

# **Interventions for Pre-school Children with Co-occurring Phonological Speech Sound Disorder and Expressive Language Difficulties: A Scoping Review**

## Abstract

*Background:* Recent evidence suggests that pre-school children with co-occurring phonological speech sound disorder and expressive language difficulties are at a higher risk of ongoing communication and literacy needs in comparison to children with these difficulties in isolation. However, to date there has been no systematic or scoping review of the literature specific to interventions for children with this dual profile.

*Aims:* To explore evidence regarding interventions for pre-school children with co-occurring phonological speech sound disorder and expressive language difficulties, including the content/delivery of such interventions, areas of speech and language targeted, and a broad overview of study quality.

*Methods:* This study used a scoping review methodology in accordance with the guidance from the Joanna Briggs Institute. Following a systematic search of Ovid Medline, Ovid Emcare, OVID Embase, CINAHL, Psychinfo and ERIC, 11 studies were included in the review. A researcher developed data extraction form was used to extract specific information about each intervention, with the JBI appraisal tools used to provide a broad overview of the quality of each study.

*Main contribution:* Included papers consisted of six RCTs, two cohort studies, two case studies, and one case series. Interventions fell into two main categories; 1) integrated interventions which combined content for both speech and language targets, and/or explicitly used the same type of technique to improve both domains, and 2) single domain interventions

which explicitly included content to target speech or language only, but also aimed to improve the other domain indirectly. Study quality varied, with detail on the content, context and delivery of interventions often underspecified, hampering replication and clinical applicability of findings.

*Conclusion:* Early emerging evidence was identified to support both integrated speech and language interventions as well as single domain interventions. However, caution should be exercised due to the variation in the quality and level of detail reported for the interventions. Future intervention studies may seek to address this by reporting in accordance with TIDieR reporting guidelines. This approach would enable clinicians to consider the applicability of the intervention to individual children within differing settings.

### **What is already known on the subject**

Pre-school children with co-occurring phonological speech sound disorder (SSD) and expressive language difficulties frequently present within speech and language therapy services. These children are at a higher risk of long-term communication and literacy difficulties compared to children with these needs in isolation. Some emerging evidence suggests that interventions for children with this co-occurring profile may exist within the literature, however this evidence may not be known to clinicians in everyday practice.

### **What this study adds**

This review is the first to systematically examine evidence of interventions for pre-school children with co-occurring phonological speech sound disorder and expressive language difficulties. The review identified a small number of intervention studies which varied in

research quality and level of detail provided regarding the content and delivery of interventions.

### **Clinical implications of this study**

The findings of this study highlight published evidence for interventions for pre-school children with co-occurring phonological speech sound disorder and expressive language difficulties. These may take the form of integrating techniques for speech/language into a single intervention, or the explicit targeting of one domain with the aim of also influencing the other. However, there is a need for further high-quality research in this area. Such studies should provide sufficient detail to enable replication. This would enable clinicians to understand the relevance and applicability of such intervention findings to the individual children they see within their clinical practice.

Interventions for Pre-school Children with Co-occurring Phonological Speech Sound Disorder and Expressive Language Difficulties: A Scoping Review

Introduction

Children accessing pre-school speech and language therapy services may present with features of Developmental Language Disorder that are unlikely to resolve by the age of 5 years (Ebbels, 2020). These features, which are not associated with biomedical conditions with a known impact on communication, such as Autism (Bishop et al., 2017), may include difficulties with verbal learning/memory, discourse, pragmatics, phonology, word finding, semantics, morphology and/or syntax (for an overview see RCSLT, 2020). As such pre-schoolers may not receive a formal diagnosis of Developmental Language Disorder until their school years, the term ‘Features of Developmental Language disorder’ (FDLD) offers a useful way to characterise this important group. Speech Sound Disorder (SSD) is also heterogenous in nature, with pre-school children with SSD presenting with difficulties relating to phonology and/or motor execution (Dodd, 2014). It is understood that although FDL and SSD may present in isolation within the pre-school population, co-occurrence of the disorders also exists. Evidence to suggest that pre-school children with this dual profile are present within clinical caseloads was initially reported by Broomfield and Dodd (2004), who identified that from a total of 320 children with a primary diagnosis of speech difficulties (85% aged 6 years or below), 25.6% had co-occurring language comprehension difficulties, 38.1% had co-occurring expressive language difficulties and 50.9% had difficulties with their vocabulary development. The co-occurrence of speech and language difficulties in pre-school children was also later demonstrated in the Early Language in Victoria Study (ELVS), where 3.4% of 1494 children met the criteria for SSD. Of this group,

40.8% also had co-occurring language difficulties (Eadie et al., 2014). Such findings suggest a marked overlap exists between SSD and FDL within the pre-school population.

