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Interventions for Pre-School Children with Co-Occurring Phonological Speech Sound Disorder and Expressive Language Difficulties: A Scoping Review Protocol

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ABSTRACT

Background: Pre-school children with co-occurring phonological speech sound disorder and expressive language difficulties are more likely to have ongoing communication and literacy needs compared 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 this frequently seen and high-risk group.

Aims: The objective of this paper is to provide a rigorous and detailed protocol for a scoping review of interventions, which target both phonological speech sound disorder and expressive language difficulties in pre-school children with primary speech, language and communication needs. The protocol includes details on the development of a search strategy, as well as the trial of an extraction tool.

Methods/Design: Included studies must aim to concurrently improve both speech production and expressive language. Children within included studies must be aged between 2:0 and 5:11 years and have communication needs with no known cause. In accordance with the Joanna Briggs institute scoping review methods guidelines, an initial search of the Ovid Emcare and Ovid Medline databases was conducted. Following this a final search strategy for these databases were produced. A draft extraction form was developed by the first author; this was then trialed by two authors on four articles each.

Discussion: Following the systematic development of an initial search strategy and extraction form, a scoping review of this topic can take place. The development of a rigorous scoping review protocol is essential in enhancing the transparency and reliability of the subsequent review. A pre-developed search strategy and trialing of an extraction form is a fundamental part of this process.

Keywords: scoping review, protocol, speech, language, intervention, pre-school

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1. Background

The aim of this paper is to provide a detailed protocol for the scoping review of interventions for pre-school children who present with co-occurring speech and language difficulties.

1.1 Terminology and prevalence

It is estimated that approximately 49.3% of paediatric speech and language therapists (SLTs) in England primarily work with pre-school children aged up to 5 years, thus forming a large part of the paediatric workforce (Roulstone et al., 2012). A large proportion of children receiving support from these services, estimated at approximately 55.4%, present with difficulties which have no known cause (Roulstone et al., 2012). At 4-5 years of age, the prevalence of children with language difficulties of unknown causation is estimated at approximately 7.6% (Norbury et al., 2016). For speech difficulties, recent findings from the Early Language in Victoria Study (ELVS) found a prevalence of 3.4% amongst a cohort of 1494 children (Eadie et al., 2014). These figures indicate a high level of need of support for children who have speech or language difficulties with no known cause.

Pre-school children with features of a Developmental Language Disorder (FDLD) present with language difficulties in the absence of an associated condition (i.e. no known cause) (Bishop et al., 2017). They are at risk of their language difficulties persisting into their later childhood years and beyond (Bishop et al., 2017). Such language difficulties are often viewed as heterogenous in nature, where an individual child may present with a combination of impairments relating to verbal learning/memory, discourse, pragmatics, word finding, understanding and use of words (semantics), grammar (morphology) and sentence building (syntax); for an overview see RCSLT (2020). Speech Sound Disorder (SSD) is an umbrella term used to describe difficulties with producing the individual speech sounds within words and sentences (Dodd, 2014). Both SSD and FDLT may present in isolation, but there is also evidence to suggest that an overlap between speech and language difficulties is observed (Shirberg & Kwiatkowski, 1994; Shriberg et al., 1999). This comorbidity was most recently highlighted in the ELVS cohort by Eadie et al. (2014), where 40.8% of 4-year olds presenting with SSD also had FDLT. The overlap of SSD with FDLT appears particularly strong when the language difficulty relates to areas of expressive language, such as use of morphology and syntax (Mortimer & Rvachew, 2010; Eadie et al., 2014; Mcleod et al., 2017). The term 'expressive language features of developmental language disorder' (eFDLD) will be used in the remainder of this paper when describing pre-school children who present with features of Developmental Language Disorder which relate to expressive language.

1.2 Relationship and long-term outlook for SSD and eFDLD

There are different sub-types of SSD, with phonology based SSDs (pSSD) being the most common subtype presenting within clinical services (McLeod & Baker, 2017). This is characterised by "an impaired ability to learn the speech-sound contrasts that discriminate words" (Dodd, 2014). Although pre-school children may present with different underlying deficits relating to SSD and eFDLD, it is suggested that phonology, that is how sounds are organised to form words, may be a key shared factor (RCSLT, 2020). This relationship was most recently highlighted by Howland et al. (2019), who illustrated the association between phonological errors (production of consonant clusters) and realisation of grammatical morphemes.

