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Interventions for childhood apraxia of speech (Review)

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[Intervention Review]

Interventions for childhood apraxia of speech

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ABSTRACT

Background

Childhood apraxia of speech (CAS) affects a child's ability to produce sounds and syllables precisely and consistently, and to produce words and sentences with accuracy and correct speech rhythm. It is a rare condition, affecting only 0.1% of the general population. Consensus has been reached that three core features have diagnostic validity: (1) inconsistent error production on both consonants and vowels across repeated productions of syllables or words; (2) lengthened and impaired coarticulatory transitions between sounds and syllables; and (3) inappropriate prosody (ASHA 2007). A deficit in motor programming or planning is thought to underlie the condition. This means that children know what they would like to say but there is a breakdown in the ability to programme or plan the fine and rapid movements required to accurately produce speech. Children with CAS may also have impairments in one or more of the following areas: non-speech oral motor function, dysarthria, language, phonological production impairment, phonemic awareness or metalinguistic skills and literacy, or combinations of these. High-quality evidence from randomised controlled trials (RCTs) is lacking on interventions for CAS.

Objectives

To assess the efficacy of interventions targeting speech and language in children and adolescents with CAS as delivered by speech and language pathologists/therapists.

Search methods

We searched CENTRAL, MEDLINE, Embase, eight other databases and seven trial registers up to April 2017. We searched the reference lists of included reports and requested information on unpublished trials from authors of published studies and other experts as well as information groups in the areas of speech and language therapy/pathology and linguistics.

Selection criteria

RCTs and quasi-RCTs of children aged 3 to 16 years with CAS diagnosed by a speech and language pathologist/therapist, grouped by treatment types.

Data collection and analysis

Two review authors (FL, AM) independently assessed titles and abstracts identified from the searches and obtained full-text reports of all potentially relevant articles and assessed these for eligibility. The same two authors extracted data and conducted the 'Risk of bias' and GRADE assessments. One review author (EM) tabulated findings from excluded observational studies (Table 1).

Main results

This review includes only one RCT, funded by the Australian Research Council; the University of Sydney International Development Fund; Douglas and Lola Douglas Scholarship on Child and Adolescent Health; Nadia Verrall Memorial Scholarship; and a James Kentley Memorial Fellowship. This study recruited 26 children aged 4 to 12 years, with mild to moderate CAS of unknown cause, and compared two interventions: the Nuffield Dyspraxia Programme-3 (NDP-3); and the Rapid Syllable Transitions Treatment (ReST). Children were allocated randomly to one of the two treatments. Treatments were delivered intensively in one-hour sessions, four days a week for three weeks, in a university clinic in Australia. Speech pathology students delivered the treatments in the English language. Outcomes were assessed before therapy, immediately after therapy, at one month and four months post-therapy. Our review looked at one-month post-therapy outcomes only.

We judged all core outcome domains to be low risk of bias. We downgraded the quality of the evidence by one level to moderate due to imprecision, given that only one RCT was identified. Both the NDP-3 and ReST therapies demonstrated improvement at one month post-treatment. A number of cases in each cohort had recommenced usual treatment by their speech and language pathologist between one month and four months post-treatment (NDP-3: 9/13 participants; ReST: 9/13 participants). Hence, maintenance of treatment effects to four months post-treatment could not be analysed without significant potential bias, and thus this time point was not included for further analysis in this review.

There is limited evidence that, when delivered intensively, both the NDP-3 and ReST may effect improvement in word accuracy in 4- to 12-year-old children with CAS, measured by the accuracy of production on treated and non-treated words, speech production consistency and the accuracy of connected speech. The study did not measure functional communication.

Authors' conclusions

There is limited evidence that, when delivered intensively, both the NDP-3 and ReST may effect improvement in word accuracy in 4- to 12-year-old children with CAS, measured by the accuracy of production on treated and non-treated words, speech production consistency and the accuracy of connected speech. The study did not measure functional communication. No formal analyses were conducted to compare NDP-3 and ReST by the original study authors, hence one treatment cannot be reliably advocated over the other. We are also unable to say whether either treatment is better than no treatment or treatment as usual. No evidence currently exists to support the effectiveness of other treatments for children aged 4 to 12 years with idiopathic CAS without other comorbid neurodevelopmental disorders. Further RCTs replicating this study would strengthen the evidence base. Similarly, further RCTs are needed of other interventions, in other age ranges and populations with CAS and with co-occurring disorders.

PLAIN LANGUAGE SUMMARY

One well-controlled study shows some evidence of effect of two interventions for childhood apraxia of speech (CAS)

Review question

What treatments help to improve the speech and language of children and adolescents with childhood apraxia of speech (CAS).

Background

Children with CAS find it difficult to produce sounds and syllables consistently and precisely in order to speak words and sentences with clarity and correct speech rhythm. As a result, children with CAS can be hard to understand with potential for negative impacts on school achievement and peer friendships. CAS affects around 0.1% of the general population. This review collates the research evidence to identify the most effective therapies for children with CAS.

Search date

The evidence is current to 6 April 2017.

Study characteristics

We found one study with 26 children aged 4 to 12 years with CAS. The children had mild to severe CAS without a known cause. Children were allocated randomly (using a method like coin tossing) to one of two treatments: the Nuffield Dyspraxia Programme - Third Edition (NDP-3); and the Rapid Syllable Transition treatment (ReST). Both therapies were delivered intensively in one-hour sessions, four days a week for three weeks. The treatments were delivered by speech pathology students in a university clinic. Outcomes

were assessed before therapy, immediately after therapy, at one month and four months post-therapy. Our review looked at one-month post-therapy outcomes only.

Study funding sources

The included study was funded by the Australian Research Council; the University of Sydney International Development Fund; Douglas & Lola Douglas Scholarship on Child and Adolescent Health; Nadia Verrall Memorial Scholarship; and a James Kentley Memorial Fellowship.

Key results

Further studies replicating these findings would strengthen available evidence.

The study provides limited evidence that the NDP-3 may improve the accuracy of production on treated items and the accuracy of connected speech. There is limited evidence that the NDP-3 has a negligible effect on speech production consistency, and the ReST a negligible effect on accuracy of production on non-treated words. The study did not measure functional communication.

Quality of the evidence

The included study was a randomised controlled trial with an overall low risk of bias. We downgraded the quality of the evidence by one level to moderate, due to imprecision, given that only one RCT was identified.

Recommendations

There is limited evidence that the NDP-3 or ReST may be helpful for children with CAS of unknown origin, aged 4 to 12 years, without other co-occurring conditions. We were not able to find out whether one of these treatment was better than the other, or whether either was better than no treatment or treatment as usual. There is currently no available evidence for other treatments.

Further RCTs - including studies comparing treatments to a no-treatment (wait-list) control group - would strengthen the evidence base. Further research is also needed for children with CAS and other disorders or diagnoses.

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [Explanation]

Nuffield Dyspraxia Programme - Third Edition (NDP-3) versus Rapid Syllable Transition Treatment (ReST) for Childhood Apraxia of Speech

Patient or population: children aged 4 to 12 years with CAS of unknown cause
Settings: University of Sydney Communication Disorders Treatment and Research Clinic
Intervention: NDP-3
Comparison: ReST

Outcomes	Summary of MD findings	Absolute MD	Number of participants (studies)	Quality of the evidence (GRADE)	Comments
Primary outcomes					
Accuracy of production on treated items Measured by: counting the number of real words produced correctly (/x) Follow-up: pre-intervention to 1 month post-intervention	NDP-3 MD of 36.0 was greater than the ReST MD of 33.9	2.1	26 (1 trial)	⊕⊕⊕◯ **Moderate**[a]	-
Accuracy of production on non-treated items Measured by: counting the number of real words produced correctly (/x) Follow-up: pre-intervention to 1 month post-intervention	ReST MD of 18.3 was minimally greater than the NDP-3 MD of 18.2	0.1	26 (1 trial)	⊕⊕⊕◯ **Moderate** [a]	-
Secondary outcomes					

Speech production consistency Measured by: calculating the number of inconsistent productions of 25 words produced 3 times using the DEAP inconsistency subtest[b] Follow-up: pre-intervention to 1 month post-intervention	NDP-3 MD of 11.1 was 0.2 greater than the ReST MD of 10.9	26 (1 trial)	⊕⊕⊕◯ **Moderate** [a]	-
Accuracy of connected speech Measured by: counting the number of correct imitations of 3 word phrases (/x) Follow-up: pre-intervention to 1 month post-intervention	NDP-3 MD of 14.3 was 2.8 greater than the ReST MD of 11.5	26 (1 trial)	⊕⊕⊕◯ **Moderate** [a]	-

GRADE Working Group grades of evidence
High quality: we are very confident that the true effect lies close to that of the estimate of the effect.
Moderate quality: we are moderately confident in the effect estimate; the true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different.
Low quality: our confidence in the effect estimate is limited; the true effect may be substantially different from the estimate of the effect.
Very low quality: we have very little confidence in the effect estimate; the true effect is likely to be substantially different from the estimate of effect

CAS: childhood apraxia of speech; **DEAP**: Diagnostic Evaluation of Articulation and Phonology; **MD**: mean difference; **NDP-3**: Nuffield Dyspraxia Programme - Third Edition; **ReST**: Rapid Syllable Transition Treatment (ReST) for Childhood Apraxia of Speech

[a] We downgraded the quality of evidence by one level, to moderate, for imprecision, as there was only one study for comparison.
[b] Note, a decrease in inconsistency is a positive outcome.