This overlap is perhaps unsurprising given that phonological deficits can be a feature of both groups (Shriberg and Kwiatkowski, 1994; RCSLT, 2020), and evidence suggests that multi-faceted and bi-directional links exist between phonological and lexical development, particularly in preschool children (Petinou et al., 2021; Stoel-Gammon, 2011). The complexity of these links are reflected in studies regarding the clinical trajectory for pre-school children with this co-occurring profile. Children with persistent SSD at 8 years are more likely to have had reported language difficulties at 2 and 3 years (Wren et al., 2016), and also 2 and 5 years (Roulstone et al., 2009). Children with reported language difficulties at 2 and 5 years are also more likely to demonstrate co-occurring speech difficulties at these same ages (Roulstone et al., 2009). Most recently, Neam et al. (2019) reported that children with a history of late talking are more likely to present with speech production difficulties at 4-5 years when compared to children who had previously demonstrated typically developing language. The heterogenous nature of co-occurring FDL and SSD is an additional complexity when considering these trajectories.

Although children with receptive language difficulties may also present with a co-occurring SSD, research to date highlights a particular connection between SSD and expressive FDL (eFDL). Eadie and colleagues (2014) reported that 36.7% of children with SSD presented with eFDL, in contrast to 20% of children with SSD presenting with receptive FDL.

A compromised linguistic system has been associated with SSD and may influence its presentation in children with co-occurring FDL. For example, children who have both SSD and eFDL may demonstrate increased linguistic (i.e., phonological) based speech error

patterns, such as increased sound omissions (Macrae and Tyler, 2014). Phonology based SSD (pSSD) is the most common subtype of SSD presenting within clinical services (McLeod and Baker, 2017), and has been linked with certain aspects of eFDLD, for example syntax and morphology (McLeod et al., 2017; Mortimer and Rvachew, 2010). The complex relationship between pSSD and eFDLD was most recently highlighted by Howland et al. (2019), who found that for pre-schoolers with phonological impairment, realisation of grammatical morphemes was influenced by phonological factors.

### *Long term risks*

Children presenting with pSSD or eFDLD in isolation may be at risk of long-term needs relating to their mental well-being, literacy skills, and educational attainment (Johnson, Beitchman and Brownlie, 2010; McCormack et al., 2010; St Clair et al., 2019). Emerging evidence suggests that pre-school children who present with a co-occurring pSSD and eFDLD profile may be at even greater risk of ongoing communication and literacy needs compared to children who have these difficulties in isolation (Hoover, 2019). In their longitudinal study Lewis et al. (2015) reported that children with co-occurring speech and language difficulties in early childhood had poorer language and literacy outcomes at adolescence in comparison to those with speech difficulties only. A heightened risk of literacy difficulties has also been described by Hayiou-Thomas et al. (2017), who reported that children presenting with co-occurring difficulties at 3;6 years were more likely to present with persistent literacy needs (e.g. difficulties with reading comprehension and phoneme awareness) at 8 years, in comparison to pre-schoolers with speech difficulties only. Given that children identified with co-occurring speech and language needs in the pre-school years may be more likely to face such long-term challenges, consideration of effective early

intervention to ameliorate the lasting impact of such difficulties is a priority area of attention for clinicians and researchers working in early years (Burgoyne et al., 2019).

### *Current practice*

Current clinical practice relating to early intervention for children with this co-occurring profile, and how this may relate to the evidence base, is unclear. Roulstone et al. (2015) investigated interventions implemented by SLTs for pre-school children with primary communication difficulties, including pSSD and eFDLD, as a part of the ‘Child Talk’ study. The research engaged in a detailed and extensive mapping of individual therapy components onto a typology of nine themes, based on clinicians’ current practice. The findings suggested that SLTs may not consider interventions specific to co-occurring eFDLD and pSSD. For example, Roulstone et al. (2015) reported positive evidence for the use of broad target recasts, where pSSD and eFDLD are targeted simultaneously by combining speech recasts with language recasts (Yoder, Camarata and Gardner, 2005), however participant SLTs did not report knowledge of this technique. Additionally, clinical review commentaries pertaining to interventions for this group are also evident within the literature (Hoover et al., 2019; Tyler, 2002; Tyler et al., 2016). Therefore, it is unclear whether extensive evidence of interventions is lacking for children with this dual profile, or if evidence does exist but is not known to clinicians. Interestingly, some researchers have reported on language-based approaches which also aim to improve speech (Tyler, 2002). However, it is not clear whether clinicians may target one domain with the expectation of improving both.

