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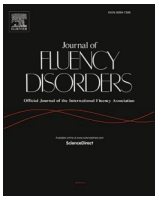

Interventions for children and adolescents who stutter: A systematic review, meta-analysis, and evidence map

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ABSTRACT

Purpose: This systematic review critically appraises and maps the evidence for stuttering interventions in childhood and adolescence. We examine the effectiveness of speech-focused treatments, the efficacy of alternative treatment delivery methods and identify gaps in the research evidence.

Methods: Nine electronic databases and three clinical trial registries were searched for systematic reviews, randomised controlled trials (RCTs) and studies that applied an intervention with children (2–18 years) who stutter. Pharmacological interventions were excluded. Primary outcomes were a measure of stuttering severity and quality assessments were conducted on all included studies.

Results: Eight RCTs met inclusion criteria and were analysed. Intervention approaches included direct (i.e. Lidcombe Program; LP) and indirect treatments (e.g. Demands and Capacities Model; DCM). All studies had moderate risk of bias. Treatment delivery methods included individual face-to-face, telehealth and group-based therapy. Both LP and DCM approaches were effective in reducing stuttering in preschool aged children. LP had the highest level of evidence (pooled effect size=-3.8, CI -7.3 to -0.3 for LP). There was no high-level evidence for interventions with school-aged children or adolescents. Alternative methods of delivery were as effective as individual face-to-face intervention.

Conclusion: The findings of this systematic review and evidence mapping are useful for clinicians, researchers and service providers seeking to understand the existing research to support the advancement of interventions for children and adolescence who stutter. Findings could be used to inform further research and support clinical decision-making.

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1. Introduction

1.1. What is stuttering?

Stuttering is a speech disorder characterised by involuntary repetition or prolongation of sounds, syllables or words, or by involuntary hesitation or pauses that disrupt the rhythmic flow of speech (World Health Organisation, 2001). Stuttering affects around 1 % of the population (Yairi & Ambrose, 2013) and has significant public health impacts. Evidence suggests later psychological difficulties may originate during the school years in children who stutter (Smith, Iverach, O'Brian, Kefalianos, & Reilly, 2014). School-aged children who stutter are at increased risk of teasing, bullying and anxiety and stuttering is also highly associated with occupational and educational under-achievement and suicidal thoughts (Nye et al., 2013). If not treated during childhood, persistent stuttering can result in lifelong social, educational and occupational reduced quality of life. A higher proportion of adults who stutter have social phobia and anxiety compared to adults who do not stutter (Blumgart, Tran, & Craig, 2010; Craig, Blumgart, & Tran, 2009; Iverach, Jones et al., 2009; Iverach, O'Brian et al., 2009; Kloth, Kraaimaat, Janssen, & Brutten, 1999; Smith et al., 2014; Stein, Baird, & Walker, 1996).

According to the WHO International Classification of Functioning, Disability and Health (ICD), treatment for stuttering should aim to make speaking easier, mainly by eliminating or reducing the quantity of stuttering symptoms (World Health Organisation, 2001). Internationally, stuttering treatment approaches have long been debated. There have been diverse theoretical approaches regarding the causes of stuttering and subsequently there are several different treatment approaches for children.

1.2. Treatment approaches for stuttering

Traditionally two main intervention approaches have been the subject of investigation for the treatment of stuttering during the preschool years. Direct intervention approaches are designed to address a child's speech directly (e.g. Lidcombe Program; LP), while indirect approaches focus on modifying the child's immediate surroundings to create a fluency-inducing environment and building the child's capacities for speaking fluently (e.g. Demands and Capacities Model; DCM). In this review, for ease of distinguishing the approaches, we will refer to direct interventions as those that sit toward the direct end of the continuum and indirect interventions as approaches as those that sit toward the indirect end of the continuum. In contrast to interventions for preschool children, there is limited research on the best interventions for school-aged children. For school-aged children, intervention approaches tend to be drawn from those designed for use with preschool children or those used for adults (e.g. speech restructuring programs).

1.3. Interventions for children who stutter

The LP is a parent-directed behavioural modification program, whereby the speech language pathologist (SLP) teaches the parent how to use the program (Onslow, Packman, & Harrison, 2003). Here the child is praised for stutter-free speech and gently corrected when they stutter. Parents complete practice at home (or in the child's usual environment) each day and receive training on how to rate the severity of the child's stutter on a scale to monitor progress. There are two stages in this treatment approach. Initially the parent and child are seen on a weekly basis where the parent is trained by the speech pathologist to deliver the intervention through everyday activities throughout the day. The intervention moves from more to less structured activities. The goal of this stage is for the child to eliminate or minimise their stutter. Once the child has achieved Stage 1 (minimisation of stutter) they move on to Stage 2, which is the maintenance phase of the intervention. Here the intervention is gradually withdrawn, and less support is provided by the parent over time. The mechanisms by which this intervention works is not fully understood. By contrast, the DCM approach trains parents to decrease the relevant motoric, linguistic, emotional or cognitive demands on the child which may be triggering stuttering, whilst simultaneously building the child's capacity across domains to enhance fluency (Franken, 2013). The aim is to modify the child's communicative environment using both interaction strategies in order to create an environment that induces fluency. While the intervention itself is standard, the strategies are individualized for the child. Parents attend weekly intervention sessions until they master to strategies and the child has reached an acceptable level of fluency. Parents practice the techniques at home each day and complete home assignments. Following this, the child completes a period of review to ensure maintenance.

Traditionally, preschool stuttering interventions are delivered one-to-one by parents who are trained by speech-language pathologists. More recently, a range of alternative treatment delivery methods have been examined, including interventions via telehealth, or delivered in groups. If the efficacy of these treatment delivery methods is found to match those of standard one-to-one / face-to-face methods, there are wide reaching implications regarding healthcare costs, and accessibility for those who cannot access treatment due to their remote location or other reasons.

In school-age children intervention tends to be less tractable and relapse is more likely (Koushik, Shenker, & Onslow, 2009; Lincoln, Packman, & Onslow, 2006). As children age during the school years, the types of interventions used to reduce stuttering typically focus less on eliminating stuttering and more on minimization and control of stuttering. For example, speech restructuring techniques may be used. The primary school years, when children who stutter are 7–11 years old, is a time when the origins of later psychological problems may also occur. Two systematic reviews have been published on interventions for stuttering in the last five years. One review focused of non-pharmacological interventions for adults and children. This review conducted electronic database searches in 2013, so an update of the research is required (Baxter et al., 2016). The second systematic review focused only on studies of adults and children that utilised telehealth (McGill, Noureal, & Siegel, 2019). The review by McGill et al. (2019) did not include the assessment of risk of bias assessments or an overall grade of the evidence.

Table 1
Search terms[a].

Concept	Definition	Search terms
Concept 1	Relates to stutter, stammer, dysfluency or PWS	Stutter* OR Stammer* OR D?sfluence OR D?sfluency OR D?sfluencies OR PWS
Concept 2	Involves some type of therapy, intervention or treatment	Therap* OR Intervention* OR Treat*
Concept 3	Includes children, adolescents or adults	Child* OR Adolescen* OR Adult*

[a] This systematic review was part of a greater project that explored both adult (Brignell et al., 2020) and childhood interventions separately; however the same search strategies were used for both with the evidence synthesis focused on the separate population groups.

Table 2
Inclusion and exclusion criteria for studies.

Dimension	Search strategy concepts	Initial screening criteria (Title/Abstract)	Full text screening
Population	Eligible populations are people who stutter, aged ≥2 years old and < = 18 years	Exclude studies where participants are (explicitly) aged <2 years old and >18 years	Exclude studies where participants have an acquired fluency disorder
Intervention	Contain some form of therapy, intervention, or treatment that is specifically designed to benefit PWS	Exclude studies that include a pharmacological intervention (including anxiolytic)	Exclude studies that did not include an aim of improving fluency outcomes
Comparator	Comparison to an alternative intervention, no intervention (control group) or usual practice	Exclude studies that included a pharmacological intervention (including anxiolytic)	
Outcome	Is a measure of stuttering using a standard assessment pre- and post-randomisation/ intervention	Exclude studies that do not include a measure of stuttering	
Study design	Following types of studies were included: Systematic Reviews, Meta-Analyses, and Randomized Controlled Clinical Trials.		
Other criteria	Study published between 1 Jan 2005 and 20 January 2019; Study published in English; Study relates to humans; Study conducted in Australia, New Zealand, Canada, UK, Europe or the US		

Thus, the current study is a systematic review, meta-analysis, and evidence mapping of the existing literature of the evidence for interventions for children who stutter. This is also the first study to systematically review a range of alternative treatment delivery methods (e.g. telehealth, groups), to synthesise the evidence and to include rigorous review of the quality of the evidence. The results of this review provide an overview of interventions for children who stutter to guide further evaluation and prioritised research during this crucial time of child development.

1.4. Purpose of this study

The primary aim is to identify and describe existing empirical evidence on interventions for children who stutter. Firstly, we explore the state of evidence surrounding interventions designed to reduce stuttering in children (preschool and school-aged), the effectiveness and efficacy of interventions and alternative delivery methods. Further, we identify gaps in the evidence where new primary studies or systematic reviews could add value and provide accessible best available evidence in the field.