BACKGROUND

Description of the condition

Childhood apraxia of speech (CAS) affects a child's ability to produce speech sounds and syllables in the right order, and to speak words and sentences with accuracy and correct speech rhythm. Over sixty years ago, Morley 1954 provided a seminal paper documenting a series of speech characteristics in children that resembled the speech production disorder of adults with acquired apraxia of speech, and the diagnosis of CAS was born. CAS is a rare condition, affecting only around 0.1% of the general population (Morley 1972; Yoss 1975). CAS is more prevalent within particular medical subgroups, however, and particularly penetrant in certain genetic syndromes (e.g. Fedorenko 2016; Mei 2017).

Historically, synonyms such as verbal dyspraxia and developmental apraxia of speech have been used. The most commonly used terms today are CAS and developmental verbal dyspraxia (DVD), with the latter used largely in the UK context (RCSLT 2011). We use the term CAS consistently throughout this review.

A deficit in motor programming or planning is thought to underlie CAS; that is, children know what they would like to say but there is a breakdown in the ability to programme or plan the required movements to accurately produce speech. The current approach to diagnosis of CAS is expert-based perception of speech symptoms (Maas 2012a). There is consensus amongst speech and language pathologists (SLPs), also known as speech and language therapists (SLTs), that three core features of CAS have diagnostic validity: (1) inconsistent error production on both consonants and vowels across repeated productions of syllables or words; (2) lengthened and impaired coarticulatory transitions between sounds and syllables; and (3) inappropriate prosody (ASHA 2007).

In addition to the core features of CAS, children may also have co-occurring impairments affecting non-speech oral motor function, language, phonemic awareness/meta-linguistics and literacy (ASHA 2007). Younger children typically present with more severe forms of the disorder, with improvement noted over time for both idiopathic CAS (Davis 2005; Jacks 2006) and individuals with CAS associated with genetic syndromes (Morgan 2017; Morgan 2018). It is not currently known how age, severity or underlying aetiology impact upon CAS treatment response or outcome.

There are no epidemiological data on the prevalence of CAS, although it occurs infrequently in comparison with other forms of developmental speech disorder such as articulation disorder and phonological disorder, which occur in around 3.5% of preschool children (Eadie 2015). A population-based estimate suggests that CAS occurs in one child per 1000 (0.1%) (Morley 1972; Yoss 1975), and is found in 3.4% to 4.3% of the children referred to clinics for speech disorder management (Delaney 2004). The diagnosis of CAS can apply to children who have a specific impairment in speech with other neurodevelopmental functions relatively more preserved (e.g. borderline or typical non-verbal cognition). Historically most cases were referred to as 'idiopathic', given limited aetiological knowledge of the condition (Morgan 2008). In recent times, however, novel insights have been gained into the genetic and neurobiological bases of CAS (Eising 2018). Variations in an increasing number of single genes have been associated with CAS (Eising 2018; Turner 2015), with the most replicated finding being disruption of the Forkhead box protein P2 or FOXP2 (Lai 2001; Morgan 2017; Vargha-Khadem 2005). Beyond single gene causes, CAS has also been associated with copy number variant syndromes, such as 16p11.2 deletion syndrome (Fedorenko 2016; Mei 2017), Koolen de Vries Syndrome (Morgan 2018), 6q25.3 deletion syndrome (Peter 2017), 7q11.23 duplication syndrome (Velleman 2011), and other genetic conditions such as Floating Harbour syndrome (White 2010). Further to genetic causes, other medical conditions associated with CAS include metabolic disorders (e.g. galactosaemia; Shriberg 2011) or epilepsy disorders (e.g. Liégeois 2012). In relation to neurobiology or brain function, there is inconsistency as regards the key brain regions and networks disrupted in CAS, with neuroimaging studies reporting both cortical and subcortical anomalies (Liégeois 2012; Liégeois 2014; Liégeois 2016).

Description of the intervention

A range of CAS treatment approaches with differing theoretical standpoints have been reported. These studies are almost exclusively in the form of uncontrolled case studies or case series. Therapeutic approaches for CAS can be grouped into the following three areas.

1. **Motor-based approaches**. These therapies are based on principles of motor learning (see Maas 2008 for a review); for example, traditional articulation-based drill therapy (Velleman 1994), the Nuffield Dyspraxia Programme (Williams 2004), the Rapid Syllable Transitions Treatment (Ballard 2010), rate control therapy (Rosenthal 1994), the PROMPT System (**P**rompts for **R**estructuring **O**ral **M**uscular**P**honetic **T**argets) (Chumpelik 1984; Dale 2013), melodic intonation therapy (Helfrich-Miller 1994), adapted cueing technique (Klick 1985), and integral stimulation or dynamic temporal and tactile cueing (Maas 2012a; Strand 2006). Motor-based therapy can also include non-speech oro-motor techniques; for example, oral form recognition training (Kingston 1987) and orofacial myofunctional therapy (Ray 2003). Motor-based therapy can also be instrumentally based, such as delayed auditory feedback (Lozano 1978), electropalatography (Carter 2004; Lundeborg 2007), and ultrasound (Preston 2013).

2. **Linguistic approaches**. Linguistic therapies address language impairments that can co-occur in children with CAS. Examples of linguistic approaches include programmes to address phonological speech production or awareness (McNeill 2009).

3. **Multi-modal communication approaches**. These approaches seek to support verbal communication. Methods can

address specific communication messages or features, such as Aided AAC (augmentative and alternative communication) Modelling (Binger 2007), or use of technological devices (Bornman 2001; Cumley 1999).

How the intervention might work

Below, we describe the ways in which the aforementioned approaches (described under Description of the intervention) might work.

1. **Motor-based approaches**. These methods use principles of motor learning, such as emphasizing a high number of successful repetitions of a task, using stimuli with high complexity, and a period of teaching followed by practice where cues and feedback are faded. Such approaches are reported to facilitate maintenance and generalisation in children with CAS (Maas 2008; Maas 2014).
2. **Linguistic approaches**. These methods are focused on the semantics, phonology or grammar of language, and not on motor speech production per se. For example, a linguistic approach may include phonological contrast therapy, where children are taught how to abstract speech sound rules for the specific language(s) they speak (Dodd 2008). Another example of a linguistic approach is core vocabulary therapy, which focuses on shaping children's word approximations whilst expanding their expressive and receptive vocabulary (Crosbie 2005).
3. **Multi-modal communication approaches**. These methods are used for children who are minimally verbal to help them communicate and reduce the frustration associated with their speech disability. Devices may include a computer, phone or tablet with applications to help children produce words, phrases and sentences. Other methods involve gesture, sign language or use of visual picture boards.

Why it is important to do this review

There is a need for clinicians and parents to be aware of the most efficacious treatments for children with CAS. To date, studies in the field are largely non-RCT (randomised controlled trials), single case series or case-control studies that are generally positive in stating improvements in speech post-therapy across motor (e.g. Baas 2008; Ballard 2010; Edeal 2011; Hall 1989; Kadis 2014; McCabe 2014; Strand 2000; Strand 2006), linguistic (e.g. McNeill 2009a; McNeill 2009b; McNeill 2010; Stokes 2010; Zaretsky 2010), and multi-modal communication approaches (e.g. Harris 1996; King 2013; Tierney 2016). Yet these non-RCT studies are inherently biased in nature and there is a need in the field for a systematic evaluation of available evidence. This review identifies best available treatments for CAS. This is an update of a Cochrane Review first published in 2008 (Morgan 2008). The previous review revealed no available RCTs for review. The first RCT in this field was published in 2015, hence it was timely to provide an updated review.

OBJECTIVES

To assess the efficacy of interventions targeting speech and language in children and adolescents with CAS as delivered by speech and language pathologists/therapists.

METHODS

Criteria for considering studies for this review

Types of studies

RCTs and quasi-RCTs (e.g. studies in which participants are allocated to intervention groups on alternate days).

Types of participants

Children aged 3 to 16 years with a diagnosis of CAS made by a speech and language pathologist/therapist.

Types of interventions

See Description of the intervention section above.
Eligible control groups were no treatment control (e.g. wait-list control), treatment as usual, or other treatment controls.

Types of outcome measures

Primary outcomes

1. Accuracy of production on treated or non-treated* items (may be associated with motor-based, linguistic or multi-modal communication approaches noted under How the intervention might work)

A desirable outcome would have been an improvement in accuracy of speech or multi-modal communication, while an undesirable outcome would have been deterioration from baseline.
*Non-treated items are stimuli (e.g. syllables, words, phrases) that have not been practised by children during intervention sessions. They are a form of control whereby we are able to measure children's performance on 'treated' items (e.g. syllables, words, phrases the child has practised during speech sessions) and compare it with performance on 'non-treated' items. In this way, we can quantify whether the child has 'generalised' their newly acquired speech skills, or improvement in speech, to non-treated stimuli,

or whether they have only improved on speech items practised during therapy.

Secondary outcomes

1. Speech production consistency across repeated words and syllables (may be associated with motor-based, linguistic or multi-modal communication approaches noted under How the intervention might work)
2. Accuracy of connected speech, including co-articulation accuracy (e.g. syllable segregation, voice onset time; most commonly associated with motor-based or linguistic approaches noted under How the intervention might work)
3. Functional communication (e.g. child- or parent-based questionnaire; may be associated with motor-based, linguistic or multi-modal communication approaches noted under How the intervention might work)

A desirable outcome would have been an improvement on outcomes one to three, whilst an undesirable outcome would have been deterioration from baseline on outcomes one to three. Outcome measurements were recorded before, immediately after and at longer-term follow-up.

Search methods for identification of studies

Electronic searches

Margaret Anderson, Cochrane Information Specialist for the Developmental, Psychosocial and Learning Problems Group, conducted the searches for this update in August 2011, June 2014 and April 2017. We searched the following list of sources which includes bibliographic databases, and international and national trials registers. We did not apply any date restrictions, but we only examined articles written in the English language. We report the search strategies for this update in Appendix 1. Earlier search strategies are in Appendix 2.