A further key finding from Child Talk was that clinicians often ‘mix and match’ different intervention components according to a child’s unique clinical profile, including those with co-occurring needs. At the time of writing, it is unknown how much evidence supports the combining of intervention components for children with co-occurring pSSD and eFDLD

(Roulstone, 2015). While clinician expertise is a key component of the evidence-based practice model, with SLTs modifying their interventions based on a child's unique needs, to date little is known about which 'active ingredients' of intervention may affect change within this population. Identifying the content of such interventions, and how this content is delivered, is the first step in identifying these ingredients. Although children with co-occurring pSSD and eFDLD are routinely seen within clinical practice, it seems that the evidence base for intervention for children with this dual profile is limited and unclear. Therefore a methodological review of the current literature would confirm these assumptions and provide more detailed information.

### *Objectives*

The overarching objective of this scoping review was to identify and describe published evidence for interventions that target both phonological SSD (pSSD) and expressive language difficulties (eFDLD). In particular, we examined the content, context and mode of delivery and undertook a broad evaluation of research quality.

### Methods

Scoping reviews may be conducted when a broad exploration of the literature is needed but the extent of current evidence on a topic unclear (Munn et al., 2018). This contrasts with systematic reviews which are narrower in focus (Peters et al., 2020). Therefore, a scoping review methodology best aligns with objectives of this study.

Although broad in nature, it is essential that a scoping review, like a systematic review, follows an explicit systematic process for literature searching and analysis. The current



scoping review has been conducted following detailed guidance from the JBI manual for evidence synthesis, chapter 11 (Peters et al., 2020).

The study was registered with the Open Science Framework (OSF) in January 2021 and can be found at <https://osf.io/u6adf>

Full details of the protocol are available through an open access journal (Rodgers et al., 2021).

### *Eligibility criteria*

The participants within included papers were required to have profiles consistent with pSSD and eFDLD as indicated by diagnostic testing and/or the intervention targets set. As the review focussed specifically on interventions within the pre-school years, 80% of children within the included papers had to be aged 2;0- 5;11 years. This is also in line with the criteria set with the ‘Child Talk’ study (Roulstone et al., 2015). Studies were excluded if they involved children whose communication needs were associated with a condition with a known impact on speech and/or language, such as Autism and Cleft Lip and Palate.

To be included in the review, the paper had to be written in English, although research conducted in languages other than English were not excluded if they met eligibility criteria. Each paper also had to report on primary, empirical research whereby change or exploration of change, in both pSSD and eFDLD, was clearly stated within the intervention aims. Papers including interventions which target other areas of speech and language, such as receptive language, were not excluded if they also targeted both pSSD and eFDLD. To avoid excluding any potentially relevant articles, a minimum publication date was not stated.

### *Information Sources*

This review did not exclude papers on account of study design, in order to provide a broad overview of interventions in the area. However, to locate papers of a minimum quality which had been subject to peer review, grey literature was excluded. This included non-peer reviewed articles, book chapters, thesis and conference abstracts. The completed search was conducted on 09/03/2021 and included CINAHL, Ovid Emcare, Ovid Embase, Ovid Medline, PsychInfo and ERIC. Where a potentially relevant article could not be retrieved, direct contact with the study authors was made. The reference lists of all included sources of evidence were screened for additional studies, as were the references from relevant non-systematic review and clinical commentary papers (Hoover, 2019; Tyler, 2002; Tyler, 2016).

### *Search*

A three-step search strategy was used in accordance with the JBI guidance (Peters et al., 2020). Initially, a set of key terms were drafted by the authors based on terminology found within relevant literature. These key terms were then refined following feedback from two independent subject experts with significant post-doctoral experience in the area; these can be found in appendix one. Following this an initial limited search Ovid Emcare and Ovid Medline was conducted. With the support of a clinical librarian, keywords and index terms were adapted and selected for each database as appropriate within the final search. Further details of development of the initial search strategy can be found in the protocol (Rodgers et al., 2021). The final search strategy for Ovid Emcare and Ovid Medline can be found in appendix two.