2. Materials and methods

2.1. Search strategy

This systematic review was conducted in accordance with the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines. A comprehensive database search was conducted (20 January 2019) using nine electronic databases and trial registries: CINAHL (Ebsco), Cochrane Reviews, Embase/Medline (Ovid: 1946 to Present with Daily Update), PsycExtra (Ovid), PsycINFO (Ovid), ClinicalTrials.gov, WHO International Clinical Trials Registry Platform, Cochrane Central Register of Controlled Trials). We also searched reference lists from included articles and existing relevant reviews and additional evidence were sourced via an intuitive and purposeful electronic search of the literature, based on selected key documents identified by review authors. Search strategies are presented in Table A1 in Appendix A.

The search terms were categorised into three main concepts (Table 1). We checked the thesaurus from each database where available to ensure all appropriate terms were included. Study titles, abstracts and inclusion criteria were independently screened by three reviewers (AB, EK, MK). A third reviewer was used to resolve any discrepancies (SR). We reviewed the full text for all studies that appeared to meet our inclusion criteria. Studies that did not meet inclusion criteria based on full text were excluded with reasons.

2.2. Inclusion and exclusion criteria for included studies

Included in this review were studies of preschool and school-aged children who stutter, aged 2–18 years and were required to have used an intervention that was designed to reduce stuttering. Excluded were studies of pharmacological interventions (Table 2). There were no limitations on the defining features of the comparator intervention (e.g. alternative intervention, control group or intervention

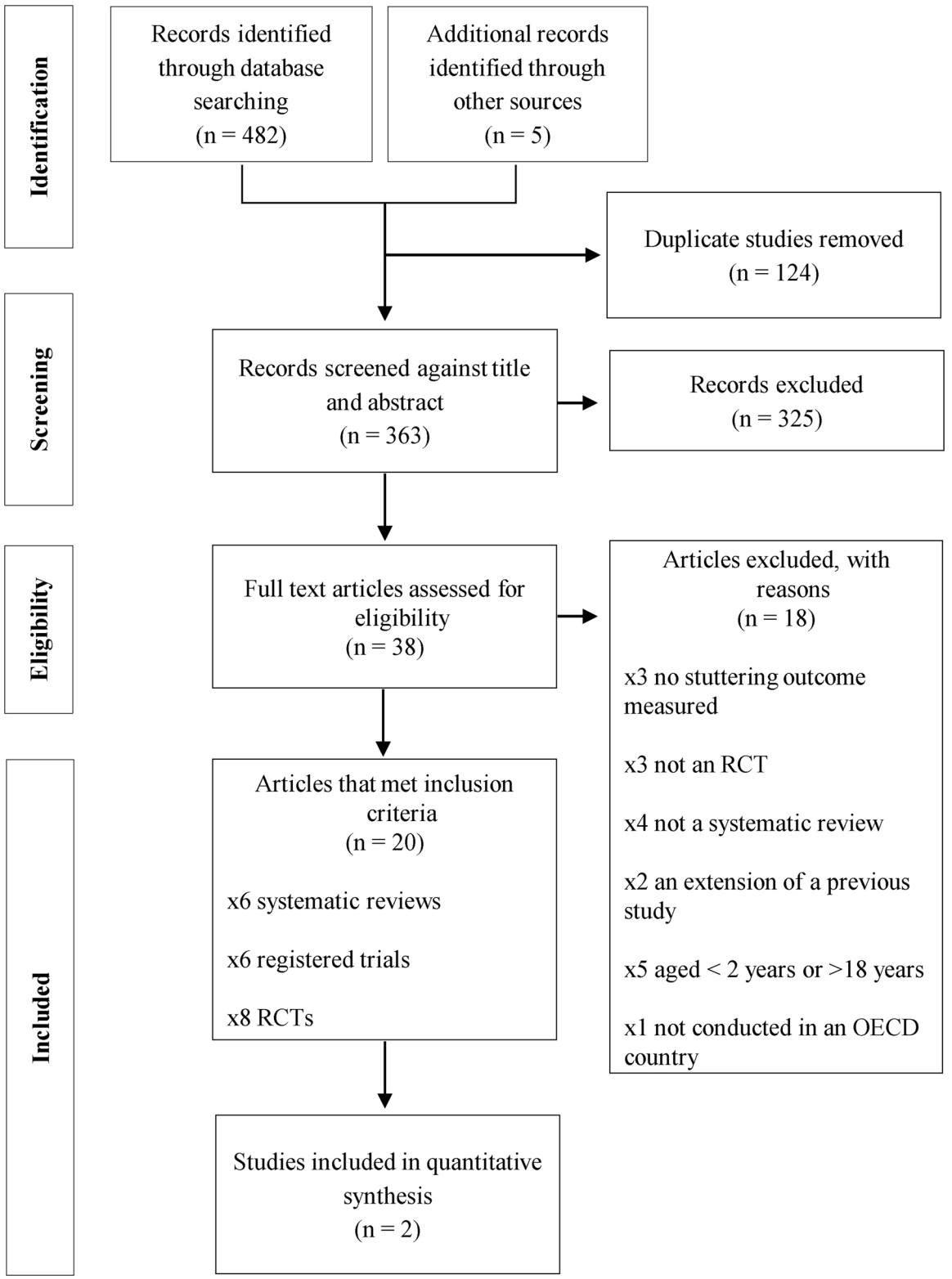

Fig. 1. The PRISMA flow diagram illustrates the sequence of identifying studies for inclusion in this review. RCTs, randomised controlled trials.

as usual) or the length of the intervention or follow-up. We included studies that were systematic reviews (Level I) and randomised controlled trials (RCTs) (Level II), representing the highest level of evidence on the National Health and Medical Research Council (NHMRC) evidence hierarchy classification (National Health & Medical Research Council, 2009).

Primary outcomes included a measure of stuttering using a standard assessment designed to measure change pre and post intervention. For example, %SS or stuttering severity ratings (SR). We limited searches to literature published since January 2005, in the English language and related to studies conducted in humans from Australia, Canada, New Zealand, United States, United Kingdom and Europe. These countries are all members of the Organization for Economic Cooperation and Development (OECD).

2.3. Data extraction and management

Data were extracted and entered in a standardised form. Data was collected on the following: authors, year, country, study design, study aim, detail of the intervention (key features), comparator/control, details of participants (age, gender and inclusion/exclusion criteria), study measures and outcomes (time points, primary and secondary outcomes and main outcomes) and funding sources. Data extraction was performed by one member of the review team (MK) and checked against the paper by a second member (AB).

2.4. Quality assessment

The methodological quality of each included study was assessed by two authors (MK, MD), based on Cochrane criteria (Higgins & Altman, 2008; Higgins et al., 2011). MK or MD have not been involved in prior studies on stuttering and neither have collaborated with any of the authors of the included studies. Risk of bias ratings focused on: selection bias, performance bias, detection bias, attrition bias, reporting bias and other biases that might be relevant. The overall quality of evidence was rated using the NHMRC evidence hierarchy (National Health & Medical Research Council, 2009).

2.5. Statistical analysis

We conducted meta-analyses where studies were sufficiently homogeneous in terms of participants, interventions, comparators and outcome measures to provide a clinically meaningful summary. We pooled continuous data using mean difference and confidence intervals (pre and post intervention) as the summary statistic. Quantitative synthesis was conducted using a random-effects model on the standardised mean difference to assess the impact of statistical heterogeneity. Where studies were not sufficiently similar, to enable meta-analysis, we provided a narrative synthesis. The analyses were carried out using STATA® V.13 statistical package (StataCorp., 2013).

2.6. Evidence mapping

The analysis of studies included in this systematic review and evidence map is primarily descriptive. Using the data extracted (as described in Section 2.3), we used frequencies and percentages to present and provide an overview of the evidence base through summaries of the study characteristics, outcomes measured and key findings. To identify gaps in the research, data were tabulated and visualised as a bubble plot, grouping studies by intervention approach, population characteristics, method of delivery, comparator and control, and reported outcomes.

3. Results

3.1. Search results

Our literature search identified 482 records and an additional five studies were identified through other sources. This was reduced to 363 records once duplicates were removed. Full text review was completed on 38 studies, of which 18 were excluded (Fig. 1). Further detail on excluded studies can be found in Table B1 in Appendix B. A total of 20 studies met the inclusion criteria. Of these, six were systematic reviews (Bate, Malouff, Thorsteinsson, & Bhullar, 2011; Baxter et al., 2015, 2016; Herder, Howard, Nye, & Vanryckeghem, 2006; Lowe, O'Brian, & Onslow, 2013; Nye et al., 2013), six were registered trials (De Sonneville-Koedoot, 2007; Onslow, 2008, 2009a, 2009b, 2013; Paterson, 2009) and eight were RCTs (Arnott et al., 2014; Bridgman, Onslow, O'Brian, Jones, & Block, 2016; de Sonneville-Koedoot, Stolk, Rietveld, & Franken, 2015; Donaghy et al., 2015; Franken, Kielstra-Van der Schalk, & Boelens, 2005; Jones et al., 2005; Lattermann, Euler, & Neumann, 2008; Lewis, Packman, Onslow, Simpson, & Jones, 2008). Systematic reviews were used to ensure we had identified all relevant studies. We contacted authors of registered trials by email to ask if they had data to share, however, no registered trials provided data.