It is known that pre-school children with isolated pSSD or eFDLD are at risk of long-term needs relating to literacy, emotional wellbeing, educational attainment and /or everyday functioning (Johnson et al., 2010; McCormack et al., 2010; St Clair et al., 2019). Although knowledge about the co-occurrence of pSSD and eFDLD is emerging, it is becoming

increasingly evidenced that pre-school children with this co-occurring profile seem more likely to present with long-term communication and literacy needs compared to young children with isolated eFDLD or pSSD (Hayiou-Thomas et al., 2017; Lewis et al., 2015; McLeod et al., 2017).

1.3 Implications for clinical practice

Targeted and effective therapeutic input is essential as early intervention has potential to ameliorate future difficulties (Burgoyne et al., 2019). Roulstone et al. (2015) investigated interventions implemented by SLTs for pre-school children with communication difficulties of unknown causation, including pSSD and eFDLD, as a part of their ‘Child Talk’ study. The findings highlighted that in the absence of flexible, evidence-based interventions, SLTs may adapt interventions developed for other clinical groups, such as those for children with isolated pSSD or eFDLD. Such intervention adaptations by SLTs have also been evidenced in other comorbid clinical groups, such as stammering and SSD (Unicomb et al., 2013). There can be great value in SLTs adapting interventions according to a child’s unique needs, with clinical expertise being a key component of evidence-based practice model (Roulstone, 2015). However, limited empirical evidence underpinning these adaptations acts as a barrier to understanding which intervention ingredients effect real change within children with co-occurring pSSD and eFDLD.

The findings from Child Talk suggest that SLTs may not be aware of interventions specifically developed for co-occurring pSSD and eFDLD, or if they are, that they do not fit the service structure in which the SLT works. For example, Roulstone et al. (2015) highlighted that whilst some evidence exists for the use of broad target recasts in supporting speech and language difficulties concurrently (Yoder et al., 2005), participant SLTs within Child Talk did not report knowledge of this approach.

To date there is a lack of knowledge concerning the number and type of published intervention studies that explicitly target pSSD and eFDLD. There is also an apparent disconnect between available evidence and clinical practice. The proposed scoping review could shed light on this emerging field of work and support clinicians to map such evidence on to their clinical practice, where appropriate.

1.4 Reviews to date

To ensure that duplicate reviews for children with co-occurring pSSD and eFDLD in the pre-school population have not been undertaken or are already proposed, a preliminary search of the Cochrane Database of Systematic Review, JBI Evidence Synthesis, Pubmed, CINAHL, PROSPERO, Figshare and Open Science Framework was first conducted in January 2021. A systematic review by Tosh et al. (2017) and scoping review by Bellon-Harn et al. (2020) were found to include both speech and language interventions. However, the population for these studies were children with speech and/or language difficulties, rather than children with comorbid pSSD and eFDLD. Additionally, these reviews were specific in nature with Tosh et al. (2017) targeting parent delivered interventions only, and Bellon-Harn et al. (2020) exclusively focusing on parent-implemented interventions involving the use of videos and digital media. Clinical commentary papers were also identified (Hoover, 2019; Tyler, 2002; Tyler, 2016), however these were not written following an explicit scoping or systematic review methodology.

No further systematic or scoping reviews were found or are currently being undertaken for pre-school pSSD and eFDLD interventions.

1.5 Review objective

Given the sparsity of systematic evidence synthesis regarding interventions for this vulnerable group, the objective of this scoping review is to explore the extent of the current literature for interventions which target both pSSD and eFDLD.

A scoping review methodology, as opposed to a systematic review methodology, has been selected as this review is exploratory and descriptive in nature. Population characteristics and intervention content, delivery and outcomes measured will be identified from relevant studies. This will enable the researchers to identify what gaps there are in the literature for subsequent exploration, as well as mapping onto current practice.