### *Selection of sources of evidence*

Following the database search 25,132 papers were found. All identified citations were collated and uploaded into Endnote and duplicates removed, leaving a remaining 20,385

papers. Once papers that were clearly unrelated to the aims of the study were removed, 611 papers were left for appraisal at abstract level. Two reviewers independently reviewed 10% of the abstracts against the inclusion criteria. As the level of agreement between the two reviewers was above 90% for the first 10%, one reviewer then reviewed the remaining abstracts. Once all abstracts had been reviewed, 18 potentially relevant sources for full text review were then retrieved with their citation imported into the Rayaan reference management database (Ouzzani et al., 2016). The two reviewers then examined these papers independently at full text level with regular consensus meetings, resulting in the final inclusion of nine papers from the database search. Following the additional screening of the included papers and clinical commentaries/non-systematic literature reviews, two out of five additional papers were found to be eligible, with a total of 11 papers going forward to data extraction phase.

#### *Data charting process and data items*

Data was extracted from the papers using a researcher-developed extraction form, which was developed as outlined within the study protocol (Rodgers et al., 2021). Key information extracted using the form (appendix 3) included participant age, aspects of speech and language targeted, and intervention content and delivery (including dosage). This information is needed to facilitate the application of such interventions within clinical practice, and is included within the Template for Intervention Description and Replication (TIDieR) guidelines (Hoffman et al., 2014).

The extraction form was updated in an iterative process. The two reviewers extracted data from each paper independently using an electronic copy of the extraction form and resolved inconsistencies via regular consensus meetings.

### *Critical appraisal of individual sources of evidence*

A broad overview of the quality of identified studies was also carried out. Although this is not essential when conducting scoping reviews quality appraisal may be undertaken if it aligns with the review aims (Peters et al., 2020). For this review, an overview of study quality was deemed as helpful to framing the clinical and research implications of the included studies. Quality review can also provide indicative evidence as to whether a subsequent systematic review on the efficacy of included papers is justifiable. Therefore, the methodology within individual papers was appraised using the corresponding tool from the Joanna Briggs Institute (2021) critical appraisal tools collection. These tools are a collection of separate checklists developed to appraise studies of differing designs, including RCTs, cohort studies and case reports. Examples of individualisation according to study design include ratings for randomisation within the RCT checklist, and ratings for case history detail within the case report checklist. Each study was appraised by two reviewers independently, using the associated study design checklist, and achieved final consensus via regular consensus meetings.

### Synthesis of results

Studies were grouped according to outcome measures used, intervention content/delivery and areas of speech and language targeted. To keep data reporting concise, the summary of findings includes only details which are specific to the purpose of this review (i.e. description of intervention content/delivery, areas targeted and measures used).

### Results

### ***Figure 1: PRISMA flowchart***

The results of the literature search, including the numbers of citations screened, duplicates removed, full text studies assessed and reasons for exclusion at full text level, are presented in the PRISMA flow diagram (Figure 1).

#### *Characteristics of sources of evidence*

One of the articles reported on Portuguese speaking participants (Lourenco et al., 2020), with the other 10 involving English speaking participants. Six articles were RCTs (Fey et al., 1994; Tyler & Watterson 1991; Tyler et al 2002; Tyler et al 2003; Tyler et al 2011; Yoder et al 2005), two were case reports (Combiths et al., 2019; Hoffman et al., 1996), two were cohort studies (Lourenco et al., 2020; Tyler & Sandoval 1994), and one was described as a case series (Bellon-Harn et al., 2004). Four of the six RCTs did not contain a control group but used randomised allocation into two experimental groups (Tyler & Watterson 1991; Tyler et al 2002; Tyler et al 2003; Tyler et al 2011).

A summary of ratings from the individual quality appraisals is given in table 1. The percentage score is derived from the number of quality standards met by each paper from its associated checklist. Full ratings for each individual study can be found within appendix 4.

#### ***Table 1: Overview of included studies***

For the six RCTs the blinding of participants and intervention deliverers was not always carried out. The authors reported that this was due to ethical reasoning or the nature of the intervention, for example when parents expressed a preference for a “wait and see” approach

(Tyler et al., 2002). Concerns of note included a lack of detail on whether outcome assessors were blinded to group allocation and how participants were randomised into groups. A common theme amongst included studies was missing information regarding participant demographics such as participant gender, and unclear reporting of intervention content/delivery. Further details can be found in table 1.