3.2. Characteristics of included studies

There were eight primary studies (Arnott et al., 2014; Bridgman et al., 2016; de Sonneville-Koedoot et al., 2015; Donaghy et al., 2015; Franken et al., 2005; Jones et al., 2005; Lattermann et al., 2008; Lewis et al., 2008) including 492 participants. The mean age of participants was 4.14 years, ranging from 2;10 to 6;11 years, and the gender ratio was 3:1 (male: female). All participants were reported to have been stuttering longer than six months prior to joining the study and all had more than two percent of syllables stuttered

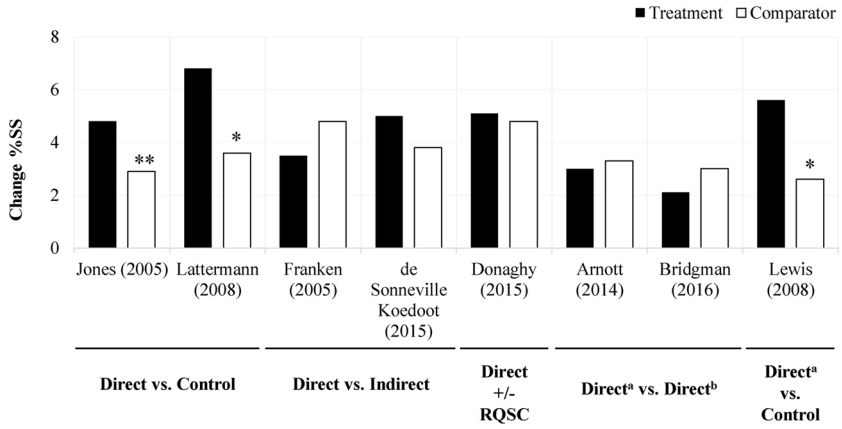

Fig. 2. Change in %SS from baseline to primary outcome timepoint for each study, as categorised by intervention approach and/or delivery method. [a]LP adapted; [b]Standard LP; %SS: percent syllables stuttered; RQSC: Request self-correction of stuttered speech. *$p < 0.05$; **$p < 0.005$.

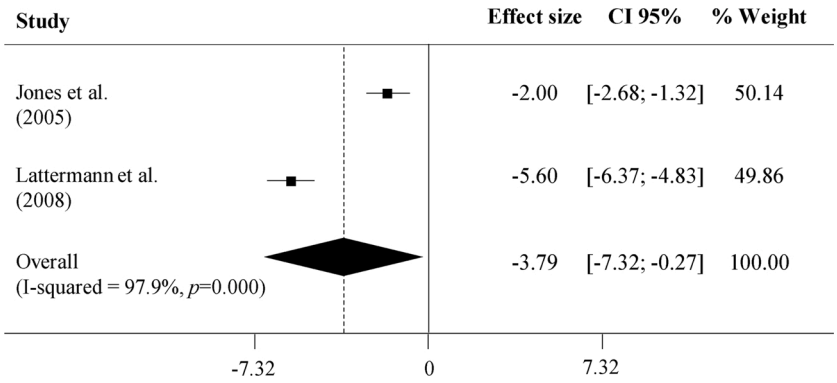

Fig. 3. Forrest plot of effect size for the LP in reducing stuttering from two trials.
Note. Scores to the left of the solid vertical line indicate a reduction in %SS. The confidence intervals for the total are represented by the diamond shape. Weights are from random effects analysis.

(%SS) (in some cases this criterion increased to three or greater). Studies were all RCTs conducted predominantly in Australia (n = 4), followed by the Netherlands (n = 2), New Zealand (n = 1) and Germany (n = 1). Most of the studies recruited through wait lists at participating speech clinics. Other sampling locations included community health services (n = 1), preschools (n = 1), GP clinics (n = 1) and advertisements in the media (n = 3). Table C1 in Appendix C contains further details of study characteristics.

3.3. Characteristics of interventions

Two main intervention approaches were identified. Seventy-five percent (n = 6) were direct treatments such as speech modification strategies that bring awareness of stuttered speech, ultimately changing moments of stuttering, and fluency-enhancing strategies that target the client's speech in order to facilitate fluency behaviours (e.g. LP). The remaining 25 % (n = 2) were indirect treatments, traditionally based on the creation of a fluency-inducing environment for the individual by promoting positive interaction (e.g. DCM). A full description of intervention characteristics is provided in Table D1 in Appendix D.

All eight studies included the LP either as an intervention and/or comparator. Two studies compared the LP to a control group (Jones et al., 2005; Lattermann et al., 2008), one compared LP with DCM (Franken et al., 2005), one compared LP with RESTART-DCM (de Sonneville-Koedoot et al., 2015), one study compared a variation of the LP (removal of the self-correct contingency aspect) to standard delivery (i.e. one-to-one / face-to-face) (Donaghy et al., 2015), two compared standard LP versus telehealth adapted (Lewis et al., 2008), (Bridgman et al., 2016) and one study compared standard LP to a group-adapted format (Arnott et al., 2014).

3.4. Effectiveness of interventions and alternative delivery methods

All studies reported reductions in %SS and some studies indicated benefits continuing or being maintained for up to 18 months post-randomisation (Fig. 2 and Table E1 in Appendix E). Both the LP and DCM approach were found to be effective, with the LP having more RCTs supporting the approach and higher levels of evidence. In all three studies (Arnott et al., 2014; Bridgman et al., 2016; Lewis

et al., 2008) that used an alternative delivery method, the intervention was found to be as effective as standard (i.e. one-to-one / face-to-face) delivery. In the following section we describe the results as categorised by intervention approach and/or delivery method. We explored the efficacy of interventions where such information was available.

3.4.1. Direct intervention compared with control group

Jones et al. (2005) compared a group of Australian children receiving the LP to a control group (delayed treatment) in an RCT. At nine months post-randomisation, the children receiving the intervention significantly reduced their stuttering frequency from 6.4 %SS (SD 4.3 %) at baseline to 1.5 %SS (SD 1.4 %). There was a significant difference between the control group and the treatment group ($p = 0.003$). In a similar study, Lattermann et al. (2008) compared the LP to delayed treatment (control), to investigate whether it was superior to natural recovery in 45 German preschool children. Analysis of spontaneous speech samples collected within the child's home environment, reported a 6.9 % decrease in the %SS in the Lidcombe group, compared with the control group (1.6 %SS) at 16 weeks post-intervention. The within-clinic speech sample measures were similar, at 6.8 % and 3.6 % reduction of syllables stuttered for the intervention and control groups, respectively. Both measures of stuttering frequency, showed significant differences between the treatment and control groups ($p = 0.003$ home and $p = 0.025$ clinic). All children completed the LP in the intervention group, reducing their dysfluency rate by a mean of 70.3 %. The control group reduced their mean dysfluency rate by 17.6 %, with 13 children decreasing stuttering frequency and nine increasing stuttering frequency.

Data from both studies (Jones et al., 2005; Lattermann et al., 2008) were pooled using the mean difference and 95 % confidence intervals (CI). The effect size for the LP was -3.795 (CI -7.323 to -0.267). That is a reduction of 3.795 %SS (Fig. 3). The heterogeneity chi-squared was 47.3 which was statistically significant ($p=<0.001$). There was substantial variation in the mean difference due to heterogeneity ($I^2 = 97.9$ %).

3.4.2. Direct intervention compared with indirect intervention

Franken et al. (2005) compared the LP with DCM in 30 Dutch preschool children. Stuttering frequency decreased from 7.2 %SS (SD 2.1 %) at baseline to 3.7 %SS (SD 2.1 %) post-intervention in the Lidcombe intervention group. In the DCM group stuttering frequency decreased from 7.9 % (SD 7.1 %) to 3.1 % (SD 2.1 %). Further analysis in this study showed a significant effect of time, but no effect of treatment and no effect of treatment x time, indicating both the LP and DCM interventions provided similar benefits in reducing stuttering frequency. In addition, the parent and therapist ratings of the children's stuttering severity were similar, with no reported difference between treatments ($p > 0.10$). In another Dutch study led by de Sonneville-Koedoot et al. (2015) a large cohort ($n = 176$) of preschool children received either the LP or DCM intervention. This study known as the Rotterdam Evaluation Study of Stuttering Therapy (RESTART-) DCM, is premised on the idea that positive changes in the child's functioning and/or in the environment will lead to a reduction in stuttering. Children who completed the RESTART-DCM treatment achieved a reduction in stuttering frequency from 6.2 % (SD 4.4 %) at baseline to 1.2 % (SD 2.1 %) at 18 months post-randomisation. This is a reduction in frequency of 71.4 %. Children who completed the LP, had a reduction in the %SS from 5.3 % (SD 4.3 %) at baseline to 1.5 % (SD 2.1 %) at follow-up, resulting in a 71.7 % reduction. There was no statistically significant difference between the two treatments ($p = 0.45$).

3.4.3. Direct intervention with and without RQSC

In a study by Donaghy et al. (2015) the role of parental RQSC in preschool children completing the LP was explored. In this RCT, children completed treatment either with ($n = 16$) or without ($n = 18$) the RQSC, achieving on average a 50 % reduction in stuttering frequency that was maintained (or further reduced) for three consecutive weeks. No significant difference was found for stuttering frequency between the two groups, suggesting the verbal contingency RQSC may not be an essential component of the intervention.

Studies that have compared standard LP to alternative delivery methods (e.g. telehealth, and group-based) have also shown reductions in %SS using the LP. These are discussed in more detail below.