In addition to standard scoping review methodology, a further objective of this review is to explore the nature of the quality of included studies. Although this is not essential when conducting scoping reviews (Peters et al., 2020), a broad overview of quality will provide indicative evidence as to whether a subsequent systematic review on the efficacy of the current evidence base may be justifiable.

1.6 Review question

What evidence exists for interventions targeting phonological SSD (pSSD) and expressive language difficulties (eFDLD) in pre-school children with this co-occurring presentation?

Within this overarching review question, the following sub questions are posed:

- 1) What is the content, context and delivery of the interventions described within included papers?
- 2) What are the broad quality characteristics of included papers?

2. Methods/Design

The scoping review will be conducted in accordance with the JBI methodology for scoping reviews (Peters et al., 2020). As a literature review ethical approval is not required. As scoping reviews cannot be registered with PROSPERO currently, this study was registered with the OSF in January 2021, with registration DOI 10.17605/OSF.IO/U6ADF.

2.1 Eligibility criteria

As in line with the JBI guidance, the eligibility for included studies will be outlined according to population, concept and context of data (Peters et al., 2020).

2.1.1 Population

The included population (pre-school children) are required to present with both pSSD and eFDLD, as indicated by their intervention targets. If pre-intervention assessments indicate typical development in either speech or language, these studies will be excluded. Studies will not be excluded if the intervention includes additional intervention targets (e.g., for receptive language). The expressive language targets could be related to vocabulary/word finding, semantics, syntax, morphology or a combination of these. The SSD targets have to be related to expressive phonology, which might include intelligibility. Children whose speech and language needs are associated with a biomedical condition with a known association with communication, such as sensorineural deafness, autistic spectrum condition or cleft lip and palate and neurological conditions affecting speech output, will be excluded. As the review aims to explore interventions for pre-school children, at least 80% of participants within the studies are required to be aged 2:0-5:11 years.

2.1.2 Concept

To be included in the review, studies must both target and assess the outcomes for speech sound production and expressive language either simultaneously or within the same episode of care or period of intervention. Anticipated change, or an exploration of change, in response to addressing both speech production as well expressive language must be stated within the aims of the intervention. To differentiate from studies focusing solely on early literacy skills, interventions targeting early sound awareness will be excluded unless they include an outcome measure of speech sound production. Studies meeting this core inclusion criteria may involve a SLT/relevant professional as a primary deliverer or as working in partnership with co-deliverers (e.g., parents, pre-school staff).

2.1.3 Context

The context for included studies will be open in that it will consider intervention studies taking place in any setting (e.g., home, clinic, nursery) and geographical location.

2.4 Information sources

As the aim of this scoping review is to provide a broad overview of evidence, it will not exclude relevant studies on account of study design. However, to maintain a minimum standard of research quality, included papers will have been published within peer reviewed journals. To locate papers with this minimum quality which have been subject to peer review, grey literature will be excluded. The included studies must present primary, empirical research. The complete search will include Ovid Medline, Ovid Emcare, OVID Embase, CINAHL, Psychinfo and ERIC. These databases have been selected because they cover a broad range of journals pertaining to medicine, psychology (including child development and education) and the allied health professions. Due to a limitation in resources, included studies will be in English. To ensure that historical intervention studies of potential relevance are not missed, the search will not include a minimum publication year. Where a potentially relevant article cannot be retrieved, direct contact with the study authors will be made.

2.5 Search strategy

In accordance with JBI protocol development guidance (Peters et al., 2020) an initial limited search of two databases was conducted prior to the full search being carried out. Initially, a set of key terms was developed by the first author, in consultation with two independent subject experts with significant postdoctoral research experience in the area. These terms were used for the initial limited search of Ovid Medline and Ovid Emcare to identify articles on the topic. With the support of a clinical librarian the text words contained in the articles and abstracts of relevant articles and the index terms used to describe the articles were used to develop a full search strategy for Medline, which can be found in Table 1. When completing the database search for the full review, keywords and index terms will be adapted for each selected database as appropriate. The reference list of all included sources of evidence will be screened for additional studies.

Table 1. Full search strategy for Medline.