1. Cochrane Central Register of Controlled Trials (CENTRAL; 2017, Issue 3) in the Cochrane Library, and which includes the Cochrane Developmental, Psychosocial and Learning Problems Specialized Register (searched 6 April 2017)
2. Ovid MEDLINE (1946 to March week 5 2017)
3. Ovid MEDLINE E-Pub Ahead of Print (searched 6 April 2017)
4. Ovid MEDLINE In Process & Other Non-indexed Citations (searched 6 April 2017)
5. Embase Ovid (1980 to 2017 week 15)
6. CINAHL EBSCOhost (Cumulative Index to Nursing and Allied Health Literature; 1937 to 10 April 2017)
7. PsycINFO Ovid (1806 to April week 1 2017)
8. PsycINFO EBSCOhost (1887 to 4 August 2011)
9. ERIC EBSCOhost (Education Resources Information Center; 1966 to 10 April 2017)
10. ERIC Proquest (Education Resources Information Center; 1966 to 6 June 2014)
11. *Cochrane Database of Systematic Reviews* (CDSR; 2017, Issue 4) part of the Cochrane Library
12. Database of Abstracts of Reviews of Effect (DARE; 2015, Issue 2) part of the Cochrane Library (not searched in previous version of review (Morgan 2008). Final issue published in 2015)
13. SpeechBITE (speechbite.com; searched 10 April 2017)
14. Australian New Zealand Clinical Trials Registry (ANZCTR; www.anzctr.org.au/BasicSearch.aspx; searched 12 April 2017)
15. Chinese Clinical Trial Registry (ChiCTR; www.chictr.org.cn; searched 10 April 2017)
16. ClinicalTrials.gov (clinicaltrials.gov; searched 10 April 2017)
17. EU Clinical Trials Register (clinicaltrialsregister.eu; searched 10 April 2017)
18. ISRCTN Registry (www.isrctn.com; searched 10 April 2017)
19. Nederlands Trial Register (trialregister.nl/trialreg/admin; searched 10 April 2017)
20. World Health Organization International Clinical Trials Registry Platform (WHO ICTRP; www.who.int/ictrp/en; searched 10 April 2017)

Searching other resources

We searched the reference lists of included reports, and requested information on unpublished trials from authors of published studies and other experts, as well as information groups in the areas of speech and language therapy/pathology and linguistics.

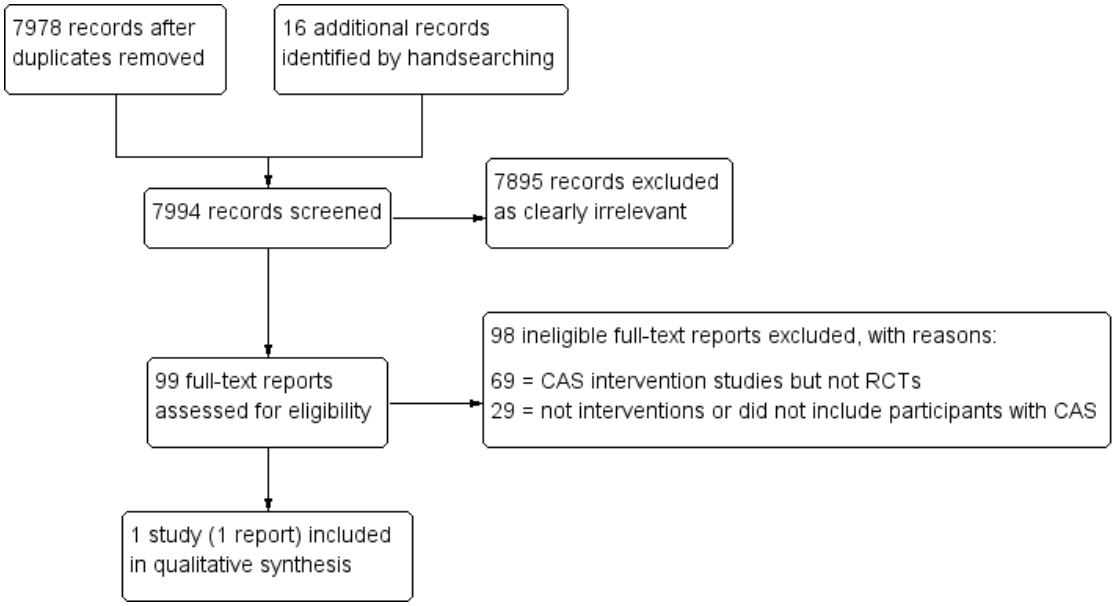
Data collection and analysis

We were unable to use many of our preplanned methods (Morgan 2006), as only one study met the inclusion criteria (Criteria for considering studies for this review). This study was published in a peer-reviewed journal and there are no other completed RCTs or quasi-RCTs at this time, published or unpublished. See Appendix 3 and Morgan 2006.

Selection of studies

Two review authors (FL and AM) independently screened all titles and abstracts yielded by the search for eligibility. In cases of uncertainty over whether an abstract met the inclusion criteria, we obtained the full-text report. Next, the same two reviewers independently evaluated each full-text report against the inclusion criteria (Criteria for considering studies for this review). In the event of disagreement over inclusion of a particular paper, FL and AM reached consensus by re-assessing the study against the inclusion criteria together. We present the results of our selection process in a PRISMA diagram; see Figure 1 (Moher 2009).

Figure 1. Study flow diagram



Data extraction and management

In addition to outcome data, we documented the following information using a data management form: participant details; setting (e.g. community clinic, school); type of intervention; length and frequency of intervention; professions involved; duration of impairment; level of severity; co-morbidity; and assessment tools employed. We requested any information that was missing or unclear from the corresponding author (Dealing with missing data). AM independently extracted and entered the outcome data into Review Manager 5 (Review Manager 2014), and FL then independently evaluated the data and entries. AM and FL discussed any disagreements until they reached a consensus. EM entered further details of excluded studies into Table 1.

Assessment of risk of bias in included studies

Two review authors (FL and AM) independently assessed the risk of bias within the one included study, using Cochrane's 'Risk of bias' tool (Higgins 2011a). Both review authors rated the risk of bias as low, high or unclear (uncertain), across each of the domains listed below. There were no cases of disagreement.

1. **Sequence generation**. Did the study describe the method used to generate the allocation sequence in sufficient detail to determine whether it produced comparable groups? In the review authors' judgment, was the sequence adequately generated?
2. **Allocation concealment**. Did the study describe the method used to conceal the allocation sequence in sufficient detail to assess whether intervention schedules could have been foreseen in advance of, or during, recruitment? In the review authors' judgment, was allocation adequately concealed?
3. **Blinding of participants and personnel**. Did the study describe any measures used to blind participants and personnel from knowledge of which intervention a given participant might have received? In the review authors' judgment, was knowledge of the allocated interventions adequately concealed from participants and relevant personnel during the study?
4. **Blinding of outcome assessment**. Did the study describe any measures used to blind outcome assessors from knowledge of which intervention a given participant might have received? In the review authors' judgment, was knowledge of the allocated interventions adequately concealed from all outcome assessors during the study?
5. **Incomplete outcome data**. Did the study report data on attrition and exclusions as well as the numbers involved

(compared with total randomised), reasons for attrition/exclusion, and any re-inclusions in analyses performed. In the review authors' judgment, did the study authors deal adequately with incomplete data?

6. **Selective outcome reporting**. Did the study make attempts to assess the possibility of selective outcome reporting? In the review authors' judgment, are reports of the study free of suggestion of selective outcome reporting determined by comparing the outcomes listed in the original study protocol with the final RCT publication?

7. **Other sources of bias**. Was the study apparently free of other problems that could put it at a high risk of bias? In the review authors' judgement, was the study free of other problems not covered by the domains above?

Measures of treatment effect

We were unable to conduct a meta-analysis due to there being only one included study. We have archived our methods for use in future updates of this review (see Appendix 3; Morgan 2006).

Unit of analysis issues

For each outcome measure, we averaged the accuracy of production (e.g. number of correct items from total items elicited) across the group. Units were mean accuracy scores for each intervention group. See Appendix 3 for additional methods archived for use in future updates of this review.

Dealing with missing data

There were missing data for 1/26 participants in the Murray 2015 RCT, due to a participant withdrawing in the middle of treatment (see Appendix 3 and Morgan 2006).

Assessment of heterogeneity

We were unable to assess heterogeneity as only one study met the inclusion criteria (see Appendix 3 and Morgan 2006).

Assessment of reporting biases

We were unable to assess reporting biases due to there being only one included study (see Appendix 3 and Morgan 2006).

Data synthesis

We could not undertake a meta-analysis as we included only one study in the review (see Appendix 3 and Morgan 2006).

Summary of findings

Using GRADEpro GDT (GRADEpro GDT 2015), we created a 'Summary of findings' table for the comparison: Nuffield Dyspraxia Programme - Third Edition (NDP-3) versus Rapid Syllable Transition Treatment (ReST) for Childhood Apraxia of Speech. In this table we report our primary (accuracy of production on treated and non-treated items) and secondary (speech production consistency and accuracy of connected speech) outcomes for one month post-treatment. We chose this time point as it is the most clinically salient time point. The time point immediately after therapy is not sufficient to determine whether the treatment effect was sustained. We did not examine the time point of four months post-therapy because the number of participants in each group (NDP-3: 9/13 participants; ReST: 9/13 participants) had returned to community SLP/SLT treatment between the one-month and four-month post-therapy period and, as such, it would be difficult to delineate between a sustained treatment effect of the RCT versus the usual therapy re-introduced. We also report in this table the quality ratings for each outcome as assessed by two review authors (AM and FL) using the GRADE approach (Schünemann 2017). They assigned ratings of high, moderate, low or very low quality, according to the presence of risk of bias (Risk of bias in included studies), indirectness of evidence, unexplained heterogeneity or inconsistency in results, imprecision of results and high probability of publication bias; they discussed any disagreements over the quality ratings until a consensus was reached.
Please see 'Summary of findings for the main comparison' for an overview of treatment effects for each outcome measure and GRADE assessment of the quality of the evidence.