3.4.4. Direct intervention delivered via standard compared to group format

In a non-inferiority study by Arnott et al. (2014) the delivery of standard LP was compared to a group format. Group delivery required parent-child dyads to attend the speech clinic for a group session, consisting on average of three parent-child pairs. This was described by the study authors as a rolling group format, where groups constantly changed their composition, with respect to number of pairs and experience using the LP techniques. No significant difference was found between the group compared to face-to-face delivery in the %SS between baseline and nine- and 18-months post-randomisation ($p = 0.80$ and $p = 0.30$). In addition, the parent-reported stuttering severity showed no difference between the two groups. This data indicates that the group delivery for the LP was non-inferior in efficacy to standard delivery. There was no significant difference in the number of clinic visits between the group compared with face-to-face delivery of the LP (both received a median of 18 visits). On average, children in the group delivery received 9.2 h of SLP time, compared to 14.3 in the one-to-one consultations. This means that the group delivery consumed 46 % (95 % CI [57-32 %]) fewer SLP hours to complete Stage 1 of the LP (75 % of children in both groups reached stage 2). In addition, the participants in the group delivery completed stage 1 in 29 weeks, compared to 25 weeks in those attending one-to-one consults.

Overall, responses from parents who participated in the face-to-face consultations provided more positive responses than from parents in the group delivery. Fewer parents in the group delivery reported satisfaction at the 'extreme' end of the scale, especially related to: receiving enough training in the LP techniques; and, feeling comfortable interacting during the group clinics.

3.4.5. Direct intervention delivered via standard compared to telehealth-adapted format

Bridgman et al. (2016) compared the telehealth adapted delivery of the LP (webcam consultations) with standard LP. This method

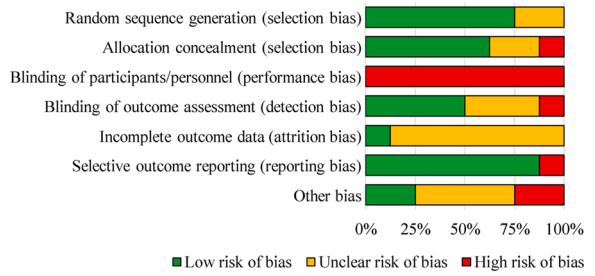

Fig. 4. Summary of risk of bias assessment of included studies.

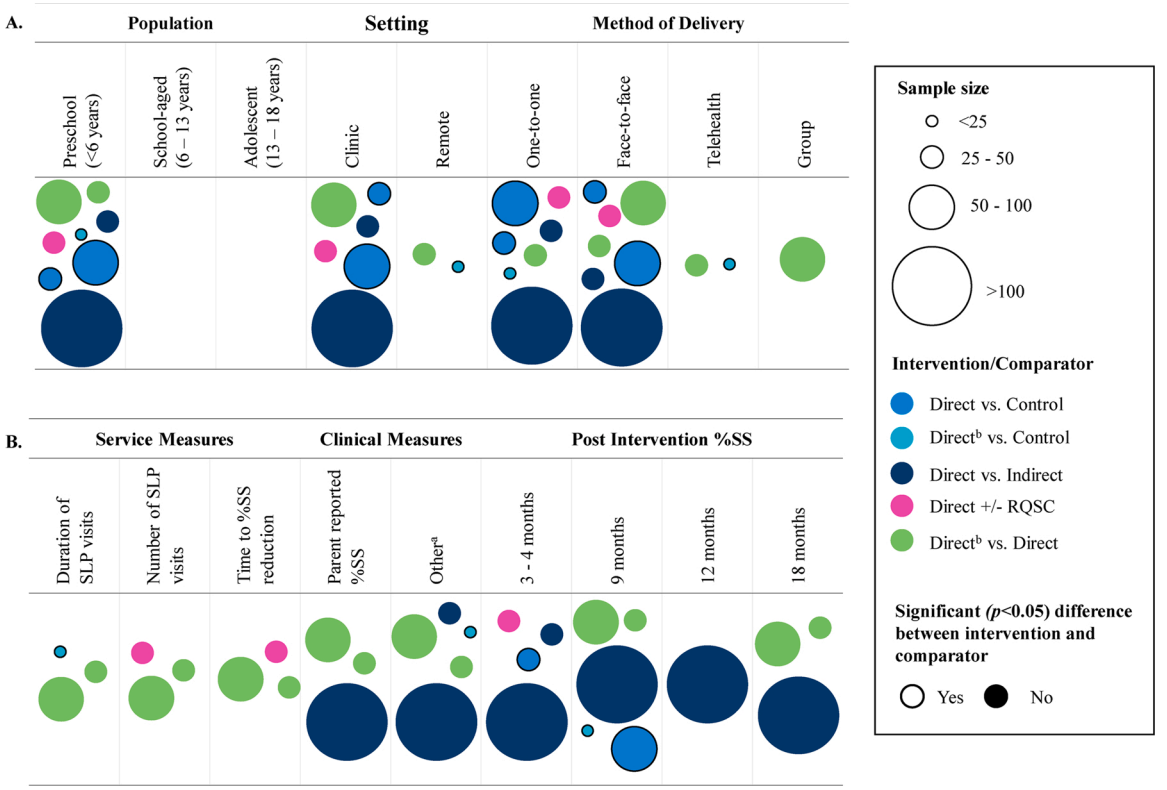

Fig. 5. A&B. Evidence map of intervention studies for children and adolescents who stutter as categorised by: (A) study characteristics; (B) outcome measures. Each bubble represents one study and the size represents participant sample size. Colours indicate intervention and comparator combinations and a solid border represents a significant difference between the reduction in %SS of the intervention and comparator group. [a]Includes: parent satisfaction survey, parent perceived relationships with SLP, KiddyCAT, CBCL, HRQoL; [b]LP adapted. Direct = LP standard unless otherwise noted.

of delivery has been previously reported to be a viable approach in a study with three preschool children (O'Brian, Smith, & Onslow, 2014). Children in the standard delivery group reduced the %SS from 4.0 % (SD 2.9) to 1.0 % (SD 1.0 %) and those in the telehealth group reduced their %SS from 3.8 % (SD 2.8 %) to 1.7 % (SD 2.0 %). Bridgman found no significant difference in stuttering frequency and parent-reported stuttering severity between the two treatment delivery methods ($p = 0.16$).

The median number of SLP consultations to complete stage 1 of the LP, was 23 for the standard delivery and 20 for those receiving webcam ($p = 0.71$). There was no evidence of an effect of age on the number of sessions ($p = 0.17$), however there was evidence that parent-reported stuttering severity at pre-randomisation was associated with 13 % more consultations required for each unit increase in stuttering severity ($p = 0.027$, 95 % CI [1–26]). The median number of weeks to complete stage 1 of the LP was 25 for both groups.

The average duration of each SLP consultation for children who had completed stage 1, was 40.4 min in the standard group and 33.4 min in the webcam group ($p < 0.001$, 95 % CI [3.4, 10.7]). Consultations with children who received telehealth delivery was 17 % shorter than those attending face-to-face clinic visits. The average weekly travel time to the clinic was reported as 66 min (range 15–180 min) and the average maximum travel time that parents reported they would consider, was 99 min (range 10–300 min).

3.4.6. Direct intervention delivered via telehealth compared with control group

A study by Lewis et al. (2008) compared a telehealth adapted delivery of the LP (telephone consultations) to no intervention. Children in the telehealth group reduced their %SS from 6.7 % to 1.1 % and those in the control group reduced their %SS from 4.5 % to 1.9 %. This phase II trial reported a treatment effect size of 73 % decrease in stuttering frequency compared to no treatment ($p = 0.02$). Telehealth delivery of the LP required on average 49 ± 26.8 (range 27–98) consultations, over 62.9 ± 31.9 (range 6.4–9.0) weeks, with a mean of 7.7 ± 0.9 (range 6.4–9.0) days. The average duration of SLP consultations was 33.1 ± 9.0 (range 26–52) minutes. The majority (87 %) of parent satisfaction questionnaires indicated that the telehealth delivery alternative had been a positive experience, regarding both the treatment process and the outcome of treatment. All parents rated themselves as "very satisfied" with their child's speech outcomes over the previous month.

3.5. Quality assessment

Fig. 4 displays the summary of the risk of bias assessment, while the individual studies' quality assessment results are presented in Table F1 in Appendix F. The overall risk of study bias was scored as moderate, mainly due to insufficient reporting on allocation concealment, inability to blind participants, non-standardised and unblinded analysis of outcome assessments, and attrition not clearly described. Only five studies explicitly mentioned the use of an independent researcher or centre to conceal allocation sequence from those assigning participants. High risk of bias is inevitable among studies in which an in-person intervention is compared to passive control, due to the inability to blind participants and speech pathologists. However, five of the nine studies attempted to minimise this high risk of detection bias by conducting baseline outcome assessments pre-randomisation with observers who were unaware of assignment. Additionally, half of the studies reported significant attrition (>15 %), with limited accounting. For 'other', four of the studies were judged as having an unclear risk of bias due to low samples sizes or lack of sample size calculation ($n = 3$) and the inclusion of participants with stuttered speech lower than the inclusion criteria ($n = 1$). Two studies were judged as having a high risk of bias for 'other', due to stopping recruitment early and a high detection bias (i.e. parents were not blinded allocation and were responsible for treatment support/delivery) that seriously weakens confidence in results. It is worth noting that six of the eight studies that investigated the LP were conducted by the authors that originally developed the approach.