The intervention comparators varied between studies, however this was not unexpected due to the differing study designs. Two studies (one RCT and one cohort) contained control groups who were free to receive support elsewhere (Lourenco et al, 2020; Yoder et al., 2005). The other cohort study (Tyler & Sandoval 1994) used pre and post intervention measures, and mid intervention generalisation probes. Three of the RCT studies included a delayed or no treatment control (Fey et al., 1994; Tyler et al., 2002; Tyler et al., 2003), and two of the RCTs used an alternative experimental group as a comparator (Tyler et al., 2011; Tyler & Watterson 1991). In one RCT support accessed by the control group, in terms of number of hours, was included as a variable within statistical analysis however the content of this was unspecified (Yoder et al., 2005). The case series by Bellon-Harn et al. (2004) used pre, mid and post treatment to measure change. The remaining case study used pre and post intervention measures and while a stable baseline was reported this was only evaluated across two timepoints (Combiths et al., 2019).

An overview of the included papers can be found in table 1. The studies involved interventions which fell into two broad categories:

- 1) INTEGRATED INTERVENTIONS (Bellon-Harn et al., 2004; Hoffman et al., 1996; Yoder et al., 2005). Integrated interventions explicitly targeted aspects of both speech and

language. Three intervention studies, which exclusively included integrated interventions, outlined how this might be done by alternating between different speech/language techniques within the same session or period of intervention, and/or by using the same technique to target both speech and language concurrently.

2) SINGLE DOMAIN INTERVENTIONS (Combiths et al., 2019; Fey et al., 1994; Lourenco et al., 2020; Tyler et al., 2002; Tyler & Watterson 1991). Although these interventions explicitly targeted speech or language, the studies had a stated aim of exploring the impact of the intervention on the other domain (e.g. the impact of speech intervention on language), with outcomes for both speech and language being assessed both before and after the intervention took place.

In addition to studies which involved either single domain or integrated interventions, three further studies compared both (Tyler et al., 2011; Tyler et al., 2003; Tyler & Sandoval 1994). One of these studies investigated the effectiveness of carrying out an intervention which targets speech before language and vice versa, in comparison to an integrated intervention which combined content for speech and language (morphosyntax) within the same session (Tyler et al., 2003). A further study compared a single domain speech only intervention to an integrated intervention, which alternated between speech and language (morphosyntax) on a weekly basis (Tyler et al., 2011). The remaining study investigated the impact of two single domain interventions for speech and language (morphosyntax) verses an integrated intervention which combined intervention content for both within individual sessions (Tyler and Sandoval, 1994).

*Population:*

Only studies which involved children with primary speech, language and communication needs were included, with all included studies stating that participants' communication needs had no known cause or associated developmental condition. These children also had co-occurring speech and language difficulties, as indicated by their intervention targets and/or pre-intervention assessments. The age range of children within included studies was 3;0 to 5;11 years. In addition to the assessment of speech production and expressive language, eight studies reported the receptive language ability of participants using results from standardised assessments. Bellon-Harn et al (2004) required participants to be at least 1.5 standard deviations below the mean for receptive language on the Clinical Evaluation of Language Fundamentals- third edition. In contrast to this, Tyler et al (2011) excluded participants who were more than 1.5 standard deviations below the mean for receptive vocabulary on the Peabody Picture Vocabulary Test-third edition. Six studies assessed receptive language skills and did not exclude children based on receptive language ability (Lourenco et al., 2019; Tyler et al., 2002; Tyler et al., 2003; Tyler and Sandoval 1994; Tyler and Watterson 1992; Yoder et al., 2005). Three studies did not refer to the receptive language skills of included participants (Combiths et al., 2019; Fey et al., 1994; Hoffman et al., 1996).

*Concept:*

As specified in the study protocol (Rodgers et al., 2021), the 'concept' of this review was the intervention content and delivery, outcome measures, and areas of speech and language targeted within included papers. For papers reporting on integrated interventions, content tended to be naturalistic (i.e., play based and non-directive) in nature, with adult scaffolding techniques playing a key role. Scaffolding techniques included broad target recasts (Yoder et al., 2005), cloze procedures (Bellon-Harn et al., 2003; Hoffman et al., 1996), and general modelling techniques such as providing contrast words (Bellon-Harn et al., 2003) and labels



(Hoffman et al., 1996). The intervention content for the remaining eight single-domain and integrated/cross domain papers also consisted of some adult scaffolding techniques to varying degrees. Common intervention techniques described within these eight papers, specific to the targeting of speech, included the use of minimal pairs (Tyler and Sandoval, 1994; Tyler and Waterson 1991), and drilling (Combiths et al., 2019; Tyler et al., 2003; Tyler et al., 2002). Focused stimulation was a common intervention technique targeting language (Tyler et al., 2002; Tyler and Sandoval, 1994). However, many of the studies also outlined how the same type of technique might be used to address both speech and language, such as auditory awareness used to heighten children's awareness of sounds and/or grammatical forms (Tyler et al., 2002; Tyler et al., 2003; Tyler et al., 2011), drilling (Fey et al., 1994), focused stimulation (Fey et al., 1994; Tyler et al., 2003), and narrative/story retell (Lourenco et al., 2019; Tyler and Sandoval 1994).