1	(phonol* or phonem*).mp. (26294)
2	Speech Sound Disorder/ (733)
3	((speech or speak* or sound* or articulat* or phonetic*) adj5 (disorder* or impair* or difficult* or delay* or immatur* or deficit* or problem* or challeng* or develop* or developmental or comprehensib* or intelligib*)).mp. (64818)
4	1 or 2 or 3 (85647)
5	"expressive language".mp. (3223)
6	(syntax or semantic* or vocabulary or gramma* or grammatically or sentence*).mp. (127849)
7	exp Language Development Disorders/ (7817)
8	((language or linguistic or talk*) adj5 (disorder* or impair* or difficult* or delay* or immatur* or deficit* or problem* or challeng* or develop* or developmental or comprehensib* or intelligib*)).mp. (63502)
9	5 or 6 or 7 or 8 (177697)
10	("pre-school*" or preschool* or "nurser*" or "early year*" or "early childhood" or kindergarten).mp. (1082001)
11	exp INFANT/ or "CHILD, PRESCHOOL"/ (1826813)
12	(toddler* or infant* or child*).mp. (3814040)
13	10 or 11 or 12 (3860106)
14	4 and 9 and 13 (14598)
15	14 use medall (9424)
16	speech sound disorder/ (733)
17	(phonol* or phonem*).mp. (26294)
18	((speech or speak* or sound* or articulat* or phonetic*) adj5 (disorder* or impair* or difficult* or delay* or immatur* or deficit* or problem* or challeng* or develop* or developmental or comprehensib* or intelligib*)).mp. (64818)
19	16 or 17 or 18 (85647)
20	"expressive language".mp. (3223)
21	(syntax or semantic* or vocabulary or gramma* or grammatically or sentence*).mp. (127849)
22	((language or linguistic or talk*) adj5 (disorder* or impair* or difficult* or delay* or immatur* or deficit* or problem* or challeng* or develop* or developmental or comprehensib* or intelligib*)).mp. (63502)
23	exp developmental language disorder/ (7817)
24	20 or 21 or 22 or 23 (177697)
25	("pre-school*" or preschool* or "nurser*" or "early year*" or "early childhood" or kindergarten).mp. (1082001)
26	(toddler* or infant* or child*).mp. (3814040)
27	child/ or infant/ or preschool child/ or toddler/ (2755158)
28	25 or 26 or 27 (3824998)
29	19 and 24 and 28 (14589)
30	29 use emcr (5165)
31	15 or 30 (14589)

2.6 Study/source of evidence selection

Following the search, all identified citations will be collated and uploaded into Endnote and duplicates removed. Titles of studies which are clearly unrelated to the population and concept of the scoping review will also be removed. Two reviewers will independently review 10% of the remaining abstracts against the inclusion criteria as stated. They will meet to compare their selection of articles. If agreement is above 90% for at least 10% of the papers, one reviewer will review the remaining abstracts. If agreement does not reach this level, then a further 10% of papers will be reviewed by the two reviewers and further discussion had. This process will be repeated until there is less than 10% disagreement, or both reviewers have reviewed all of the abstracts. Once all abstracts have been reviewed, potentially relevant sources for full text review will be retrieved in full and imported into the JBI system for the Unified Management, Assessment and Review of Information (SUMARI) (Munn et al., 2019). The two reviewers will examine all selected papers independently at full text level with regular consensus meetings. Reasons for the exclusion of sources at full text level will be recorded and reported in the scoping review. Any disagreements that arise between the reviewers at each stage of the selection process will be resolved through either discussion or with an additional reviewer/s. The results of the search and the study inclusion process will be reported in full in the final scoping review and presented in a Preferred Reporting Items for Systematic Reviews and Meta-analyses extension for scoping review (PRISMA-ScR) flow diagram (Tricco et al., 2018).

Following extraction of included papers, critical appraisal tools (Joanna Briggs Institute, 2021) will be used to provide a broad overview of the quality of included papers. As the included studies may vary in study design, each study will be appraised using the corresponding study design checklist on SUMARI (Munn et al., 2019). Two reviewers will individually appraise each study, with regular consensus meetings to confirm ratings. If consensus cannot be met, a third reviewer will be consulted. Due to the likely variation in study design, which could include RCTs and individual case reports, it may not be deemed appropriate to make direct comparisons between some papers. This will be accounted for within the narrative synthesis of findings, where the authors will clearly state the study design for each paper with the corresponding commentary on level of quality based on its associated standards.