We used the NHMRC levels of evidence hierarchy (National Health & Medical Research Council, 2009) to provide an overall rating of the levels of evidence. The overall grade of recommendation was determined through consideration of five domains: the evidence base, consistency of the findings, generalisability, clinical impact and applicability. Based on the available evidence, for preschool children, the LP was rated 'Grade A' (excellent) and was considered 'excellent' across four of the five domains. The clinical impact was rated 'Grade B' (good) for preschool aged children to account for the smaller sample sizes of these studies and higher attrition. The RESTART-DCM approach was rated 'good' for all domains, except for generalisability which was rated 'excellent'. There were no studies that included RCTs for older school-aged children/adolescents so the level of evidence for this group was rated poor.

3.6. Gaps in the evidence

The resulting evidence maps show study characteristics and outcome measures as distributed by intervention approach (Fig. 5A&B). Each study is represented as a bubble and the size is proportional to the evidence base (sample size). The first evidence map (Fig. 5A) highlights the paucity of studies to inform effectiveness of interventions for school-aged children and adolescents who stutter, with no RCTs for these age groups. Additionally, only two studies have examined interventions delivered in settings outside of the clinic as telehealth adapted methods. One study involved group-format delivery. All were direct interventions. The second evidence map (Fig. 5B) highlights the variation of reported outcome measures across studies, with very few including measures of service use.

4. Discussion

This systematic review and evidence map aimed to evaluate the effectiveness of interventions for children who stutter and examine whether alternative methods of delivery were as effective as interventions delivered using standard methods. Overall, our main finding is that at a global level, the evidence is concentrated on interventions for preschool children who stutter, using direct approaches. More specifically, the search identified studies that were grouped under two types of intervention approaches: direct intervention and indirect intervention. Two studies compared the LP to a control group, one compared a modified version of the LP to the traditional version (removal of the component that requests self-correction) and two studies compared the RESTART-DCM approach to the LP.

The LP was found to have the best available evidence for children under 6 years of age. It was found to be effective in reducing stuttering in children in all eight studies, with the %SS ranging from 3.8 to 9.4% at baseline to 0.9–3.7 % at follow-up across studies. In those studies that compared the LP to a control group, there was a statistically significant difference between the groups in %SS at follow up. The pooled effect size for these studies showed a reduction of around 4 %SS from pre to post intervention. For the two RESTART-DCM interventions, %SS were reduced from between 5.3–7.9 % at baseline to 1.5–3.1 % at follow up. The RESTART-DCM study was found to have similar outcomes to the LP in the two studies that compared both approaches side by side, however there are fewer studies investigating RESTART-DCM. The body of evidence regarding interventions for older school-aged children who stutter, is limited because there are no RCTs. This review did not report on studies that were below the highest level of evidence.

All eight studies included in this review included the LP but used various alternative methods of delivery with standard LP delivery used as a comparator. Telehealth, webcam and group format were compared to standard delivery (i.e. one-to-one / face-to-face).

Alternative methods of delivery for the LP were shown to be as effective at reducing stuttering as standard delivery. The efficacy of interventions was not reported consistently for all studies but overall, participants in the webcam and group delivery required a smaller number of consultations and less SLP time to complete Stage 1 of the LP, although not significantly different. Importantly, the adapted methods of delivery did not take a significantly greater amount of time. This finding has key implications for the delivery of interventions for those who do not have access to local intervention services due to location or availability. While some studies provided information on the efficacy of the intervention, a challenge of the extant literature is that few studies consider efficiency. This is an important future direction in the field.

Overall, risk of bias was rated as moderate across the eight studies. All studies were at high risk of bias in the 'blinding of participants' domain. This is not surprising given the methodological challenges of blinding participants to intervention allocation in these types of studies. The studies were generally rated at 'low risk' for both the selection and detection bias. It should be noted that six of the eight studies were associated with the authors who originally developed the LP, which has the potential to produce bias. Overall, based on the available evidence we found LP to have the highest level of evidence and rated it Grade A (excellent). The RESTART-DCM model was rated as Grade B (good) for the effectiveness of stuttering interventions.

4.1. Implications for future research

Addressing the evidence gaps identified in this study requires support from researchers and clinicians to ensure efforts to strengthen the evidence-base surrounding interventions for children and adolescence who stutter are informed by the most effective and efficacious approaches. To ensure the relevance, uptake and use of such new evidence, a strategic or prioritised research agenda should be informed by the findings of this and other reviews and co-developed with researchers, service providers and clinicians in speech and language.

To improve the body of evidence through research, we make the following recommendations:

1 Research to date has concentrated on interventions for preschool aged children, with no RCT interventions for school-aged children or adolescents. Future research should focus on interventions for school-aged children and adolescents and include independent replication of current findings.
2 Most studies had small samples sizes and large attrition rates, suggesting that more robust study design is needed (i.e. larger studies with at least 80 % power).
3 Studies should integrate treatment comparison designs, and investigation of the influence of self-selection and environmental influences could be addressed.
4 Studies should respond to our lack of understanding of how-to deliver interventions most efficiently by investigating the relationship between dose and response to intervention and/or follow up individuals over mid to long term to test how well intervention gains are maintained over time.
5 Harmonisation of outcome measures across studies would be valuable in order to make cross study comparisons.
6 Typically treatments are developed and tested with compliant families/individuals who complete the prescribed course. However, families sometimes have complex and competing needs meaning they are not able to complete treatment as prescribed. Research is required to understand how best to address these issues.
7 Studies examining the cost-effectiveness and economic evaluation of interventions are needed to elucidate the most effective and sustainable methods of delivering stuttering interventions for children in the context of publicly provided services.
8 Finally, in the community, children who stutter may have additional speech and language impairments and/or comorbidities such as intellectual disability. Studies that include such children will improve the evidence base for children who have different presentations and more complex needs.

4.2. Limitations

A limitation of this review was the inclusion of studies that were published in English over the past 14 years, in order to retrieve the most up-to-date studies. There were some Phase I and Phase II studies published prior to the dates in this review (e.g. Onslow, Menzies, & Packman, 2001) and the current review builds on this existing body of evidence. In addition, studies of pharmacological interventions in combination with speech interventions were included, albeit none were identified. Whilst historically treatment of stuttering in children and adolescents has been dominated by speech therapy, there is a paucity of evidence regarding response to pharmacological agents. As such, current treatment options with pharmacological intervention may offer synergistic effects and warrants consideration. Due to the small number of studies that could be combined for meta-analysis, we were not able to perform subgroup analysis or investigate potential sources of heterogeneity between studies. Included studies varied in the length and dose of the LP intervention. In the standard LP it took a median of 16 (range 11–23) clinic visits to reach Stage 2. The length of Stage 2 also varied but children were typically monitored for 44–52 weeks. Several studies included in this review were conducted over shorter periods than those in the standard LP. For example, Franken et al. (2005) conducted a trial over 12 weeks and Lattermann et al. (2008) conducted a trial over 16 weeks. Neither study included a maintenance stage. Such diversity in study methods may have produced different outcomes.

5. Conclusions

In conclusion, this systematic review and mapping of the evidence revealed that both direct and indirect interventions are effective in reducing stuttering in preschool children. The direct approach had a higher quantity and quality of evidence, relative to the indirect approach. Several different methods of delivery were identified, including; one-to-one, group, face-to-face, and telehealth. This review did not identify an absolute indication for the most effective method of delivery and there was no evidence that one-to-one (i.e. face-to-face) interventions were any more, or less effective than the alternative methods. Currently, there is insufficient evidence from RCTs to rigorously evaluate interventions for school-aged children and adolescents who stutter. With increasing pressure on health funding, the results of this review may help to guide future research and prioritise precious clinical resources.

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Data availability

Data are available on reasonable request. All data relevant to the study are included in the article or uploaded as online supplementary information.

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Appendix A

Table A1
Search strategy[a].

CINAHL EBSCOhost	**Search Terms**
Concept 1	Stutter* OR Stammer* OR D?sfluence OR D?sfluency OR D?sfluencies OR PWS
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	'Systematic Review' OR Review OR Randomi?ed Control* Trial* 01 Jan 2005 - 20 Jan 2019
MEDLINE Ovid	**Search Terms**
Concept 1	Stutter* OR Stammer* OR D#sfluenc* OR PWS OR exp Stuttering/
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	Systematic Review OR Review Random* Control* Trial*.pt. OR exp Randomized Controlled Trials as Topic/ OR exp Randomized Controlled Trial/ limit to yr="2005 -Current"
PsycEXTRA Ovid	**Search Terms**
Concept 1	Stutter* OR Stammer* OR D#sfluenc* OR PWS
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	Systematic Review or Review or Randomi*Control* Trial*
PsycINFO Ovid	**Search Terms**
Concept 1	Stutter* OR Stammer* OR D#sfluenc* OR PWS
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	Systematic Review or Review or Randomi*Control* Trial*
Embase	**Search Terms**
Concept 1	Stutter* OR Stammer* OR Disfluencies OR Dysfluencies OR Dysfluency OR Disfluency OR PWS
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	Systematic Review OR Review OR Randomi*Control* Trial*
Cochrane	**Search Terms**
Concept 1	Stutter* OR Stammer* OR D?sfluenc* OR PWS OR Stuttering(mesh)
Concept 2	Therap* OR Intervention* OR Treat*
Concept 3	Child* OR Adolescen* OR Adult*
Other Filters	Systematic Review OR Review OR Randomi*Control* Trial* Publication Year from 2005 (not groups)

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Table A1 (*continued*)

World Health Organisation	**Search Terms**
Concept 1	Stutter OR Stammer OR Stuttering OR Stammering OR Disfluencies OR Dysfluencies OR Dysfluency OR Disfluency
Concept (clinicaltrials.gov)	**Search Terms**
Concept 1	Stutter OR Stammer OR Stuttering OR Stammering OR Disfluencies OR Dysfluencies OR Dysfluency OR Disfluency
Concept (Google Scholar)	**Search Terms**
	Stutter OR Stammer OR Stuttering OR Stammering OR Disfluencies OR Dysfluencies OR Dysfluency OR Disfluency OR Systematic Review OR Review OR Randomised control trial

[a] This systematic review was part of a greater project that explored both adult and childhood (Brignell et al., 2020; published elsewhere) interventions separately; however, the same search strategies were used for both with the evidence synthesis being focused on the separate population groups.