Details on intervention dosage varied across papers; ten reported on the length and number of intervention sessions with three providing further detail on the dosage of the individual content of these sessions (e.g., number of child imitations aimed for, or number of adult recasts given) (Tyler et al., 2002; Tyler et al., 2003; Yoder et al., 2005). Where this detail was available, frequency of dosage ranged from 3 weekly sessions over 4 weeks (Bellon-Harn et al., 2003) to twice weekly sessions over 2 sets of 12-week blocks (Tyler et al., 2002).

Speech targets predominantly focused on developing a broader range of speech sounds, with one study also targeting speech intelligibility, with the percentage of intelligible speech calculated from a language sample (Yoder et al., 2005). Developing a broader range of sounds was commonly measured through Percentage Consonants Correct (PCC) (Bellon-Harn et al., 2003; Combiths et al., 2019; Fey et al., 1994; Hoffman et al., 1996; Lourenco et

al., 2020; Tyler et al., 2011; Tyler and Watterson 1991) and/or by conducting individual phoneme production probes elicited using pictures before, during and after an intervention (Combiths et al., 2019; Tyler and Sandoval 1994). Some studies included additional calculations, such as the Target Generalisation Composite (TGC), which is a percentage derived from the number of times a child accurately generalised the target sounds divided by the total number of opportunities to use the sound (Tyler et al., 2003).

With regard to studies targeting expressive language, ten of the eleven studies focused on development of sentence length with a particular emphasis on morpheme use. The most common measure for this was Mean Length of Utterance in morphemes (MLUm) (Combiths et al., 2020; Hoffman et al., 1996; Tyler et al., 2002; Tyler et al., 2011; Tyler and Sandoval 1994; Tyler and Watterson 1991; Yoder et al., 2005). Alternative and additional measures of expressive language included Mean Length of Utterance in words (MLUw) (Lourenco et al., 2020), the Developmental Sentence Score (DSS) (Fey et al., 1994), Finite Morpheme Composite (FMC) (Tyler et al., 2003; Tyler et al., 2011), and the results/scores of individual morpheme and/or morphophonological probes elicited before, during, and/or after the intervention (Combiths et al., 2019; Tyler and Sandoval 1994). One study focused on developing semantic complexity levels with pre and post intervention probes (Bellon-Harn et al., 2003).

*Context:*

One study took place in a university clinic (Lourenco et al., 2020), one in an educational setting (Combiths et al., 2019), and one in a “small therapy room” (Yoder et al., 2005). The location of the intervention was not explicitly stated in the remainder of the studies. All interventions, except one, were delivered by a trained professional in psychology or speech

and language therapy. A parent-delivered intervention, overseen by a clinician, was compared to a fully clinician delivered intervention within one study (Fey et al., 1994).

*Reported results of individual sources of evidence:*

*Integrated intervention studies:* Positive change in speech production, measured by PCC, and expressive language (MLUm and semantic complexity levels) was reported in two of the integrated studies that used baseline measures as a comparator (Bellon-Harn et al., 2003; Hoffman et al., 1996). Yoder et al (2005) reported improved expressive language (MLUm) and speech production (intelligibility) by children who had relatively low pre-treatment speech accuracy as a result of an integrated intervention, when compared to a control group.

*Single domain intervention studies:* Tyler et al (2002) reported that a speech only intervention led to improvement in just speech, and not speech and expressive language, when compared to a control group within their RCT. In contrast to this, Tyler and Watterson (1991) reported positive cross domain effects from speech to language in a speech only intervention based on participants' pre and post treatment assessment. One case study which focused on speech production reported generalised learning to untreated consonants and clusters across word positions but no generalisation to language (morpheme production) (Combiths et al., 2019).

