it also identifies children with SSD at younger ages as well as the persistent group. If an association is not found for children with transient SSD when younger rather than persistent SSD, then this could act as a useful clinical marker for persistent SSD.

Strong associations were also observed between persistent SSD and combining words at age 24 months and use of word morphology at 38 months, as reported by the mother. Combining words was important at age 24 months but not at age 38 months suggesting that this risk factor is age dependent. Highman et al. (2008) also found word combinations to be a predictor of later speech status, in their case of Childhood Apraxia of Speech (CAS), as well as difficulties with gross motor development, feeding and

dribbling. This suggests a possible motor component to this delay. It is possible that children at risk of persistent SSD may have greater difficulty in making the sequenced fine movements of the articulators required for speech and have particular trouble with making the transition to word combinations, where even greater coordination of movement is required. The evidence from the school-age ‘other developmental predictors’ discussed below, which found an association between suspected coordination problems and PSD status, provide further support for this idea.

Correct use of word morphology at 38 months was based on parental report of 12 items. The odds ratio reported showed that children scoring higher on this were less likely to be case children. Whether this reflects a language difficulty per se or a difficulty in expressing word morphology due to restrictions in speech production is not clear from these results and needs further investigation.

The results from this study suggest an association between poor performance on non-word repetition and persistent SSD status. Non-word repetition is well recognized as a measure of phonological working memory (Gathercole, Willis, Baddeley, & Emslie, 1994) and has also been associated with measures of vocabulary development in typically developing children and with poor performance by children with language impairment (see Coady & Evans, 2008 for a review). Yet non-word repetition requires a number of processing skills beyond that of memory, including speech perception and discrimination, phonological encoding, phonological assembly, motor planning, and articulation – skills associated more typically with speech processing and production than language (Stackhouse & Wells, 1997). Indeed, a recent investigation by Farquharson (2015) found children with remediated SSD aged between 9 and 13 performed significantly worse on a test of non-word repetition. It is not yet clear to what extent language skills may also influence the associations observed.

The remaining variables in this group, WOLD comprehension, DDK accuracy and phoneme deletion, were important at the univariable stage but were lost when considered alongside difficulty pronouncing sounds and nonword repetition. Dropping such factors from the model does not necessarily imply that they do not reflect important underlying characteristics, only that there is shared variance among certain variables (for example, nonword repetition and phoneme deletion) and influences are better represented by other (more statistically dominant) measures.

Literacy and learning performance

Variables relating to literacy and learning performance did not remain in the final model in either the early childhood or school-age predictors groups when factors unrelated to literacy and learning were included. This suggests that although there is an association between literacy and learning and persistent SSD, there is considerable shared variance with other variables within the model which emerge as more statistically dominant in the analysis.

Other early developmental

While a range of measures remained important at the within groups stage (measures of gross and fine motor skills at 42 months, dribbling at 4 weeks, intelligence and memory) only weak sucking at four weeks, suspected coordination problems, and variables related to hearing (presence of hearing impairment and previous insertion of tympanostomy tubes) remained in the final model.

With regard to hearing, whilst there is the suggestion that there may be an impact on some subtle language skills such as aspects of phonological processing or verbal working memory (Majerus et al., 2005; Nittrouer & Burton, 2005), there is a strong body of evidence to suggest negligible impact of otitis media and associated hearing loss on the development of speech and language (Paradise et al., 2005, 2007; Roberts, Rosenfeld

& Zweisel, 2004a; Roberts et al., 2004b). The contrast in these findings may relate to the differences in when the measures were taken. While Roberts et al., (2004a) carried out a meta-analysis of 14 studies and Paradise and colleagues collected longitudinal data, the measure used in the analysis reported here was a single hearing assessment and parental report of whether or not tubes had been fitted. Complementary data provided by successive hearing tests over time and information on the dates and timings of tube insertion would provide a more complete picture and no doubt provide clearer data relating to hearing history rather than performance at a single point in time.

Oral sucking habits have been associated with SSD in other studies of risk factors for SSD (Highman et al., 2008; Tomblin et al., 1991). Moreover, there is evidence that associates poor sucking with other developmental factors such as early growth faltering, low IQ and gross motor development (Emond, Drewett, Blair, & Emmett, 2007; Motion, Northstone, Emond, Stucke, & Golding, 2002). It is thought that sucking difficulties in the first few weeks of life may be a marker of subtle neurological impairment, accounting for the lowered IQ score.