Appendix B

Table B1
Studies relevant to the topic of interventions for children who stutter that did not meet the inclusion criteria.

Study	Reason for exclusion
Carey B, O'Brian S, Onslow M, Block S, Jones M, et al. Randomized Controlled Non-Inferiority Trial of a Telehealth Treatment for Chronic Stuttering: The Camperdown Program. Int J Lang Commun Disord. 2010;45(1):108-20.	Participants were aged >18 years
Chesters J, Möttönen R, Watkins KE. Transcranial direct current stimulation over left inferior frontal cortex improves speech fluency in adults who stutter. Brain: A Journal of Neurology. 2018;141(4):1161-71.	Participants were aged >18 years
Cream A, O'Brian S, Jones M, Block S, Harrison E, et al. Randomized Controlled Trial of Video Self-Modeling Following Speech Restructuring Treatment for Stuttering. J Speech Lang Hear Res. 2010;53(4):887-97.	Participants were aged >18 years
de Sonneville-Koedoot C, Bouwmans C, Franken MC, Stolk E (2015) Economic evaluation of stuttering treatment in preschool children: The RESTART-study. J Commun Disord 58:106-118	An economic evaluation of de Sonneville-Koedot 2015
de Veer S, Brouwers A, W E, Tomic W. A Pilot Study of the Psychological Impact of the Mindfulness-Based Stress Reduction Program on Persons who Stutter. European Psychotherapy. 2009;9(1):39-56.	Stuttering/speech is not reported as the outcome in this study
Franklin D, Taylor CL, Hennessey NW, Beilby JM. Investigating factors related to the effects of time-out on stuttering in adults. International Journal of Language & Communication Disorders. 2008;43(3):283-99.	No random assignment of participants to groups
Hewat S, Onslow M, Packman A, O'Brian S. A phase II clinical trial of self-imposed time-out treatment for stuttering in adults and adolescents. Disabil Rehabil. 2006;28(1):33–42.	No random assignment of participants to groups
Jones M, Onslow M, Packman A, O'Brian S, Hearne A, Williams S, Ormond T, Schwarz I (2008) Extended follow-up of a randomized controlled trial of the Lidcombe Program of Early Stuttering Intervention. International Journal of Language & Communication Disorders 43:649-661	A follow-up study of Jones 2005
Ingham RJ, Ingham JC, Bothe AK, Wang Y, Kilgo M. Efficacy of the Modifying Phonation Intervals (Mpi) Stuttering Treatment Program with Adults Who Stutter. Am J Speech Lang Pathol. 2015;24(2):256-71.	Participants were aged >18 years
Iverach L, Jones M, O'Brian S, Block S, Lincoln M, Harrison E, et al. Corrigendum to "The relationship between mental health disorders and treatment outcomes among adults who stutter". Journal of Fluency Disorders. 2009;34(4):301.	Results are reported based on mental health disorders AND treatment outcomes of participants from an existing study
Lincoln M, Packman A, Onslow M. Altered auditory feedback and the treatment of stuttering: A review. Journal of Fluency Disorders. 2006;31(2):71-89.	Not a systematic review
Lu C, Zheng L, Long Y, Yan Q, Ding G, Liu L, et al. Reorganization of brain function after a short-term behavioral intervention for stuttering. Brain Lang. 2017;168:12-22.	Not conducted in English
McAllister J, Gascoine S, Carroll A, Humby K, Kingston M, Shepstone L, et al. Cognitive bias modification for social anxiety in adults who stutter: a feasibility study of a randomised controlled trial. BMJ Open. 2017;7(10):e015601	Stuttering was not confirmed at intake
Menzies RG, O'Brian S, Onslow M, Packman A, St Clare T, et al. An Experimental Clinical Trial of a Cognitive-Behavior Therapy Package for Chronic Stuttering. J Speech Lang Hear Res. 2008;51(6):1451-64.	Participants were aged >18 years
Neumann K, Euler HA, Bosshardt H-G, Cook S, Sandrieser P, Sommer M. The Pathogenesis, Assessment and Treatment of Speech Fluency Disorders. Deutsches Aerzteblatt International. 2017;114(22/23):383-90.	Summarizes some results of a systematic review which was published in German
	Not a systematic review

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Table B1 (*continued*)

Study	Reason for exclusion
Packman A, Onslow M. Investigating optimal intervention intensity with the Lidcombe Program of early stuttering intervention. International Journal of Speech-Language Pathology. 2012;14(5):467-70.	
Rousseau I, Packman A, Onslow M, Harrison E, Jones M (2007) An investigation of language and phonological development and the responsiveness of preschool age children to the LP. J Commun Disord 40:382-397	No random assignment of participants to groups
Sidavi A, Fabus R. A review of stuttering intervention approaches for preschool-age and elementary school-age children. Contemporary Issues in Communication Science & Disorders. 2010;37:14-26.	Not a systematic review

Appendix C

Table C1
Characteristics of included studies.

Study details	Intervention	Participant details	Outcomes assessed	Main results
Arnott et al. (2015) Aim To investigate the efficacy and efficiency of standard delivery of the LP compared with group delivery Country Australia Study Type Non-inferiority RCT Level of Evidence: II Data Collection Audio recordings of conversational speech sample with parent and non-parent questionnaire Funding National Health and Medical Research Council, Australia	Intervention LP - group adapted Comparator Standard LP	Recruitment Via a waiting list of the speech clinic N = 54 (27 intervention / 27 comparator) Attrition 11/54 (20 %) randomised withdrawn due to various reasons Demographics Aged 3;0–5;11 years, 42 male and 12 female Stuttering History: > 6 months since onset Severity: ≥ 2 %SS, observed by SLP	Primary Outcome SLP hours per child to complete stage 1 of the LP Secondary Outcome - %SS - Number of clinic visits and weeks to complete stage 1 - Parent-reported stuttering SR - Parent satisfaction survey Time points/Follow-up Pre-randomisation, 9- and 18-months post-randomisation Response Criteria Three consecutive clinic visits where SLP %SS score is <2 %SS and parent-reported %SS is < 2 %SS	At baseline the mean %SS in the intervention group was 4.0 ± 2.0 (range 0.9–8.5) and 4.4 ± 4.0 (range 0.9–21.5) in the control group. After 9- and 18-months post-randomisation, the mean %SS was reduced to 0.9 ± 0.8, 0.9 ± 0.7 and 1.1 ± 1.3, 0.6 ± 0.4, respectively. Analysis of covariance indicated no evidence of difference between the intervention and control groups at 9 months ($p = 0.80$) or 18 months ($p = 0.30$) post-randomisation, hence the group delivery results in non-inferior %SS outcomes. Parent-reported stuttering SR showed no difference between the intervention and control groups at 9 months ($p = 0.59$) and 18 months ($p = 0.87$) post-randomisation, again supporting that group delivery of the LP is non-inferior to one-to-one delivery.
Bridgman et al. (2016) Aim To compare outcomes of clinic and webcam delivery of the LP intervention for early stuttering Country Australia Study Type Open plan, parallel group, RCT Level of Evidence: II Data Collection Audio recordings of speech sample by parents at the home, parent questionnaire Funding National Health and Medical Research Council, Australia	Intervention LP – webcam adapted Comparator Standard LP	Recruitment Via the waiting list of the speech clinic and local community health services N = 49 (25 intervention / 24 comparator) Attrition 5/49 (10 %) randomised did not complete stage 1 at 9 months; 16/49 (32 %) randomised, did not complete stage 1 at 18 months Demographics Aged 3;0–5;11 years, gender not reported Stuttering History: > 6 months since onset Severity: not specified	Primary Outcome - %SS at 9 months post-randomisation - Number of SLP consultations to complete stage 1 of the LP Secondary Outcome - %SS at 18 months post-randomisation - Parent-reported stuttering SR - Number of weeks to achieve stage 2 - Mean duration of consultations - Parent satisfaction survey Time points/Follow-up Pre-randomisation, 9- and 18-months post-randomisation	A total of 37 children completed stage 1 of the LP, 14 in the intervention group and 13 in the control group. At baseline the %SS were similar between the two groups (intervention: 3.8 ± 2.8; control: 4.0 ± 2.9) and there was also no significant difference between groups at 9 months post-randomisation (1.7 ± 2.0 vs. 1.0 ± 1.0, p 0.16, 95% CI [-0.3, 1.7]) and 18 months post-randomisation (0.8 ± 0.9 vs. 0.7 ± 0.5, $p = 0.72$, 95% CI [-0.4, 0.6]). Similarly there were no differences in parent-reported SR at pre-randomisation, 9-

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Table C1 (*continued*)