Language only interventions were reported to impact on both speech production and language in two RCT studies (Fey et al., 1994; Tyler et al., 2002). In contrast to this, Lourenco and colleagues (2020) cohort study reported that narrative intervention had a less marked impact on speech. This cohort study included a control group which could access non-intervention specific speech and language therapy support, however the nature of this intervention is not

stated within the paper. Tyler and Waterson (1991) reported finding a regression in speech when employing a language only intervention in their RCT. The authors speculate that this finding may have been a consequence of differences in the linguistic profiles between experimental groups.

*Studies which included both integrated and single domain interventions:*

An early study reported that an integrated intervention resulted in greater improvements in both speech and expressive language (morphosyntax) when compared to speech or morphosyntax only interventions (i.e., single domain) (Tyler and Sandoval, 1994). In their study which compared the effectiveness of a single domain speech intervention with an integrated intervention, Tyler et al. (2011) reported finding that children receiving both interventions made significant gains in all measures (including both MLUm and PCC), aside from in the FMC which approached significance. Tyler et al (2003) focused on the impact of targeting speech and language (morphosyntax) in differing orders, by comparing 4 experimental intervention groups to each other and a control: 1) alternated speech and morphosyntactic intervention content on a weekly basis, 2) integrated speech and morphosyntactic intervention content within the same sessions, 3) a 12 week block of morphosyntax only intervention followed by a further 12 week block of speech only intervention, and 4) a 12 week block of speech only intervention by a further 12 week block of morphosyntax only intervention. The control was a no treatment group. The authors reported finding that morphosyntactic change was greatest for children in group 1. They reported no significant differences between the groups in terms of speech change, however when compared to the control group they found that groups 1, 2 and 3 made the best gains.

Discussion

The aim of this scoping review was to explore the population, concept and context of interventions which aim to improve both pSSD and eFDLD in pre-school children with this co-occurring presentation. In addition, a broad overview of study quality has been reported. Of the 11 papers eligible for inclusion within this review, three integrated speech/language content within interventions and five included interventions with content for either speech or language (i.e. single domain). An additional three papers outlined studies which included both integrated and single domain interventions. All were peer reviewed as stated within the inclusion criteria, but variability amongst the quality and level of detail of each study was noted.

#### *Clinical profiles of participants*

Included studies involved children with co-occurring speech and expressive language difficulties. However, variability was noted regarding the inclusion and reporting of children with differing clinical characteristics within this dual profile. For children presenting with both pSSD and eFDLD, the inclusion/exclusion of children with receptive language difficulties is a particularly important consideration, given that it could be a potential confound when drawing conclusions from individual response to therapy, and three studies did not report on this. For clinicians to determine which study findings have relevance to children within their everyday practice, it is important that such information is reported. Children with pSSD and eFDLD are a heterogenous group, and as such information on the wider clinical profile of participants would enable researchers and practitioners to develop theories about why children with varying presentations may respond differently to the same intervention.

### *Linking to practice: intervention content*

It is interesting to note that the individual content of interventions described in the included papers are present within the wider evidence base for intervention studies which focus on either speech or language. Within the current paper we have used the term ‘technique’ as a general term, which encompasses both named techniques such as auditory bombardment and strategies such as recasting. Techniques within the included papers, which present in the wider evidence base for speech or language intervention studies, include auditory bombardment for speech (Bowen and Cupples, 1999), and focused stimulation for language (Bruinsma et al., 2020). They also include adult scaffolding within naturalistic contexts (e.g., cloze procedures) for speech intervention, (Camarata, 1993) and for language intervention (Falkus et al., 2016). The noted exception is the technique of broad target recasts (Yoder et al., 2005), which are exclusively used to target both speech and language concurrently, rather than speech or language in isolation (Camarata et al., 2006). The use of the same type of technique to address multiple areas is in keeping with reports on current practice (Roulstone et al., 2015). However, specific details of techniques, including dosage and whether they are being used to target speech, language, or both together, was limited within the included papers within this review. This lack of detailed reporting acts as a barrier to judging the effectiveness of these combinations in regard to targeting both speech and language within a single intervention or episode of therapy.