Links between intelligence and memory and speech development have been shown in previous studies (Shriberg et al., 1999; Keating et al., 2001). However, this study suggests that coordination skills are more important in children with persistent SSD at age 8. This is consistent with reports in the literature of links between general coordination problems and speech impairment (Gaines & Missiuna, 2007; Gibbon, 2002; Hill, 2001; Hill & Bishop, 1998; Visscher et al., 2007, 2010; Robinson, 1991; Webster et al., 2005).

Nature of persistent SSD

The pattern of predictor variables which emerge as important in this dataset help to further our understanding of the nature of persistent SSD. The findings relating to

motor skill, as evidenced by a number of variables, suggest that this could be a feature common to many children identified as persistent SSD. Problems with weak sucking as a baby and suspected coordination disorder point to a more motorically based deficit of speech and while DDK, another measure relating to oromotor skill, was not important in the final model, it was identified as a distinguishing feature in previous work using the same dataset (Wren et al., 2012). In contrast, most measures of cognition did not remain in the final model. The exception to this was non-word repetition which, as has already been discussed, encompasses a wide range of skills including memory, phonological processing and speech motor skill. While the findings of this study support the concept of SSD as being multifactorial in nature, and the sample included in the study were undoubtedly heterogeneous, the results hint at the possibility that when SSD persists it is multifactorial in nature and there is involvement across more than one domain of motor, cognition and language.

Conclusion

This study has investigated persistent SSD in children in a population study and obtained an estimated prevalence of 3.6%. The final model of risk factors described in the paper provides useful information on what factors might be important to consider in assessing an individual child's risk for persistent SSD in the clinical setting. In the early years, limited combining of words at 24 months and use of word morphology at 38 months as well as difficulty being understood by strangers at age 3 could be useful clinical markers alongside demographic factors relating to home ownership and gender and difficulties with nonword repetition at school-age.

The predictor variables also provide useful information on the nature of persistent SSD. While it is known that speech development requires intact motor, cognitive and

linguistic skills, difficulty with any one of these areas might lead to differences in the timing and pattern of SSD while problems in more than one area may be an important factor in determining why some children's problems with speech persist. Further research is needed to investigate this hypothesis and to determine the degree to which intervention can impact on these underlying skills to remediate SSD before it can be classified as persistent.

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References

- Bird, J., Bishop, D. V. M., & Freeman, N. (1995).** Phonological awareness and literacy development in children with expressive phonological impairments. *Journal of Speech and Hearing Research, 38*, 446-62.

- Bishop, D. V., & Adams, C.** (1990). A prospective study of the relationship between specific language impairment, phonological disorders and reading retardation. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 31, 1027.
- Bishop, D. V., & Edmundson, A.** (1987). Language-impaired 4-year-olds: Distinguishing transient from persistent impairment. *Journal of Speech & Hearing Disorders*, 52, 156-173.
- Boyd, A., Golding, J., MacLeod, J., Lawlor, D., Fraser, A., Henderson, J., Molloy, L., Ness, A., Ring, S. & Davey-Smith, G.** (2012). Cohort profile: The ‘Children of the 90s’ – the index offspring of the Avon Longitudinal Study of Parents and Children. *International Journal of Epidemiology*, 42, 111-127.
- Bralley, R. C., & Stoudt, R. J.** (1977). A 5-year longitudinal study of development of articulation in elementary school children. *Language Speech and Hearing Services in the Schools*, 8, 176-180.
- Broomfield, J., & Dodd, B.** (2004). The nature of referred subtypes of primary speech disability. *Child Language Teaching and Therapy*, 20, 135-151.
- Browning, G.G., Rovers, M.M., Williamson, I., Lous, J. & Burton, M.J.** (2010) Grommets (ventilation tubes) for hearing loss associated with otitis media with effusion in children. *Cochrane Database of Systematic Reviews*. Issue 10. Art. No.: CD001801.
- Campbell, T. F., Dollaghan, C. A., Rockette, H. E., Paradise, J. L., Feldman, H. M., Shriberg, L. D., . . . Kurs-Lasky, M.** (2003). Risk factors for speech delay of unknown origin in 3-year-old children. *Child Development*, 74, 346-357.
- Choudhury, N., & Benasich, A. A.** (2003). A family aggregation study: The influence of family history and other risk factors on language development. *Journal of Speech, Language & Hearing Research*, 46, 261-272.
- Coady, J.A. & Evans, J.L.** (2008) Uses and interpretations of non-word repetition tasks in children with without specific language impairments (SLI). *International Journal of Language and Communication Disorders*, 43, 1-40.