Subgroup analysis and investigation of heterogeneity

We were unable to perform any subgroup analyses as we included only one study in the review. See Appendix 3 and Morgan 2006.

Sensitivity analysis

We were unable to perform a sensitivity analysis as we included only one study in the review. See Appendix 3 and Morgan 2006.

RESULTS

Description of studies

Results of the search

We identified a total of 7978 records once duplicates were discarded. EM identified a further 16 records through handsearching. Of these 7994 titles and abstracts, we excluded 7895 as clearly

irrelevant, and assessed the full texts of the remaining 99 reports against our inclusion criteria (Criteria for considering studies for this review). From these 99 reports, only one study met our inclusion criteria for this review (Included studies); we excluded the remaining 98 reports as irrelevant (see Excluded studies). We did not identify any non-English abstracts for inclusion. Please see Figure 1.

Included studies

The one included study, Murray 2015, was an RCT that compared treatment effects for two interventions, each delivered intensively (one hour for four days a week for three weeks): the Nuffield Dyspraxia Programme - Third Edition (NDP-3; Williams 2004) and the Rapid Syllable Transition treatment (ReST; Ballard 2010). Twenty-six children (13 allocated to each therapy group), aged 4 to 12 years (18 males) with CAS diagnosed by a SLP/SLT participated in the study, which took place at the University of Sydney Communication Disorders Treatment and Research Clinic. The primary outcomes were per cent accuracy on treated and untreated pseudo-words and real words and phrases.

The research was funded (as published in the article) by: Douglas and Lola Douglas Scholarship on Child and Adolescent Health; Nadia Verrall Memorial 2010; and Postgraduate Student Scholarship 2011 through Speech Pathology Australia, James Kentley Memorial Scholarship, Postgraduate Research Support Schemes and Faculty of Health Sciences; University of Sydney International Development Program Fund, and the Australian Research Council Future Fellowship.

Please see the Characteristics of included studies table for further detail of the nature of these interventions.

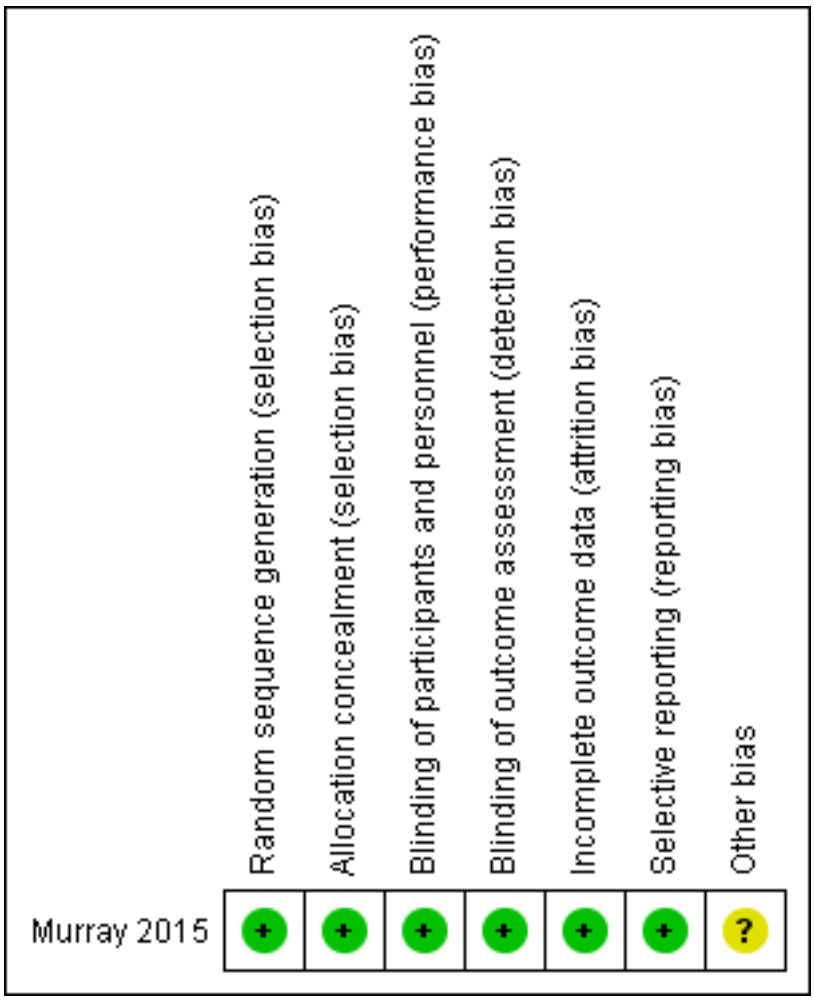
Excluded studies

We excluded 98 full-text reports. Of these, 29 studies were either not interventions (e.g. diagnostic studies), or did not include participants with CAS (e.g. focused on other speech disorders or adult-acquired apraxia of speech). The remaining 69 excluded papers were CAS intervention studies but were not RCTs, and are tabulated in Characteristics of excluded studies tables. Further detail on the excluded CAS studies is provided in Table 1.

Risk of bias in included studies

We examined the one included study, Murray 2015, for risk of bias. We judged the study to be at low risk of bias for all domains except 'other sources of bias', which we judged to be at unclear risk of bias. Please see the 'Risk of bias' table (beneath the Characteristics of included studies table) for further detail on the basis of our decisions, and Figure 2 for a summary of ratings.

Figure 2. Risk of bias summary: review authors' judgements about each risk of bias item for each included study.



Effects of interventions

See: **Summary of findings for the main comparison**

We downgraded the quality of the evidence by one level to moderate due to imprecision, given that only one RCT was identified.

Primary outcome: accuracy of production

The Murray 2015 study compared the number of real words produced correctly (out of the total elicited words) at pre-treatment with one month post-treatment for treated and non-treated items.

Treated items

The study authors reported that, compared to pre-treatment baseline, the NDP-3 MD of 36.0 was greater than the ReST MD of 33.9 at one month post-treatment, with an absolute mean difference of 2.1 between groups.

Non-treated items

The study authors reported that, compared to pre-treatment baseline, the ReST MD of 18.3 was minimally greater than the NDP-3 MD of 18.2 at one month post-treatment with an absolute mean difference of 0.1 between groups.

Secondary outcomes

Speech production consistency

The Murray 2015 study compared treatment gains in speech production consistency (measured by 25 real words repeated three times using the inconsistency subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP) test (Dodd 2006)), at pre-treatment with one month post-treatment for treated items. The study authors reported that, compared to pre-treatment baseline, the NDP-3 MD of 11.1 was minimally greater than the ReST MD of 10.9 at one month post-treatment, with an absolute mean difference of 0.2 between groups.

Accuracy of connected speech

The Murray 2015 study compared treatment gains in the accuracy of connected speech (as assessed by imitated word accuracy in connected speech of at least three word combinations), at pre-treatment with one month post-treatment for treated items. The study authors reported that, compared to pre-treatment baseline, the NDP-3 MD of 14.3 was greater than the ReST MD of 11.5 at one month post-treatment, with an absolute mean difference of 2.8 between groups.

The study did not measure functional communication.

DISCUSSION

Summary of main results

We sought to investigate the effectiveness of targeted speech and language interventions for children and young people, aged 3 to 16 years of age, with a diagnosis of CAS made by a speech and language pathologist/therapist. We found only one study, Murray 2015, which met our inclusion criteria (Criteria for considering studies for this review). This RCT recruited 26 children aged 4 to 12 years, and compared two interventions: the Nuffield Dyspraxia Programme-3 (NDP-3); and the Rapid Syllable Transitions Treatment (ReST). Treatments were delivered intensively in one-hour sessions, four days a week for three weeks, in a university clinic in Australia. Speech pathology students delivered the treatments in the English language.

We considered all core domains to be at low risk of bias. Both the NDP-3 and ReST therapies demonstrated improvement at one month post-treatment. A number of cases in each cohort had recommenced usual treatment by their speech and language pathologist between one month and four months post-treatment (NDP-3: 9/13 participants; ReST: 9/13 participants). Hence we could not analyse maintenance of treatment effects to four months post-treatment without significant potential bias, and so we did not include this time point for further analysis in this review.

Overall there is limited evidence that, when delivered intensively, both the NDP-3 and ReST may effect improvement in word accuracy in 4- to 12-year-old children with CAS, measured by the accuracy of production on treated and non-treated words, speech production consistency and the accuracy of connected speech. The study did not assess functional communication. We are unable to say whether either treatment is better than the other, or better than no treatment or treatment as usual. No evidence currently exists to support the effectiveness of other treatments for children aged 4 to 12 years with idiopathic CAS, without other comorbid neurodevelopmental disorders. No formal analyses were conducted to compare NDP-3 and ReST by the original study authors, hence one treatment cannot be reliably advocated over the other. Further RCTs replicating this study would strengthen the evidence, which we currently rate as low using the GRADE evidence rating system (i.e. that 'further research is very likely to have an important impact on our confidence in the estimate and is likely to change the estimate').

Further well-controlled studies investigating the effectiveness of other treatments for CAS are also needed across other motor-based therapies, and also across linguistic and multi-modal approaches. As noted earlier in the Why it is important to do this review section, non-RCT case series or case-control studies examining motor, linguistic and multi-modal interventions for CAS have described positive effects of intervention, but RCTs are required to strengthen the evidence base for these approaches. Further, there is also a need for trials that examine interventions for CAS compared to no treatment (e.g. wait-list control group). A no-treatment comparison is arguably difficult to achieve in this field however, given the typically severe presentation of speech disorder and reticence of parents or clinicians (or both) to withhold treatment from children. Finally, RCTs are also needed on populations with CAS and co-occurring neurodevelopmental or medical disorders.