2.7 Data extraction

Data will be identified from relevant papers using a researcher-developed extraction form. This form was adapted from guidance provided by the Joanna Briggs institute Reviewer's Manual (Peters et al., 2020) in order to meet the specific requirements of the proposed review. The form was piloted by two independent reviewers on 4 relevant studies identified from the initial limited search. A final draft was agreed following a consensus meeting between the two reviewers and can be found in Table 2. This final draft was amended to include specific details about the population and concept as relevant to the aims of this review. Population details include age and key speech and language characteristics. Concept details include areas of speech/language addressed, intervention content and delivery and outcomes assessed. The data extraction tool will be revised if necessary during the process of extracting data from each included information source.

Table 2. Extraction form.

DATA CHARTING	
Evidence source details and characteristics	
Citation details (reference)	
Type of intervention study	
Country	
Participant details, including: <ul style="list-style-type: none"> • Number of • Age range in months Inclusion/exclusion, including: <ul style="list-style-type: none"> • Stated characteristics of speech + language difficulties • How were these characteristics identified? (e.g., speech sample, standardised assessment) • Exclusion criteria and how identified 	
Aims	
Extracted content and delivery details	
CONTENT: What aspects of speech production are targeted within the study aims? (e.g., PCC, intelligibility, phonological processes)	
CONTENT: What aspects of expressive language are targeted within the study aims? (e.g., morphemes, MLU, vocabulary)	
CONTENT: Key approaches, activities and strategies stated <ul style="list-style-type: none"> - Speech specific? - Language specific? - Combined speech/language? 	
FORMAT: Approaches/activities/strategies combined/integrated, or <u>sequential</u> for speech and language?	
DELIVERY: <ul style="list-style-type: none"> • Setting (e.g., home, clinic, nursery) • Deliverer (e.g., SLT consultative with parent, SLT only) • Dosage • Duration 	
OUTCOME MEASURES: <ul style="list-style-type: none"> - Name/brief description - Aspect of speech and/or language measured - Inclusive of Validated, Unvalidated e.g., adapted MLU, PCC, Intelligibility rating, measures of functional impact/participation 	

DATA COLLECTION TIME POINTS	
FINDINGS: As provided in relation to the measures identified in the outcome measures cell/row	

2.8 Analysis of the evidence

The results of each included paper will not be independently reported as this study is not being conducted within a systematic review methodology (Peters et al., 2020). However, as a broad overview of study quality has been included within this scoping review process, a brief synthesis of overall study findings will be reported narratively. This will help indicate if there are any findings of note which could be investigated further if a systematic review were to be conducted. As in line with a scoping review methodology, analysis of the findings will be largely descriptive, with frequency counts relating to the concept and context of studies where appropriate.

2.9 Presentation of the results

The overall study information with concept and context data will be presented in tabular form (Tables 3 and 4) with a corresponding narrative summary for each section. The findings from the quality appraisal will be discussed narratively, with tables summarising reviewer appraisal ratings attached to the appendix. As the presentation of data is an iterative process dependent on study findings (Peters et al., 2020), these presentation approaches may be further refined at review stage according to the content of the findings.

Table 3. Presentation of overarching study information.

Overarching study information and population					
Reference (country)	Type of study	Aims (as relevant to the review)	Comparison	No children	Age range at baseline

Table 4. Presentation of concept and context characteristics.

Concept (i.e. intervention features)				Context
Speech outcomes	Expressive language outcomes	Format	Intervention approaches	Delivery and dosage

3. Conclusion

Pre-school children with co-occurring pSSD and eFDLD are a group presenting frequently within clinical services who are at risk of long-term literacy and communication needs. To date, systematic/scoping reviews have only focused on interventions for speech or language in isolation, and there is a need for an explicit and systematic review of the literature on interventions for children with this dual presentation. This protocol has described the initial limited search process, the development of a usable extraction tool, as well as an overview of how evidence will be analysed and presented. The next stage will be to conduct the full review and report on the findings as in accordance with this protocol.

Declarations

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