- Dale, P. S., Price, T. S., Bishop, D. V. M., & Plomin, R.** (2003). Outcomes of early language delay: I. predicting persistent and transient language difficulties at 3 and 4 years. *Journal of Speech, Language & Hearing Research, 46*, 544-560.
- Delgado, C. E. F., Vagi, S. J., & Scott, K. G.** (2005). Early risk factors for speech and language impairments. *Exceptionality, 13*, 173-191.
- Emond, A., Drewett, R., Blair, P., & Emmett, P.** (2007). Postnatal factors associated with failure to thrive in term infants in the Avon Longitudinal Study of Parents and Children. *Archives of Disease in Childhood, 92*, 115-119.
- Farquharson, K.** (2015) After dismissal: Examining the language, literacy and cognitive skills of children with remediated speech sound disorders. *Perspectives on School-Based Issues, 16*, 50-59.
- Felsenfeld, S., Broen, P., A., & McGue, M.** (1992). A 28-year follow-up study of adults with a history of moderate phonological disorder: Linguistic and personality traits. *Journal of Speech and Hearing Research, 35*, 1114-1125.
- Felsenfeld, S., Broen, P. A., & McGue M.** (1994). A 28-year follow-up of adults with a history of moderate phonological disorder: Educational and occupational results. *Journal of Speech and Hearing Research, 37*, 1341-53.
- Felsenfeld, S., & Plomin, R.** (1997). Epidemiological and offspring analyses of developmental speech disorders using data from the Colorado adoption project. *Journal of Speech, Language, and Hearing Research: JSLHR, 40*, 778-791.
- Fox, A. V., Dodd, B., & Howard, D.** (2002). Risk factors for speech disorders in children. *International Journal of Language & Communication Disorders / Royal College of Speech & Language Therapists, 37*, 117-131.
- Gaines, R., & Missiuna, C.** (2007). Early identification: Are speech/language-impaired toddlers at increased risk for developmental coordination disorder? *Child: Care, Health & Development, 33*, 325-332.

- Gathercole, S. E., Willis, C., Baddeley, A., & Emslie, H.** (1994) The children's test of nonword repetition: A test of phonological working memory. *Memory*, 2, 103-127.
- Gibbon, F.** (2002). *Investigations in clinical phonetics and linguistics*. Mahwah: Lawrence Erlbaum Associates.
- Gillon, G. T., & Moriarty, B. C.** (2007). Childhood apraxia of speech: Children at risk for persistent reading and spelling disorder. *Seminars in Speech and Language*, 28, 48-57.
- Glogowska, M., Roulstone, S., Enderby, P. & Peters, T.** (2000) Randomised controlled trial of community based speech and language therapy in preschool children. *British Medical Journal*, 321, 923.
- Glogowska, M., Roulstone, S., Peters, T. J., & Enderby, P.** (2006). Early speech- and language-impaired children: Linguistic, literacy, and social outcomes. *Developmental Medicine & Child Neurology*, 48, 489-494.
- Harrison, L. J., & McLeod, S.** (2010). Risk and protective factors associated with speech and language impairment in a nationally representative sample of 4- to 5-year-old children. *Journal of Speech, Language & Hearing Research*, 53, 508-529.
- Hesketh, A.** (2004). Early literacy achievement of children with a history of speech problems. *International Journal of Language & Communication Disorders*, 39, 453-468.
- Highman, C., Hennessey, N., Sherwood, M., & Leitao, S.** (2008). Retrospective parent report of early vocal behaviours in children with suspected childhood apraxia of speech (sCAS). *Child Language Teaching & Therapy*, 24, 285-306.
- Hill, E. L.** (2001). Non-specific nature of specific language impairment: A review of the literature with regard to concomitant motor impairments. *International Journal of Language & Communication Disorders*, 36, 149-171.
- Hill, E. L., & Bishop, D. V. M.** (1998). A reaching test reveals weak hand preference in specific language impairment and developmental co-ordination disorder. *Laterality: Asymmetries of Body, Brain and Cognition*, 3, 295-310.