Cochrane Reviews are often criticised in the SLP/SLT field because they do not allow consideration of lower levels of evidence, such as case studies or case series, which are more commonly performed in the field. Recognising these concerns we have provided a summary of the observational studies of CAS interventions excluded from this review (see Table 1), to encourage future, rigorous and controlled investigation of the efficacy of these methods. The lack of RCT intervention data in the CAS field to date is reinforced by challenges of: (1) the low incidence of the disorder; (2) the lack of a universally applied diagnostic classification system; (3) a lack of understanding of the aetiology of CAS; and (4) the challenge of designing trials for children with co-occurring clinical features (e.g. non-verbal cognitive impairment) or disorders (e.g. intellectual disability, autism spectrum disorder).

Overall completeness and applicability of evidence

We identified only one small RCT for inclusion in this review, indicating that there is an urgent need for further RCTs in this field. The interventions examined are currently in use and therefore results are applicable to clinical practice.

Quality of the evidence

We considered the overall quality of the evidence to be moderate using the GRADE approach; see Summary of findings for the main comparison. We downgraded the quality of the evidence by one level to moderate, due to imprecision, given that only one RCT was identified.

Potential biases in the review process

We carefully managed potential conflicts of interest, as described below under Contributions of authors and Declarations of interest. There is a possible risk of language bias given that we only included studies written in English.

Agreements and disagreements with other studies or reviews

There are no other systematic reviews examining only RCT and quasi-RCT evidence for efficacy of treatment for CAS.

AUTHORS' CONCLUSIONS

Implications for practice

The present review concluded that there is only one RCT examining interventions for CAS in the literature to date, which requires replication. This study provides some evidence that the NDP-3 may improve the accuracy of production on treated items (words) and connected speech, but limited evidence that the NDP-3 improves speech production consistency or that the ReST improves accuracy of production on non-treated words. The study did not measure functional communication.

There are a range of further therapies reported in the literature (Table 1), but the effectiveness of these interventions has not been rigorously examined; that is, other existing studies involve case study or case-series investigations and not RCTs, limiting the ability to interpret and generalise findings to a broader population of children with CAS. At present the evidence supports the use of NDP-3 or ReST intervention programmes for children with idiopathic CAS, aged 4 to 12 years, without other co-occurring neurodevelopmental deficits. Further well-controlled studies investigating the effectiveness of other treatments for CAS are urgently needed. There is a substantial range of treatments available for CAS; however, these require comparison with each other and to a no treatment (e.g. wait-list control) group before their efficacy is rigorously demonstrated. Further trials are also needed that examine the efficacy of therapies for children with CAS with a range of co-occurring neurodevelopmental impairments or diagnoses.

Implications for research

There is a critical need for further rigorously controlled studies of treatment efficacy for CAS. Replication of the work by Murray 2015 is required. Further work should also rigorously examine other CAS treatments reported in the literature. RCTs and quasi-RCTs are difficult to conduct given the heterogeneity of presentation of individuals with CAS, and due to the low incidence of the disorder. However, the work of Murray 2015 shows RCTs are possible.

Future studies may also investigate further therapy implementation variables to increase our understanding of treatment response in this population, in particular considering dose, delivery, uptake and context, with examples given below.

1. Duration, dose, delivery, uptake and intensity of treatment (e.g. intervention once a week over 12 weeks or three sessions over five weeks)

2. Response of particular subgroups of participants to treatment (e.g. subgroups based on age, genetic diagnosis, specific speech symptomatology), or dependent upon similarity of co-occurring features (e.g. gross and fine motor or cognitive presentation)

3. Impact of timing of treatment (e.g. intervention at three years versus six years)

4. Effect of the administrator of treatment (e.g. clinician, parent, teacher's aide or even participant-administered therapy for older children)

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* *Indicates the major publication for the study*

CHARACTERISTICS OF STUDIES

Characteristics of included studies *[ordered by study ID]*

Murray 2015

Methods	Parallel-group randomised controlled trial
Participants	**Sample size**: 26 children **Dropouts/withdrawals**: 1 child in the NDP-3 group dropped out mid-treatment yet was included in the analysis using intention-to-treat analysis **Sex**: 18 males, 8 females **Mean age**: 5 years and 6 months (SD = 25 months) **Inclusion criteria** 1. Clinical diagnosis of confirmed CAS, specified as having all 3 features of the ASHA 2007 consensus-based position paper, and at least 4 out of 10 features from the 'Strand' checklist (Shriberg 2010) 2. Aged between 4 and 12 years at time of treatment 3. Standard score of $\geq$ 85 for receptive language of CELF-IV or CELF-P2 4. Normal or adjusted-to-normal hearing and vision 5. Child and at least 1 parent being native Australian-English speakers 6. No other diagnosed developmental or genetic disorders (e.g. dysarthria, autism or intellectual disability) No information was collected on race, ethnicity or socioeconomic status
Interventions	**Process** Participants were randomly assigned to 1 of the 2 treatments: ReST or NDP-3. Concealed allocation was revealed after baseline assessment was completed. No significant differences between groups for any baseline variables (age, sex, primary or secondary outcome measures or CAS severity). Dose was controlled. Treatment was delivered for both ReST and NDP-3 over 12 x 1-hour sessions, scheduled 4 days/week for 3 weeks in school vacation time in January 2011 and January 2012, with a maximum of 10 participants per block. Treatments were provided as per intervention manuals and published protocol (Murray 2012). ReST sessions had an average of 100.4 production trials (SD = 0.9) and NDP-3 had an average of 101.3 (SD = 1.2), with no significant difference in number of production trials between groups. Therapy was provided by student SLPs under the supervision of Murray and McCabe. Several days of training were provided for both treatments and in transcription and data collection until reaching inter-rater reliability > 85%. Further detail on each treatment is provided below 1. **ReST**: this treatment is based on principles of motor learning. There were 3 goal levels within the treatment: (1) 2-syllable C1V1C2V2 (e.g. bagu or fabi), (2) 3-syllable C1V1C2V2C3V3 (e.g. baguti or fabitu), (3) 3-syllable pseudo words as final nouns within carrier phrases (e.g. "Can I have a baguti?"). Children were required to practise production of 20 pseudo words, with a goal of 80% accuracy of production in perceptually rated articulation, coarticulation and prosody over 2 consecutive sessions before stepping up to the next goal level. The child's initial goal level was selected dependent upon initial diagnostic testing prior to the pre-treatment experimental probe. Consonants in the stimuli were individually selected for each child to ensure all target sounds were at least 10% stimulable and were maximally different fricative and plosive sounds (e.g. /b/, /f/, /t/, /g/), again based on pre-treatment data. Stimuli were

	designed so that half had a strong - weak pattern and the remainder a weak - strong pattern, with the third syllable being either strong (using "ee" (/i/)) or weak (using "er", the Australian schwa). All pseudo words had a high phonotactic probability and were orthographically biased. Sessions consisted of pre-practice and practice components. In pre-practice, which lasted 10 to 15 minutes, the clinician aimed to elicit at least 5 correct productions of any of the 20 stimuli using imitation, phonetic placement cues, tapping of stress pattern, segmenting and blending and prosodic cues in addition to 'knowledge of performance' feedback after each production. In practice, which lasted around 50 minutes, the participant worked toward the goal of 80% accuracy with no cues given across 100 trials. Trials were delivered in 5 blocks of 1 trial of each of the 20 treated stimuli, presented in random order. 'Knowledge of results' feedback was provided 50% of the time on a decreasing scale (i.e. on 9 of the first 10 trials, down to only 1 of the final 10 trials). See Murray 2012 and Murray 2015 for further detail 2. **NDP-3**: the NDP-3 intervention was conducted as described in the manual (Williams 2004) and subsequent publication (Williams 2010). Treatment goals targeted unknown segments as single sounds or syllable shapes using known sounds. Each goal was targeted during a game-based activity, treated in a separate block of 18 minutes and was associated with 5 individualised stimuli. Children were required to achieve 90% accuracy for each target stimulus before moving on to different stimuli within the same goal. Verbal instructions, modelling and articulation, and visual - tactile cues were provided as needed. 'Knowledge or results' and 'knowledge of performance' feedback was provided 100% (i.e. after every production attempt). If the production was correct, the child was then asked to repeat the response a further 3 times, again with immediate knowledge of results and knowledge of performance feedback by the clinician
Outcomes	**Timing of outcome assessment** Outcome assessments were conducted prior to treatment and within 1 week, 1 month and 4 months post-treatment. No therapy was reported between study onset and 1 month post-treatment yet over half the cohort resumed community SLP services between 1 and 4 months post-treatment (ReST = 9, NDP-3 = 9) **Primary outcomes** The primary outcomes included: 1. treatment gains; 2. maintenance of treatment gains; and 3. expected response generalisation to untreated real words and pseudo words using experimental probe items at the child's individualised generalisation level Outcomes were measured based on a 292-item experimental probe of treated and untreated stimuli. 162 items from NDP-3 assessment and 80 pseudo words from ReST treatment, and an additional 50 untreated 1-, 2- and 3-syllable real word stimuli were used to test for generalisation of treatment effects in both groups. The probe assessed impairment level speech outcomes for simultaneous accuracy for articulation and prosody. For further detail on scoring, see Murray 2015. **Secondary outcomes** A number of secondary measures of generalization were made to further explore potential differences in the treatments' effects 1. Imitated word accuracy in untreated connected speech of at least 3 words (as per