Study details	Intervention	Participant details	Outcomes assessed	Main results
				and 18-months post-randomisation.
Donaghy et al. (2015) Aim To investigate the role of the specific contingency RQSC in the LP Country Australia Study Type RCT Level of Evidence: II Data Collection Audio recordings of speech samples collected within and outside of the clinic Funding Partly by the National Health and Medical Research Council, Australia	Intervention LP – RSQC adapted Comparator Standard LP	Recruitment Via word of mouth and study advertisements placed in local clinics, community health centres and preschools N = 38 (19 intervention / 19 comparator) Attrition 4/38 (10 %) randomised, 4 children were not eligible and excluded, 1 withdrew but was included in analysis by log-rank test in survival analysis Demographics Aged 2;10 - 5;10 years, 26 male and 12 female Stuttering History: ≥ 6 months since onset, no previous intervention with LP of child or their siblings Severity: ≥3 %SS	Primary Outcome Number of clinic visits and number of weeks to achieve a 50 % reduction in %SS Time points/Follow-up Pre-randomisation, intervention response achieved, and intervention response maintained for 3 weeks Response Criteria Achievement of a 50 % reduction in %SS, which is sustained (or further reduced) for 3 weeks	A total of 33 participants completed the study, achieving a 50 % reduction in stuttering that was stabilised (or further reduced) for 3 weeks. At baseline, the intervention group mean %SS was 7.1 ± 4.8 (range 3.2–19.5) and 7.0 ± 2.8 (range 3.5–12.8) in the control group ($p = 0.91$). A 72% (2 ± 1.7, range 0.5-7.4) and 69% (2.2 ± 1.1, range 0.6-4.6) reduction in %SS was achieved in the intervention and control groups, respectively which was not significant ($p = 0.72$). The parent SR were not statistically different between groups at baseline or at intervention response. The control group achieved a 68 % reduction in typical stuttering SR and 67 % in the intervention group ($p = 0.51$).
Franken et al. (2005) Aim To compare the effectiveness of the LP with the DCM Country Netherlands Study Type RCT Level of Evidence: II Data Collection Audio recordings of speech sample by parents at the home, questionnaires Funding Friends of Sophia Hospital Foundation, Netherlands	Intervention Standard LP Comparator DCM	Recruitment Via SLPs and GPs N = 30 (15 intervention / 15 comparator) Attrition 4/30 (13 %) did not complete intervention, 3/30 (10 %) did not collect required data Demographics Aged <6;0 years, 17 male and 6 female Stuttering History: ≥ 6 months since onset Severity: ≥2 %SS, rated by parent and SLP	Primary Outcome %SS Secondary Outcome - Stuttering SR - Bristol Stammering questionnaire Time points/Follow-up Pre-intervention and 3 months post-intervention Response Criteria Stuttering frequency <1 % SS, stuttering SR in the past 3 weeks was 1–2 %SS and parent confident in maintain procedures at home	The stuttering frequency for the LP intervention decreased from 7.2 ± 2.0 %SS to 3.7 ± 2.1 %SS. The DCM intervention group decreased from 7.9 ± 7.1 %SS to 3.1 ± 2.1 %SS. A mixed-design ANOVA showed a significant effect of time (pre- and post-intervention) $F(1, 21) = 15.18$, ($p < 0.01$), but no effect of intervention and no effect of intervention x time interaction ($p > 0.10$). Parent and therapist ratings for stuttering SR were similar, with an effect of time for the parent, $F(1, 21) = 85.50$, ($p < 0.01$) and for the therapist, $F(1, 21) = 73.73$, ($p < 0.01$). There was no effect that involved intervention ($p > 0.10$).
Jones et al. (2005) Aim To evaluate the efficacy of the LP of early stuttering intervention by comparison to a control group Country New Zealand Study Type Pragmatic, open plan, parallel group, RCT Level of Evidence: II Data Collection Audio recordings of speech samples collected by parents outside of the clinic	Intervention Standard LP Comparator Delayed treatment - parents were advised they could receive intervention during the trial at other clinics, but not the LP	Recruitment Via presentation at the study speech clinics for intervention N = 54 (29 intervention / 25 comparator) Attrition 7/54 (13 %) randomised did not complete intervention Demographics Aged 3;0–6;0 years, 42 male and 12 female Stuttering History: ≥ 6 months since onset Severity: ≥2 %SS	Primary Outcome %SS Time points/Follow-up Pre-intervention and 9 months post-randomisation Response Criteria Clinical difference at 9 months after randomisation of 1 %SS	The mean baseline SR of stuttering in the intervention and control groups were similar, 6.4 ± 4.3 %SS and 6.8 ± 4.9 %SS respectively. A highly significant decrease in the %SS at 9 months after randomisation was reported between the intervention and control groups (95 % CI [0.8 %–3.9 %] $p = 0.003$). The result was also similar after adjusting for intervention site, baseline SR, age, sex and family history of stuttering. A logistic regression model

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Table C1 (*continued*)

Study details	Intervention	Participant details	Outcomes assessed	Main results
Funding Nil				exploring the proportion of children with <1 %SS at 9 months post-randomisation, revealed the proportion was higher in the intervention group when adjusted for baseline SR score (95 % CI [0.03–0.63] $p = 0.01$).
Lattermann et al. (2008) Aim To investigate whether the short-term reduction of stuttered speech following intervention with the LP is significantly greater than natural recovery in German-speaking preschool children Country Germany Study Type RCT Level of Evidence: II Data Collection Audio recordings of speech samples collected within and outside of the clinic Funding Partly by the Rotary Club, Wiesbaden, Germany	Intervention Standard LP Comparator Delayed treatment - placed on a waiting list and assigned intervention after trial was completed	Recruitment Via media advertising N = 46 (23 intervention / 23 comparator) Attrition 1/46 (2.2 %) recruited, 1 did not complete required data post treatment Demographics Aged 3;0–5;11 years, 42 male and 4 female Stuttering History: ≥ 6 months since onset Severity: ≥3 %SS	Primary Outcome %SS in the home and in the clinic Time points/Follow-up Pre-intervention and 4-months post-randomisation Response Criteria Clinical difference at 4 months post-randomisation in %SS	Outside the clinic: The baseline %SS mean was 9.5 ± 5.5 %SS (range 2.2–26.6) in the intervention group and 7.5 ± 4.7%SS (range 1.8–20.2) in the control group. At 4 months post-randomisation the intervention group was 2.6 ± 1.9%SS (range 0.0–7.3) and 6.2 ± 4.7%SS (range 0.7–17.4) in the control group. The mean decrease in the intervention group was 6.8%SS and an average reduction of 70.3% in dysfluency rate. The control group had an average decrease in %SS score of 3.6% and a reduction in dysfluency rate by a mean of 17.6%. In the control group 13 children had a natural decrease in their stuttering frequencies, whereas 9 children had increases. Within the clinic: The baseline mean %SS was 9.4 ± 4.5 (range 2.3–19.7) in the intervention group and 10.0 ± 7.7 (range 3.6–39.6) in the control group. At 4 months post-randomisation the intervention group reduced their %SS by 6.8% (2.6 ± 1.7) and the control group by 1.6% SS (6.4 ± 3.7). Overall the children receiving the LP reduced their dysfluency rate by 70.6% compared to 25.4% in the control group. Fifteen children in the control group showed a natural decrease in their stuttering and 7 displayed increases. ANCOVA analysis showed that the difference between the groups was significant.
Lewis et al. (2008) Aim To evaluate the efficacy of telehealth delivery of the LP of early stuttering intervention compared with a control group Country Australia Study Type Open plan, parallel group, RCT Level of Evidence: II Data Collection	Intervention LP – telephone adapted Comparator Delayed treatment - parents were placed on the wait list and offered intervention after the trial was complete	Recruitment Advertised in the local press N = 22 (9 intervention / 13 comparator) Attrition 4/22 (18 %) randomised were withdrawn due to non-compliance Demographics Aged 3;0–4;6 years, 14 male and 8 female Stuttering History: > 6 months since	Primary Outcome %SS Secondary Outcome - Intervention time - Parent satisfaction Time points/Follow-up 1 week prior to randomisation and 9 months post-randomisation Response Criteria A > 80 % reduction in %SS from time of randomisation to 9 months.	The baseline mean, pooled % SS scores were 6.7 in the intervention and 4.5 in the control group. The mean 9-month post-randomisation scores for the intervention group was 1.1 and 1.9 %SS for the control group. ANCOVA showed a 69 % decrease in the frequency of stuttering in the intervention group at 9 months post-randomisation, compared to

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Table C1 (*continued*)