- Howard, S.** (2004). Connected speech processes in developmental speech impairment: Observations from an electropalatographic perspective. *Clinical Linguistics & Phonetics*, *18*, 405-417.
- Howard, S.** (2007). The interplay between articulation and prosody in children with impaired speech: Observations from electropalatographic and perceptual analysis. 4th international electropalatography (EPG) symposium, held in Edinburgh in september 2005. *Advances in Speech Language Pathology*, *9*, 20-35.
- James, D.** (2001). Use of phonological processes in Australian children ages 2 to 7 11 years. *Advances in Speech-Language Pathology*, *3*, 27.
- Jessup, B., Ward, E., Cahill, L., & Heating, D.** (2008). Prevalence of speech and/or language impairment in preparatory students in northern Tasmania. *International Journal of Speech-Language Pathology*, *10*, 364-377.
- Keating, D., Turrell, G., & Ozanne, A.** (2001). Childhood speech disorders: Reported prevalence, comorbidity and socioeconomic profile. *Journal of Paediatrics and Child Health*, *37*, 431-436.
- Kirkpatrick, E., & Ward, J.** (1984). Prevalence of articulation errors in new south wales primary school pupils. *Australian Journal of Human Communication Disorders*, *12*, 55-62.
- Larrivee, L. S., & Catts, H. W.** (1999). Early reading achievement in children with expressive phonological disorders. *American Journal of Speech-Language Pathology*, *8*, 118-28.
- Law, J., Boyle, J., Harris, F., Harkness, A., & Nye, C.** (2000). Prevalence and natural history of primary speech and language delay: Findings from a systematic review of the literature. *International Journal of Language & Communication Disorders*, *35*, 165-188.
- Law, J., Rush, R., Schoon, I., & Parsons, S.** (2009). Modeling developmental language difficulties from school entry into adulthood: Literacy, mental health, and employment outcomes. *Journal of Speech, Language & Hearing Research*, *52*, 1401-1416.
- Leitao, S., Fletcher, J., & Hogben, J.** (2000). Speech impairment and literacy difficulties: Underlying links. *The Australian Educational and Developmental Psychologist*, *17*, 63-75.

- Lewis, B. A., & Freebairn, L.** (1997). Subgrouping children with familial phonologic disorders. *Journal of Communication Disorders, 30*, 385.
- Lewis, B.A., Freebairn, L.A., Hansen, L.A., Stein, C.M., Shriberg, L.D., Iyengar, S.K., & Taylor, H.G.** (2006). Dimensions of early speech sound disorders. *Journal of Communication Disorders, 39*, 139--157.
- Lewis, B. A., Freebairn, L. A., Hansen, A. J., Miscimarra, L., Iyengar, S. K., & Taylor, H. G.** (2007). Speech and language skills of parents of children with speech sound disorders. *American Journal of Speech-Language Pathology, 16*, 108-118.
- Long, S. H., Fey, M. E., & Channell, R. W.** (2006). *Computerized profiling (MS-DOS version 9.7.0)*. . Cleveland, OH: Case Western Reserve University.:
- Majerus, S., Amand, P., Boniver, V., Demanez, J., Demanez, L., & Linden, M. V. D.** (2005). A quantitative and qualitative assessment of verbal short-term memory and phonological processing in 8-year-olds with a history of repetitive otitis media. *Journal of Communication Disorders, 38*, 473-498.
- McKinnon, D. H., McLeod, S., & Reilly, S.** (2007). The prevalence of stuttering, voice, and speech-sound disorders in primary school students in Australia. *Language, Speech & Hearing Services in Schools, 38*, 5-15.
- Motion, S., Northstone, K., Emond, A., Stucke, S., & Golding, J.** (2002). Early feeding problems in children with cerebral palsy: Weight and neurodevelopmental outcomes. *Developmental Medicine & Child Neurology, 44*, 40-43.
- Nathan, L., Stockhouse, J., Goulandris, N., & Snowling, M. J.** (2004). The development of early literacy skills among children with speech difficulties: A test of the "critical age hypothesis". *Journal of Speech, Language & Hearing Research, 47*, 377-391.
- Nittrouer, S., & Burton, L. T.** (2005). The role of early language experience in the development of speech perception and phonological processing abilities: Evidence from 5-year-olds with histories of otitis media with effusion and low socioeconomic status. *Journal of Communication Disorders, 38*, 29-63.