### *Linking to practice: intervention delivery*

Internationally it is becoming more widely recognised that significant others, including parents and education staff, play an important role with supporting a child’s communication needs (Klatte et al., 2020). As such, interventions are increasingly evolving to include

training for these significant others in delivering some aspects of the intervention within the child's everyday environment (Hegarty et al., 2021; Heidlage et al., 2020). Such training may enable significant others to collaborate with clinicians to enable children to work towards their goals (Sugden et al., 2020). In the majority of included studies, the intervention content was delivered by either a trained professional in psychology or speech and language therapy. Additionally, the dosage of intervention sessions often spanned several months, with multiple sessions a week. For clinicians working within public services, it may not be possible to deliver such interventions with the professional as the sole deliverer and at the stated dosage, as they are often required to follow predefined treatment pathways (Roulstone et al., 2015). Additionally, emerging evidence suggests that increased dosage may not always result in greater outcomes, and that less frequent, longer sessions as well as more frequent, shorter sessions, may be most beneficial for language change (Frizelle et al., 2021). In order to address this challenge for children with co-occurring pSSD and eFDLD, future research may benefit from being tailored to typical service provisions and may focus on how to best provide effective training to significant others working with children within their everyday environments. Such research is becoming increasingly evident within intervention studies which focus solely on SSD (Sugden et al., 2020) or FDL (Roberts et al., 2019).

### *Outcomes*

Although the described presentation of both pSSD and eFDLD may be considered more specific than 'SSD' and 'DL', children with this co-occurring pSSD and eFDLD nevertheless constitute a heterogeneous group. Therefore, interventions targeting both pSSD and eFDLD may address differing aspects of these difficulties. For language, most studies included within this review focused on morphology and syntax. This is perhaps unsurprising given the established link between these aspects of language development and pSSD (McLeod

et al., 2017; Mortimer and Rvachew, 2010). With the exception of one study, which targeted speech intelligibility (Yoder et al., 2005) the impact of each intervention was not measured in terms of everyday functioning (i.e., the impact of the child's pSSD and eFDLD on daily life). Although the measuring of outcomes relating directly to speech and language skill are important for addressing whether an intervention 'works', it is recognised that addressing the impact on the child's participation in daily life is as valuable, with both parents and children expressing appreciation for intervention considering functional outcomes and personal experiences (Roulstone, 2015). This stance is reflected within recent intervention studies which target either speech or language, where functional outcome measures are used alongside measures of communication impairment (Cunningham et al., 2019; Sugden et al., 2020). Future intervention research for children with co-occurring pSDD and eFDLD could seek to include such broader outcome domains in addition to improving speech/language skill. Such an approach would ensure that interventions have a meaningful impact on the lives of whom they aim to support.

### *Limitations*

A scoping review, in comparison with a systematic review, does not give an in-depth evaluation of intervention effectiveness due to the diversity of the interventions and measures within included studies. In the present scoping review, while the synthesis of reported study findings is informative caution needs to be applied to the reported findings of individual papers due to varying study quality. The JBI tools were fit for the purpose of providing a broad overview of study quality. However, a future systematic review aiming to investigate the effectiveness of interventions for this clinical group could include the application of additional rigorous tools for evaluation of bias, such as the Cochrane risk of bias (Higgins et al., 2011). This would complement the quality appraisal.



### *Conclusion and recommendations*

Building on the reported findings from previous clinical reviews/commentaries on interventions for young children with co-occurring pSSD and eFDLD (Hoover et al., 2019; Tyler et al., 2002; Tyler et al., 2016), the current scoping review offers additional recommendations for research studies going forward. This guidance is in line for with general best practice within intervention research, and below has been tailored towards our population of interest.

Recommendations include:

- Providing sufficient detail to replicate findings, using the TIDieR guidelines (Hoffmann et al., 2014). This should include details, wherever possible, on dosage within intervention activities, specific equipment used and a clearly described context for the intervention delivery. As intervention studies for this clinical group may involve combining techniques typically used for speech and/or language, such detail would facilitate understanding about which combination of techniques work best for which children, and in what context.
- Providing sufficient detail about the control group/pre-intervention baseline and experimental groups. This should include information regarding the key speech and language characteristics of study participants. This would ensure that there is a detailed comparator which accounts for maturation. Additionally, this detail would support conclusions to be drawn on what interventions work best for children with this co-occurring profile, which is heterogenous in nature and may vary according to additional difficulties (i.e., with receptive language).

In summary, this scoping review draws together knowledge on interventions for pre-school children with pSSD and eFDLD by identifying relevant studies from a scattered literature and providing a summary of the intervention content, areas of speech and language targeted, and an overview of the quality of current evidence. This knowledge provides a foundation for developing the evidence base for this much under-researched clinical group. Although there is much more work to be done within this field, inspiration may be taken from well-established interventions for pSSD or eFDLD.

#### Data availability statement

The data that supports the findings of this study are available in the supplementary material of this article

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