Study details	Intervention	Participant details	Outcomes assessed	Main results
Audio recordings of speech samples by parents in the home, parent questionnaire Funding National Health and Medical Research Council, Australia		onset, no previous or current intervention Severity: not defined		the control group (95 % CI [13 %–89 %] $p = 0.04$). The adjusted intervention effect (gender, age, family history and baseline SR) estimated a 73% decrease (95% CI [25%-90%] $p = 0.02$) in stuttering. In the intervention group, 6 of the 8 children achieved the response criterion at 9 months post-randomisation and 2 of the 10 children in the control through natural recovery ($p = 0.054$).
de Sonneville-Koedoot et al. (2015)				
Aim To compare the effectiveness of direct versus indirect stuttering intervention in preschool children Country Netherlands Study Type Parallel group, RCT Level of Evidence: II Data Collection Audio recordings of speech sample with a parent in and out of the home and with a non-parent away from the home, questionnaires Funding The Netherlands Organization for Health Research and Development, Netherlands	Intervention Standard LP Comparator RESTART-DCM	Recruitment Via a registration list at 20 participating study sites N = 199 (100 intervention / 99 comparator) Attrition 23/199 (11 %), 22 did not complete intervention, 1 control was missing recordings Demographics Aged 3;0–6;3 years, 138 male and 61 female Stuttering History: ≥ 6 months since onset Severity: ≥2 %SS	Primary Outcome Percentage of non-stuttering children at 18 months post-randomisation Secondary Outcome At 3, 6, 12 and 18 months: - %SS - Parent-reported stuttering SR - Parent evaluation of child's HRQoL At baseline and 18 months: - Speech attitude of child (KiddyCAT) - Emotional and behavioural assessment (CBCL) At 18 months: - SLP and child rated stuttering SR Time points/Follow-up Pre-randomisation, 3, 6, 9 and 18-months post-randomisation Response Criteria ≤1.5 %SS at 18 months post-randomisation	In total, 76.5 % (65/85, 95 % CI [66.4–84.2]) of children in the intervention group, achieved a %SS score ≤ 1.5 by 18 months post-randomisation and were classified as non-stuttering. Compared to the children in the control group (71.4%, 65/91, 95% CI [61.4-79.7]). This was not statistically different ($p = 0.45$). The effect of intervention was non-significant for %SS from baseline to 18 months, however a significant interaction between time and intervention type was detected ($p = 0.008$). This result indicates that the %SS differed between groups at different time points and the effect of time indicates that the %SS in both the intervention and control groups decreased significantly over time ($p = 0.002$). In both groups most of the decrease in %SS occurred within the first 3 months. There was no effect of intervention on outcomes for EQ-VAS and KiddyCAT.

LP: Lidcombe Program; RESTART: Rotterdam Evaluation Study of Stuttering Therapy; DCM: Demands and Capacities Model; RQSC: Request self-correction of stuttered speech.

Appendix D

Table D1
Description of interventions and methods of service delivery for included studies.

Intervention	Approach	Setting	Method of Delivery[a]	Description
LP - Standard	Direct	Clinic	One-to-one Face-to-face	The LP is a commonly used stuttering treatment for preschool children. It is based on behavioural reinforcement, whereby the child is praised for stutter-free speech and gently corrected when they stutter. Stutter-free speech generalises to the child's everyday conversations over time. This treatment approach consists of two stages and is administered by parents daily within the child's natural environment.

(*continued on next page*)

Table D1 (*continued*)

Intervention	Approach	Setting	Method of Delivery[a]	Description
LP – Group adapted	Direct	Clinic	Face-to-face Group	• Stage 1: The parent and child are required to attend the clinic on a weekly basis where the parent is trained by the SLP to deliver the intervention. Intervention is delivered in 15-minute blocks every day through activities that are gradually moved from highly structured to unstructured conversation. • Stage 2: Once the stuttering has been minimised, the child progresses to stage 2, where intervention is gradually withdrawn. The objective of this stage is for the child to maintain intervention effects with reduced support from their parent. The Standard LP intervention is adapted to group delivery: • Regular (weekly) scheduled SLP consultations in groups of child-parent dyads, instead of one-to-one clinic visits. • The target is to maintain up to three child-parent pairs per visit. • Although not encouraged, parents can move between groups at different days/ times of the week. • Groups constantly change their composition and comprised of parents with varying Lidcombe experience. The basic structure of a group clinic visit involves: free play, parent report and discussion of stuttering SR, practice intervention during structured and unstructured conversations and reflection and recommendations for the coming week.
LP – Webcam adapted	Direct	Remote	One-to-one Face-to-face Telehealth[b]	The Standard LP is adapted to webcam delivery: • Regular (weekly) scheduled SLP consultations via webcam, instead of clinic visits. • Children and parents receive training within their homes using a personal computer, webcam and a video calling program. • Weekly scheduled webcam consultations are 45−60 min duration, as per standard protocol.
LP – Telephone adapted	Direct	Remote	One-to-one Telehealth[b]	The Standard LP is adapted to telehealth delivery: • Regular (weekly) scheduled SLP consultations over the telephone, instead of clinic visits. • Video demonstrations of SLPs conducting intervention and detailed discussions, replace one-to-one demonstration of intervention in the clinic. • Additional support via telephone/e-mail is provided to parents. • %SS are made from audio-recordings, rather than face-to-face and in real-time. • Parent training via audio-recordings and telephone, rather than face-to-face. • The SLP observes parents applying intervention via audio-recordings, rather than face-to-face.
LP – RQSC adapted	Direct	Clinic	One-to-one Face-to-face	The Standard LP is adapted by removing the RQSC of stuttered speech contingency.
DCM	Indirect	Clinic	One-to-one Face-to-face	The DCM is a stuttering intervention for preschool children. It aims to modify the child's communicative environment using a combination of interaction and family strategies to create a fluency-inducing environment for the child. This intervention simultaneously builds the child's capacity across domains to enhance fluency. The intervention is based on the premise that the factors that trigger stuttering are different for every child. • Intervention is the same for every child while the specific strategies selected and implemented are individualised. • Families are required to attend the clinic on a weekly basis until the parents have demonstrated competence implementing family and interaction strategies and the child has reached a level of stutter-free speech that satisfies the parents. Once an acceptable level of stutter-free speech is established, children complete a review period to ensure they maintain intervention effects.

LP: Lidcombe Program; SLP: Speech Language Pathologist; RQSC: Request for Self-Correction of stuttering; DCM: Demands and Capacity.
Webcam delivery where the SLP delivers treatment over the internet via a computer, using a live video calling program (Skype) and the SLP and client can see and speak to one another; Telehealth delivery where the SLP delivers treatment via information technology and telecommunication. The treatment is adapted to telephone delivery.

[a] One-to-one: Treatment that is delivered by the SLP directly to the client. This is also known as individual treatment; Group: Treatment that is delivered by the SLP directly to two or more clients simultaneously; Face-to-face: Treatment that is delivered by the SLP to the client whilst in direct contact with one another in a clinic setting.

[b] Telehealth includes a collection of technologies used to deliver medical care. This includes:

Appendix E

Table E1
Summary of findings of change in stuttering frequency.

Study	Intervention	Pre-treatment Mean %SS (SD)	+ 3–4 months Mean %SS (SD)	+ 9-months Mean %SS (SD)	+ 12-months Mean %SS (SD)	+ 18-months Mean %SS (SD)
Arnott et al. (2014)	LP – group adapted	3.9 (2.0)	–	0.9 (0.8)	–	0.9 (0.7)
	Standard LP	4.4 (4.0)	–	1.1 (1.3)	–	0.6 (0.4)
Bridgman et al. (2016)	LP – webcam adapted	3.8 (2.8)	–	1.7 (2.0)	–	0.8 (0.9)
	Standard LP	4.0 (2.9)	–	1.0 (1.0)	–	0.7 (0.5)
Donaghy et al. (2015)	LP - RQSC adapted	7.1 (4.8)	2.0 (1.7)[b]	–	–	–
	Standard LP	7.0 (2.8)	2.2 (1.1)[b]	–	–	–
Franken et al. (2005)	Standard LP	7.2 (2.0)	3.7 (2.1)	–	–	–
	DCM	7.9 (7.1)	3.1 (2.1)	–	–	–
Jones et al. (2005)	Standard LP	6.4 (4.3)	–	1.5 (1.4)	–	–
	Delayed Treatment	6.8 (4.9)	–	3.9 (3.5)	–	–
Lattermann et al. (2008)	Standard LP	9.4 (4.5)	2.6 (1.7)	–	–	–
	Delayed Treatment	10.0 (7.7)	6.4 (3.7)	–	–	–
Lewis et al. (2008)	LP - telephone adapted	6.7 (3.0)[a]	–	1.1 (0.5)[a]	–	–
	Delayed Treatment	4.5 (3.5)[a]	–	1.9 (1.0)[a]	–	–
de Sonneville-Koedoot et al. (2015)	Standard LP	6.2 (4.4)	2.5[a]	2.8[a]	1.4[a]	1.2 (2.1)
	RESTART-DCM	5.3 (4.3)	3.2[a]	2.2[a]	1.5[a]	1.5 (2.1)

LP: Lidcombe Program; DCM: Demands and Capacities Model; RESTART: Rotterdam Evaluation Study of Stuttering Therapy; RQSC: request for self-correction of stuttered speech; %SS: percent syllables stuttered.

[a] Value estimated from graph.

[b] Time to 50 % reduction in %SS (~ 12 weeks).

Appendix F

Table F1
Risk of bias assessment within and across all studies included in final synthesis.

Bias Category								
Study	Random sequence generation (Selection)	Allocation concealment (Selection)	Blinding of participants and personnel (Performance)	Blinding of outcome assessment (Detection)	Incomplete outcome data (Attrition)	Selective outcome reporting (Reporting)	Problems not covered elsewhere (Other)	Overall Quality
Arnott et al. (2014)	+	+	-	+	?	+	?	Low
Bridgman et al. (2016)	+	+	-	+	?	+	?	Low
de Sonneville-Koedoot et al. (2015)	+	?	-	?	?	+	+	Moderate
Donaghy et al. (2015)	+	+	-	?	+	-	?	Moderate
Franken et al. (2005)	?	?	-	-	?	+	-	Moderate
Jones et al. (2005)	+	+	-	+	?	+	-	Moderate
Lattermann et al. (2008)	?	-	-	?	?	+	?	Moderate
Lewis et al. (2008)	+	+	-	+	?	+	+	Low

(+) Low risk of bias; (?) Unclear risk of bias; (-) High risk of bias.

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