- Pagel Paden, E.** (1994). Otitis media and disordered phonologies: Some concerns and cautions. *Topics in Language Disorders, 14*, 72-83.
- Paradise, J.L., Dollaghan, C.A., Campbell, T.F., Feldman, H.M., Bernard, B.S.,...Smith, C.G.** (2005) Otitis media and tympanostomy tube insertion during the first three years of life: developmental outcomes at the age of four years. *Pediatrics, 112*, 265-277.
- Paradise, J. L., Feldman, H. M., Campbell, T. F., Dollaghan, C. A., Rockette, H. E., Pitcairn, D. L., . . . Pelham, W., J.** (2007). Tympanostomy tubes and development outcomes at 9 to 11 years of age. *New England Journal of Medicine, 356*, 248-261.
- Patel, R.R., Peters, T.J., Murphy, D.J. and the ALSPAC Study Team.** (2005) Prenatal risk factors for caesarean section. Analyses of the ALSPAC cohort of 12944 women in England. *International Journal of Epidemiology, 34*, 353-367.
- Peters, T.J.** (2008) Multifarious terminology: multivariable or multivariate? Univariable or univariate? *Paediatric and Perinatal Epidemiology, 22*, 506.
- Peterson, R. L., Pennington, B. F., Shriberg, L. D., & Boada, R.** (2009). What influences literacy outcome in children with speech sound disorder? *Journal of Speech, Language, and Hearing Research, 52*, 1175-1188.
- Raitano, N. A., Pennington, B. F., Tunick, R. A., Boada, R., & Shriberg, L. D.** (2004). Pre-literacy skills of children with speech sound disorders. *Journal of Child Psychology & Psychiatry, 45*, 821-835.
- Records, N. & Tomblin, J.B.** (1994) Clinical decision making: Describing the decision rules of practicing speech-language pathologists. *Journal of Speech and Hearing Research, 37*:144--156.
- Reilly, S., Wake, M., Bavin, E. L., Prior, M., Williams, J., Bretherton, L., . . . Ukoumunne, O. C.** (2007). Predicting language at 2 years of age: A prospective community study. *Pediatrics, 120*, e1441-9.
- Reilly, S., Wake, M., Ukoumunne, O. C., Bavin, E., Prior, M., Cini, E., Conway, L., Eadie, P., & Bretherton, L.** (2010) Predicting language outcomes at 4 years of age: Findings from Early Language in Victoria Study. *Pediatrics, 126*, e1530-1537.

- Rescorla, L.** (2002). Language and reading outcomes to age 9 in late-talking toddlers. *Journal of Speech, Language & Hearing Research, 45*, 360-371.
- Rice, M. L., Taylor, C. L., & Zubrick, S. R.** (2008). Language outcomes of 7-year-old children with or without a history of late language emergence at 24 months. *Journal of Speech, Language & Hearing Research, 51*, 394-407.
- Roberts, J. E., Rosenfeld, R. M., & Zeisel, S. A.** (2004a). Otitis media and speech and language: A meta-analysis of prospective studies. *Pediatrics, 113*, e238-48.
- Roberts, J., Hunter, L., Gravel, J., Rosenfeld, R., Berman, S., Haggard, M., . . . Wallace, I.** (2004b). Otitis media, hearing loss, and language learning: Controversies and current research. *Journal of Developmental and Behavioral Pediatrics, 25*, 110-122.
- Robinson, R. J.** (1991). Causes and associations of severe and persistent specific speech and language disorders in children. *Developmental Medicine & Child Neurology, 33*, 943-962.
- Roulstone, S., Law, J., Rush, R., Clegg, J., & Peters, T.J.** (2011) *Investigating the role of language in children's early educational outcomes*. Research Report DFE-RR134.
- Roulstone, S., Miller, L. L., Wren, Y., & Peters, T. J.** (2009). The natural history of speech impairment of 8-year-old children in the avon longitudinal study of parents and children: Error rates at 2 and 5 years. *International Journal of Speech Language Pathology, 11*, 391.
- Rust, J.** (1996). *Weschler Objective Language Dimensions Manual*. . London: The Psychological Corporation.
- Rvachew, S.** (2007). Phonological processing and reading in children with speech sound disorders. *American Journal of Speech-Language Pathology, 16*, 260-270.
- Shriberg, L. D.** (1993). Four new speech and prosody-voice measures for genetics research and other studies in developmental phonological disorders. *Journal of Speech and Hearing Research, 36*, 105-140.
- Shriberg, L. D., Austin, D., Lewis, B. A., McSweeney, J. L., & Wilson, D. L.** (1997a). The percentage of consonants correct (PCC) metric: Extensions and reliability data. *Journal of Speech, Language & Hearing Research, 40*, 708-722.