	NDP-3 manual; Williams 2004, p 143) 2. DEAP (Dodd 2006) inconsistency subtest 3. Single Word Test of Polysyllables (Gozzard 2004) (only administered at pre-treatment and 1-month post-treatment) 4. GFTA-2 (Goldman 2000) was administered at pre-treatment and 1-month post-treatment to document changes in segmental accuracy using per cent phonemes correct (PPC), per cent vowels correct (PVC), per cent consonants correct (PCC) as well as per cent lexical stress (prosodic) matches for untreated single words in these clinically available assessments. For further detail on scoring, fidelity, reliability and recording, see Murray 2015 **Comparisons** 3 comparisons for each primary and secondary outcome measure were conducted 1. Pre-treatment compared with 1 week post-treatment to assess acquisition of treatment and generalization effects 2. 1 week versus 1 month post-treatment to assess short-term maintenance of these effects 3. 1 week versus 4 month post-treatment to test longer-term maintenance with exception of the Single Word Test of Polysyllables (Gozzard 2004) and GFTA-2 (Goldman 2000), which were only administered pre-treatment and 1 month post-treatment
Notes	**Funding** Douglas and Lola Douglas Scholarship on Child and Adolescent Health; Speech Pathology Australia funded Nadia Verrall Memorial 2010 and Postgraduate Student Scholarship, James Kentley Memorial Scholarship, Postgraduate Research Support Schemes and Faculty of Health Sciences; University of Sydney International Development Program Fund; and Australian Research Council Future Fellowship **Conflicts of interest**: none known **Study start date**: January 2010 **Study end date**: July 2012

Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Clarification was sought from the corresponding author by phone who confirmed that each envelope had a note within it specifying the treatment condition to which the child was allocated (Murray 2015). The authors could not see through the envelopes. Envelopes were placed in a container and an independent person (corresponding author's husband) not involved in the study selected an envelope that was then given a participant number (P1, P2, etc.) until all participants were allocated to an arm of the study. Allocation was not revealed until after the pre-treatment evalua-

Murray 2015 (*Continued*)

		tion
Allocation concealment (selection bias)	Low risk	Clarification was sought from corresponding author (Murray 2015), who confirmed via email that envelopes were sequentially numbered based on the random order in which they were selected from a container (i.e. randomised and not based on any identifying variable)
Blinding of participants and personnel (performance bias) All outcomes	Low risk	SLP could not be blinded to type of intervention
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Blinded, independent assessors
Incomplete outcome data (attrition bias) All outcomes	Low risk	No attrition
Selective reporting (reporting bias)	Low risk	All outcome measures reported in the original protocol, Murray 2012, were reported. A lexical stress measure was added in final outcome ratings but not mentioned in protocol but this was an addition and not a failure to report
Other bias	Unclear risk	1. Maintenance findings. Some children resumed their usual therapy in the 4-month period to maintenance assessment. Whilst the number of children resuming usual treatment was similar between both groups, this variable may have led to increased maintenance results across both treatments 2. No control group without intervention (i.e. no wait-list control group) 3. Pre- and post-treatment assessors Qualified SLPs who had not seen the children previously conducted the 1 week, 1 month and 4 month post-assessments. In some cases, final-year undergraduate SLP students (4th-year students) conducted post-assessments. The same SLP or student SLP must not have seen/rated the children before. One researcher performed all of the pre-assessments, including probes, before allocation was revealed

CAS: childhood apraxia of speech;**CELF-IV**: Clinical Evaluation of Language Fundamentals - Fourth Edition; **CELF-P2**: Clinical Evaluation of Language Fundamentals - Preschool 2; **DEAP**: Diagnostic Evaluation of Articulation and Phonology; **GFTA-2**: Goldman-Fristoe Test of Articulation 2;**NDP-3**: Nuffield Dyspraxia Programme - Third Edition; **ReST**: Rapid Syllable Transitions Treatment; **SD**: standard deviation; **SLP**: speech language pathologist

Characteristics of excluded studies *[ordered by study ID]*

Study	Reason for exclusion
Baas 2008	Not RCT or quasi-RCT (case study)
Ballard 2010	Not RCT or quasi-RCT
Beathard 2008	Not RCT or quasi-RCT (case study)
Binger 2007	Not RCT or quasi-RCT (case study series)
Binger 2008	Not RCT or quasi-RCT (case study)
Binger 2011	Not RCT or quasi-RCT (case study)
Bornman 2001	Not RCT or quasi-RCT (case study)
Bose 2001	Not RCT or quasi-RCT (case study series)
Carter 2004	Not RCT or quasi-RCT (case study series)
Chappell 1973	No experimental treatment data included in study
Culp 1989	Not RCT or quasi-RCT (single case [ABA] design)
Cumley 1999	Not RCT or quasi-RCT (case series)
Dale 2013	Not RCT or quasi-RCT (case series)
Daly 1972	Not RCT or quasi-RCT (case study)
Dworkin 1988	Study examined adult participant with AAOS
Edeal 2011	Not RCT or quasi-RCT
Forrest 2001	Study focuses on children with speech disorder, not specifically DAS. No experimental treatment data included in study
Groenen 1996	No experimental treatment data included in study

(*Continued*)

Hadar 1984	Study examined adult participant with AAOS
Hall 1989	Not RCT or quasi-RCT (case study)
Hall 1990	Not RCT or quasi-RCT (longitudinal case study)
Harris 1996	Not RCT or quasi-RCT (case study)
Hayden 2006	Study uses a hypothetical treatment case only. No experimental treatment data
Head 1975	Study focuses on intervention for a group of participants with a range of speech disorders without dissociating between participants with subtypes of speech disorders. Does not report treatment efficacy specific to participants with DAS
Helfrich-Miller 1994	Not RCT or quasi-RCT (case study series)
Iuzzini 2010	Not RCT or quasi-RCT (case study)
Jaroma 1984	Study does not specify whether child has diagnosis of DAS or only some features of dyspraxia
Kadis 2014	Not RCT or quasi-RCT (case study series)
Katz 2006	Study examined adult participants with AAOS
King 2013	Not RCT or quasi-RCT (case study series)
Kingston 1987	Study focused on articulation disorders, not specifically DAS
Klick 1985	No experimental treatment data included in study
Krauss 1982	Not RCT or quasi-RCT (case study)
Lagasse 2012	Not RCT or quasi-RCT (case study)
Lozano 1978	Study examined adult participant with AAOS
Lundeborg 2007	Not RCT or quasi-RCT (case study)
Lüke 2016	Not RCT or quasi-RCT (case study)
Maas 2012a	Not RCT or quasi-RCT (case study)
Maas 2012b	Not RCT or quasi-RCT (case study)
Martikainen 2011	Not RCT or quasi-RCT

(*Continued*)

Martin 2016	Not RCT or quasi-RCT (case study series)
McCabe 2014	Not RCT or quasi-RCT (case study)
McNeill 2009a	Not RCT or quasi-RCT (case series)
McNeill 2009b	Not RCT or quasi-RCT (case study)
McNeill 2010	Not RCT or quasi-RCT (case study series)
Morgan Barry 1995	Not RCT or quasi-RCT (case study series)
Moriarty 2006	Not RCT or quasi-RCT (case study)
Namasivayam 2013	Not RCT or quasi-RCT (case study series)
Namasivayam 2015	Not RCT or quasi-RCT (pre-post group design)
Preston 2013	Not RCT or quasi-RCT
Preston 2016	Not RCT or quasi-RCT (case study)
Preston 2017	Not RCT or quasi-RCT (case study)
Ray 2003	Study examined adult participant with AAOS
Richardson 2004	Study focus on motor dyspraxia or developmental coordination disorder not apraxia of speech
Rosenbek 1974	Not RCT or quasi-RCT (case study)
Rosenthal 1994	Study combined a number of treatment methods and grouped individuals. Could not determine individual participant outcomes related to specific treatment methods
Skelton 2014	Not RCT or quasi-RCT (case study)
Square 1994	No experimental treatment data included in study
Stokes 2010	Not RCT or quasi-RCT (case study)
Strand 2000	Not RCT or quasi-RCT (case study)
Strand 2006	Not RCT or quasi-RCT (case series)
Thomas 2014	Not RCT or quasi-RCT (case study)
Thomas 2016	Not RCT or quasi-RCT (case study)

(*Continued*)

Tierney 2016	Not RCT or quasi-RCT (case study)
Vashdi 2013	Not RCT or quasi-RCT
Vashdi 2014	Not RCT or quasi-RCT
Velleman 1994	Not RCT or quasi-RCT (case series)
Yoss 1974	Not RCT or quasi-RCT
Zaretsky 2010	Not RCT or quasi-RCT (case study)

AAOS: acquired apraxia of speech.
ABA: applied behaviour analysis
DAS: developmental apraxia of speech.
RCT: randomised controlled trial.