- Shriberg, L. D., Austin, D., Lewis, B. A., McSweeney, J. L., & Wilson, D. L.** (1997b). The Speech Disorders Classification System (SDCS): Extensions and lifespan reference data. *Journal of Speech, Language & Hearing Research, 40*, 723-740.
- Shriberg, L. D., Tomblin, J. B., & McSweeney, J. L.** (1999). Prevalence of speech delay in 6-year old children and comorbidity with language impairment. *Journal of Speech, Language & Hearing Research, 42*, 1461-1481.
- Smit, A. B.** (1993). Phonologic error distributions in the Iowa-Nebraska articulation norms project: Consonant singletons. *Journal of Speech and Hearing Research, 36*, 533-547.
- Smit, A. B.** (1993). Phonologic error distributions in the Iowa-Nebraska articulation norms project: Word-initial consonant clusters. *Journal of Speech and Hearing Research, 36*, 931-947.
- Stackhouse, J., & Wells, B.** (1997). *Children's speech and literacy difficulties: A psycholinguistic framework*. Chichester: Whurr.
- Stanton-Chapman, T. L., Chapman, D. A., Bainbridge, N. L., & Scott, K. G.** (2002). Identification of early risk factors for language impairment. *Research in Developmental Disabilities, 23*, 390-405.
- Sutherland, D., & Gillon, G. T.** (2007). Development of phonological representations and phonological awareness in children with speech impairment. *International Journal of Language & Communication Disorders, 42*, 229-250.
- Templin, M. C.** (1957). *Certain language skills in children; their development and interrelationships. (monograph series no.26)*. Minneapolis: University of Minnesota, The Institute of Child Welfare.
- Tomblin, J. B., Hardy, J. C., & Hein, H. A.** (1991). Predicting poor-communication status in preschool children using risk factors present at birth. *Journal of Speech & Hearing Research, 34*, 1096-1105.
- Tomblin, B., Smith, E., & Zhang, X.** (1997) Epidemiology of specific language impairment: Prenatal and perinatal risk factors. *Journal of Communication Disorders, 30*, 325-344.

- Tuomi, S., & Ivanoff, P.** (1977). Incidence of speech and hearing disorders among kindergarten and grade 1 children. *Special Education in Canada, 51*, 5-8.
- Visscher, C., Houwen, S., Moolenaar, B., Lyons, J., Scherder, E. J., & Hartman, E.** (2010). Motor proficiency of 6- to 9-year-old children with speech and language problems. *Developmental Medicine & Child Neurology, 52*, e254-8.
- Visscher, C., Houwen, S., Scherder, E., Moolenaar, B., & Hartman, E.** (2007). Motor profile of children with developmental speech and language disorders. *Pediatrics, 120*, e158-63.
- Webster, R. I., Majnemer, A., Platt, R. W., & Shevell, M. I.** (2005). Motor function at school age in children with a preschool diagnosis of developmental language impairment. *Journal of Pediatrics, 146*, 80-85.
- Winitz, H., & Darley, F. L.** (1980). Speech production. In F. M. Lassman, R. O. Fisch, D. L. Vetter & E. S. LaBenz (Eds.), *Early correlates of speech, language and hearing* (pp. 232-265). Littleton, MA: PSG Publishing.
- Wolke, D., & Meyer, R.** (1999). Cognitive status, language attainment, and prereading skills of 6-year-old very preterm children and their peers: The bavarian longitudinal study. *Developmental Medicine & Child Neurology, 41*, 94-109.
- Wren, Y. E., McLeod, S. M., White, P., & Miller, L. L., Roulstone, S.E.** (2013). Speech Characteristics of 8-year-old children with atypical speech: Findings from a prospective population study. *Journal of Communication Disorders, 46*, 53-69.
- Wren, Y. E., Roulstone, S. E., & Miller, L. L.** (2012). Distinguishing groups of children with persistent speech disorder: Findings from a prospective population study. *Logopedics, Phoniatrics and Vocology, 37*, 1-10.
- Yliherva, A., Olsén, P., Mäki-Torkko, E., Koironen, M., & Järvelin, M.R.** (2001). Linguistic and motor abilities of low-birthweight children as assessed by parents and teachers at 8 years of age. *Acta Paediatrica (Oslo, Norway: 1992), 90*, 1440-1449.
- Zubrick, S. R., Taylor, C. L., Rice, M. L., & Slegers, D. W.** (2007). Late language emergence at 24 months: An epidemiological study of prevalence, predictors, and covariates. *Journal of Speech, Language, and Hearing Research, 50*, 1562-1592.