ADDITIONAL TABLES

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control)

Study	Participants	Methodology/ paper type	Intervention	Intervention approach	Intervention intensity and duration	Outcome measures	Treatment outcomes	Timing of outcome measures	Methodological considerations
Baas 2008	1 male aged 12.8 years with CAS and charge syndrome	Not quasi-/RCT (Single case (AB) design)	Dynamic Temporal and Tactile Cueing	Motor	Phase I and II: sessions 4 × per week; Phase III: weekly therapy. Study over 25 months. Home practice not reported	Articulation accuracy on 2-item scale for treated items; speech rate	Phase I (core vocabulary) : change on 4/6 targets. Maintained at last probe Phase II (core vocabulary): reached 100% accuracy for 3/5 words. Reduction	Baseline and during treatment. No longer-term follow-up data	Lack of experimental control, multiple baselines, control, longer-term follow-up or generalisation data. Clinical file data used. No replication across par-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							of stereotypies Phase III: decreased speech rate from 94 to 71 SPM		ticipants. Assessors, participants, therapists not blinded
Ballard 2010	3 siblings (2 males, 1 female) aged 7.8 and 10. 10 years with CAS	Not quasi-/ RCT (Single subject multiple baseline design across behaviours and participants)	Rapid Syllable Transition Treatment (ReST)	Motor	60-minute sessions (100-120 trials per session), 4 × per week for 12 sessions. Home practice not reported	Reading aloud 10 treated and 10 non-treated non-word strings; real word generalisation data; perceptual analysis of prosodic pattern and acoustic analysis using pairwise variability index	3/3 had significant gains in treated items and generalisation to same level of treated complexity. 2/3 generalised to lower and higher complexity non-word items. Minimal generalisation to real words	Baseline data taken at beginning of every 4th session and at 4 weeks post-treatment	No long-term follow-up data. Limited participants for generalisation of outcomes. No blinding of assessors, participants or therapists. No stimulus generalisation measures
Beathard 2008	1 female aged 3 years with CAS	Not quasi-/RCT (Case description)	Music therapy	Other (alternative interventions)	30-minute sessions over 9 months. 24 sessions in total	Descriptive data only	Commenced non-verbal. At end, had 11 phonemes in inventory	Pre-treatment and post-treatment. No follow-up data	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No replicable outcome measures. No statistical anal-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									ysis. No blinding of assessors, participants or therapists. No follow-up or generalisation data. Unclear which aspect of treatment provided outcomes or affect of maturation, schooling, etc. No replication across participants. No long-term follow-up data
Binger 2007	2 males aged 4.2 and 4.4 years with CAS and language disorder	Not quasi-/RCT (Single case multiple baseline across participants)	Aided AAC Modeling	Augmentative and alternative communication	15-minute sessions, 1 to 3 × per week for 10 to 15 sessions	Frequency of use of multi-symbol messages in play scenarios	Significantly more frequent use of multi-symbol messages using aided AAC as well as different types of messages. Maintained and generalised gains. Increased participation	Baseline × 3, every 2nd treatment session, and at 2, 4 and 8 weeks post-treatment	CAS diagnosis unclear and not replicable. Limited outcome measures. No blinding of assessors. No response generalisation data taken (only stimulus generalisation)

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

Binger 2008	1 female (Latino) aged 3.4 years with CAS and suspected velocardiofacial syndrome	Not quasi-/RCT (Single case multiple baseline across participants)	Aided AAC Modeling	Augmentative and alternative communication	10-minute sessions, 1 to 3 × per week for 10 to 15 sessions	Frequency of use of multi-symbol messages in play scenarios	Significantly more frequent use of multi-symbol messages using aided AAC. Parental response to training excellent. Maintained and generalised gains	Baseline × 3, every 2nd treatment session, and at 2, 4 and 8 weeks post-treatment	CAS diagnosis unclear and not replicable. No blinding of assessors. No response generalisation data taken (only stimulus generalisation)
Binger 2011	1 female aged 6 years with CAS and language disorder	Not quasi-/RCT (Single case multiple baseline across behaviours)	Aided AAC Modeling	Augmentative and alternative communication	15-minute sessions, 1 to 3 × per week for 10 to 15 sessions	Frequency of use of grammatical morphemes	Significantly more frequent use of grammatical morphemes using aided AAC. 2nd intervention period needed for 2/3 targets. Maintained gains	Baseline × 3, every treatment session, and 2, 4 and 8 weeks post-treatment	CAS diagnosis unclear and not replicable. No blinding of assessors. No response generalisation data taken (only stimulus generalisation)
Bornman 2001	1 male aged 6.6 years with CAS, hemiplegia and seizures	Not quasi-/RCT (Single case (ABA) design)	Voice output devices (Macaw)	Augmentative and alternative communication	60-minute sessions for 2 sessions (training). Home practice focus	Frequency of appropriate responses to questions in structured discourse	Mother provided greater frequency and type of questions. Frequency of appropriate responses in-	2 × baseline, 2 × practice period, 1 × post-treatment, and 4 weeks post-treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							creased		and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. Unclear dosage of home practice. No generalisation data. No long-term follow-up data
Carter 2004	1 male and 1 female aged 12 and 8 years respectively diagnosed with CAS. Additional 8 children (7 males) aged 4 to 7 years with persistent articulation errors	Not quasi-/ RCT (Case series - single group study)	Electropalatography (EPG) on /t, d, k, g, s, z/	Motor	30-minute sessions, 1 × per week for 10 weeks	Per cent consonants correct (PCC) and Probe Scoring System (PSS) on probe of 43 words	Significant difference noted for PSS for whole group. PCC scores improved in percentage	Pre-treatment (baseline first session) and post-treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No follow-up or generalisation data. No blinding of assessors
Culp 1989	1 female aged 8 years with CAS and intellectual disability	Not quasi-/RCT (Single case (ABA) design)	Partners in Augmentative Communication Training (PACT)	Augmentative and alternative communication	30 to 90-minute sessions daily after 3 days of intensive training. Home	Ratio of parent vs participant messages; ratio of successful/intelli-	Participant had greater frequency of messages compared to parent, and	Pre-treatment and 2 months post-treatment	Lack of experimental control, multiple baselines or control data. CAS diagno-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

					practice focus	gible messages from child	slightly higher frequency of successful measures (high baseline accuracy) . Increased participation		sis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No immediate post-treatment data or generalisation data. No replication across participants
Cumley 1999	2 females and 1 male aged 3.4, 8 and 12. 9 years respectively, with CAS (2 with intellectual disability and 1 with submucous cleft)	Not quasi-/RCT (3 case studies/ reports)	Combined communication boards and voice output devices	Augmentative and alternative communication	3.4-year-old: 2 to 3 × per week for 12 weeks 8-year-old: daily for 6 months 12-year-old: not reported	3.4-year-old: MLU. 8-year-old: assessment of phonological processes; communication repairs 12-year-old: description of functional communication	3.4-year old: minimal speech improvement, MLU increased to WNL 8-year old: no change in speech, parent report of greater communication repairs, and less frustration 12-year old: supplemented natural speech	Pre-assessment and treatment descriptions	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No immediate post-treatment data

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							to initiate, maintain and repair communication		or generalisation data. No replication across participants
Dale 2013	3 males and 1 female aged 3.6 to 6 years diagnosed with CAS	Not quasi-/ RCT (Single subject (ABB or ABC) design)	Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) - full programme (FP) for 8 weeks versus PROMPT without tactile-kinaesthetic-proprioceptive cueing for 4 weeks and FP for 4 weeks	Motor	50-minute session, 2 × per week for 8 weeks	Trained words on probe, untrained words. Pre-post testing on the DEAP, TOCS+, VMPAC focal motor and sequencing subtests and Vineland socialization scales	2/4 improved on DEAP. 4/4 improved on TOCS+, VMPAC subtests and Vineland. All 4 showed greater improvement on easier targets and majority maintained to 3 months post-treatment. Generalisation to untrained items noted	Probe words: baseline × 3, treatment × 4, post-treatment, and 3 months post-treatment	Lack of experimental control as control data changed and interpreted as generalisation but no other control used (e.g. multiple baselines). CAS diagnosis concerning prosody unclear. Blinded assessors for only some outcomes. No withdrawal period between treatment phases and participant differences made comparison between

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									conditions difficult. All measures not taken at consistent times
Edeal 2011	2 males aged 6.2 and 3.4 years with CAS (1 case with repaired cleft lip and palate and language disorder)	Not quasi-/RCT (Single case (AB) design)	Integral Stimulation (Dynamic Temporal and Tactile Cueing)	Motor	Varied across participants. 40-minute sessions (15 minutes each condition plus probes) . 1 case: 3 × per week for 11 weeks. 1 case: 2 × per week for 5 weeks	Probe data on targeted phonemes (articulation) in words for each participant. 1 phoneme targeted with high production frequency = 100 trials and another with moderate production frequency = 60 trials. Articulation and language sample taken at 2 weeks post-treatment	Large effect sizes for high production frequency and moderate for moderate production frequency. Improvement in PCC and phoneme inventory post-treatment. Some generalisation	Baseline × 3, each treatment session, and 1 probe post-treatment	Lack of experimental control, multiple baselines or control data. No long-term follow-up data. No blinding of assessors. Accuracy based on if target phoneme was correct (including cognate pair substitution) not if whole word was correct
Hall 1989	1 female aged 9 years with mild CAS (followed until 12 years)	Not quasi-/ RCT (Case study/ report)	Articulation therapy, motor-programming remedial model	Motor	5 school semesters	Templin-Darley Tests of Articulation	Remediation of all 31 items for /r/, /ɜ˞/ and /ə˞/	Test completed each semester	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up data or generalisation data. No replication across participants. No stimulus generalisation measures
Harris 1996	1 male aged 5 years with CAS and language disorder	Not quasi-/RCT (Multiple baseline across discourse contexts)	Computer-based AAC	Augmentative and alternative communication	4-minute sessions, 2 × per week for 22 sessions over 4 months	Frequency of noun/verb phrases in reciprocal book reading and structured discourse	Improvement in both contexts but more so in book reading than discourse. Some generalisation	Baseline, treatment, and withdrawal probes	CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No follow-up data. No blinding of assessors. No replication across participants
Helfrich-Miller 1994	3 children (2 males, 1 female) aged 2.9 to 8 years with	Not quasi-/ RCT (Case study series)	Melodic Intonation therapy (MIT)	Linguistic and motor	Varied. 37 to 71 sessions	Varied. Description of skills, consonant in-	Child 1: all consonants in inventory Child 2:	Pre- and post-treatment	No experimental control. Lack of information

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	CAS					ventories, sequencing error rates and intelligibility compared to typical development	spoke in complex sentences, poor intelligibility, and articulation errors present Child 3: sequencing error rate dropped from 75% to 22%. 13/18 consonant sounds improved		on diagnosis of CAS. Primarily descriptive measures - not reliable or tested using statistics. No control, maintenance or generalisation data
Iuzzini 2010	4 children (2 males, 2 females) aged 3.7 to 6.10 years with CAS	Not quasi-/ RCT (Single case design)	Stimulability (STP) and modified Core Vocabulary (mCVT) used concurrently	Linguistic and motor	55-minute sessions (10 minutes STP, 45 minutes mCVT), 2 × per week for 20 sessions. No home practice	Per cent phonemes correct, phonetic inventory and inconsistency	PCC increased on average 20% after combined therapy (range 9% to 32%). Inventory gained 5 phones on average (range 1 to 10). 3/4 had greater consistency on CSIP and ISP after therapy; 1 had greater inconsistency	Pre- and post- treatment	Poor experimental control as stable baseline not established, lack of control data. CAS diagnosis unclear and not replicable. No statistical analysis. No blinding of assessors. No immediate post-treatment data or generalisation data
Jaroma 1984	1 male aged 5.5 years with	Not quasi-/	Sensory integrative	Motor	Daily sessions for 2	(SP only) Illi-	Test not completed	Pre-treatment only	Lack of experimen-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	"some dyspraxic features" (CAS diagnosis not explicit)	RCT (Case study)	therapy and speech therapy		months	nois Test of Psycholinguistic Abilities	post-treatment. Observation of greater self-monitoring and correction of speech		tal control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures and no post-treatment data. No blinding of assessors. No immediate post-treatment data or generalisation data. No replication across participants. Lack of information on speech therapy provided
Kadis 2014	14 children (9 males, 5 females) aged 3 to 6 years with diagnosed CAS (compared to 14 age-matched controls)	Not quasi-/ RCT (Case series pre-post design)	Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT)	Motor	2 × per week for 8 weeks (16 sessions in total)	GFTA2, HCAPP, VMPAC, MRI	Significant gains as a group for all speech measures	1-week pre-treatment (baseline), 1-week post-treatment	CAS diagnosis unclear and not replicable. Age-matched control group older than CAS

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									group. Limited information on PROMPT targets selected for replication. No blinding of assessors. No stimulus generalisation measures
King 2013	3 males aged 4.1, 5.8 and 8.6 years diagnosed with CAS. 1 of the 3 diagnosed with Opitz FG syndrome and another with PDD-NOS	Not quasi-/ RCT (Single subject multiple baseline across participants design)	Integrated Multimodal Intervention (structured book reading, drill and play activities with AAC devices present and speech encouraged)	Augmentative and alternative communication	1-hour sessions, 2 × per week for 3 to 6 weeks	Category (e.g. vocalisation, AAC or both), type of word and accuracy targets Case 1: final consonants. Case 2: initial /s/ clusters then /f/. Case 3: initial /s/ clusters	Increases in vocalisations/spoken speech noted for 3/3. Speech accuracy improved on targets for 1/3 cases but all showed some generalisation to more accurate everyday speech	Baseline probes, probes every 2nd treatment session, 1-month post-treatment	Poor experimental control for case 1 and some change on control data noted. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No generalisation data. No long-term treatment data
Klick 1985	1 female aged 5.6 years with CAS	Not quasi-/RCT (Case description)	Adapted Cueing Technique	Motor	30 minutes of therapy per day for 6 months	Number of single words/ utterances	From 2 to 4 words to 12 words and several carrier	Description of progress during treatment	Lack of experimental control, multiple baselines

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							phrases. After 6 months began to produce novel sentences		or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up or generalisation data. No replication across participants
Krauss 1982	2 males aged 5 and 6 years diagnosed with CAS	Not quasi-/ RCT (Single case (ABAA) design)	Concurrent Melodic Intonation Therapy (MIT) and traditional therapy (20% and 80% of sessions respectively)	Linguistic and motor	2 × per week over 2-month period	Pre- and post-treatment gains on word-morpheme usage, auditory comprehension, naming, describing function, sentence completion, imitation of word phrases and articulation. Tested using language	Significant gains were found in phrase length (MLU), picture naming, and verbal imitation tasks. Little change in articulation	Pre-treatment, post-traditional therapy, and post-MIT therapy	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No blinding of assessors. No immediate post-treatment data or generalisation data. No long-term follow-up.

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

						sampling and Porch Index of Communicative Ability in Children			There were no reliability data reported for language sample analysis, a subjective measure
Lagasse 2012	2 males aged 5 and 6 years with suspected CAS	Not quasi-/RCT (Single case (AB) design)	Melodic Intonation Therapy (MIT) compared to 'traditional speech-language therapy'	Linguistic and motor	Ongoing 1 × per week speech therapy (traditional articulation sessions) and 40-minute MIT music sessions over 4 weeks (both treatments concurrent)	GFTA2; KLPA2 and speech production on stimulable sounds in 1- or 2-syllable words	Case 1 made greater gains in MIT sessions (but only 2% gain). Case 2 made greater gains on traditional articulation therapy (15% gain)	Pre- and post- treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up or generalisation data
Lüke 2016	1 German-speaking male aged 2.7 years with severe CAS	Single case design (A-B design with 3 follow-up assessments post-treatment with some treatment sessions between assessments)	Speech Generating Devices - fixed display (Gotalk 20+) and dynamic display (DynaVox V)	Augmentative and alternative communication	45-minute sessions × 50 treatment sessions. Treatment sessions 2 to 28 days apart	Means of communication (oral versus SGD), intelligibility of speech productions, consistency of speech productions, lex-	Significantly more communication initially with SGD than speech; significant increase in speech intelligibil-	Baseline × 3, every 2nd treatment session, and 2, 4 and 8 weeks post-treatment	Lack of baseline data for consistency. CAS diagnosis unclear and not replicable. No blinding of assessors. No

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

						ical development, and grammatical development	ity; consistency (however reduced data in baseline period); amount of words used; and increased MLU and inflections after 8 to 9 sessions		clear withdrawal phase after treatment with SGDs for control and no generalisation data
Lundeborg 2007	1 female aged 5.1 years with CAS	Not quasi-/ RCT (Single case cross-over design)	Intra-oral stimulation and electropalatography	Motor	25-minute sessions (5 minutes intra-oral stim, 20 minutes EPG); daily at home, total of 195 sessions in 12 months	Per cent consonants correct, per cent phonemes correct, per cent words correct, intelligibility, visual deviancy	Significant treatment outcomes on all measures	Pre-testing, A1 (baseline), B (intervention: oral stimulation therapy), A2 (withdrawal for 3 months), B (intervention: EPG), and A3 (follow-up)	Cross-over design, no control group or data taken to control for maturation. No replication across participants. No long-term follow-up or generalisation data taken
Maas 2012a	4 children (2 males, 2 females) aged 5.4 to 8.4 years with CAS (2 also with dysarthria and a third with language disorder); 3 also in Maas	Not quasi-/ RCT (Single case alternating treatments design with multiple baselines across behaviours over 2 phases)	Dynamic Temporal and Tactile Cueing (high versus moderate feedback frequency in cross-over design)	Motor	50-minute sessions 3 × per week for 3 participants but 1 had 60-minute sessions 2 × per week	Per cent accuracy on 2-point scale of segmental and suprasegmental aspects of target words and phrases with 2 words	2 responded better to low frequency feedback, 1 to high frequency feedback, and 1 to no condition. No generalisation effects	Weekly probes: 3 to 4 × baseline, 4 × treatment. Phase 1: 4 to 5 × withdrawal, 4 × treatment. Phase 2: 2 × withdrawal and 1 month	Small sample size with heterogeneity. Cross-over conditions made comparison difficult regarding targets chosen. No control group.

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	2012b, as below							post-treatment	Effect sizes used not interpretable or comparable to others. Different doses across all participants. Treatment fidelity < 80%. No stimulus generalisation measures
Maas 2012b	4 children (2 males and 2 females) aged 5.0 to 7.9 years with CAS. 2 cases had additional dysarthria diagnoses 1 other case had multiple co-occurring disorders	Not quasi-/ RCT (Single case alternating treatments design with multiple baselines across behaviours over 2 phases)	Dynamic Temporal and Tactile Cueing (random versus blocked practice compared in cross-over design)	Motor	2 × 4 week blocks of therapy	Per cent accuracy on 2-point scale of segmental and suprasegmental aspects of entire target words and phrases with 2 words	3/4 responded to both conditions. 2 responded relatively better to blocked practice, 1 to random practice, and 1 to no condition. 2/4 demonstrated generalization	Weekly probes: 3 to 4 × baseline, 4 × treatment. Phase 1: 4 to 5 × withdrawal, 4 × treatment. Phase 2: 2 × withdrawal and 1 month post-treatment	Small sample size with heterogeneity. Cross-over conditions made comparison difficult regarding targets chosen. No control group. Effect sizes used not interpretable or comparable to others. Treatment fidelity < 80%. No stimulus generalisation measures

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

Martikainen 2011	1 female aged 4.7 years with CAS	Not quasi-/RCT (Multiple baseline across behaviours - cross-over treatment design)	Combined Melodic Intonation Therapy (MIT) and Touch Cue Method (TCM)	Motor and linguistic	3 sessions for 6 weeks for 18 sessions for MIT. 6 weeks no therapy. 3 sessions for 6 weeks for 18 sessions for TCM	Articulation accuracy: PVC, PCC. Also, overall word accuracy scores: PMLU, PWP, PWC All calculated from responses to 46 picture cards	1/5 measures significant post-MIT (per cent vowels correct). Per cent consonants correct also reduced 3/5 significant post-TCM (PVC, PCC, PMLU). PVC, PCC and PMLU maintained. Greater changes for both therapies after withdrawal. PCC and PMLU only significant after MIT withdrawn	Beginning and end of 6-week baseline, beginning and end of both treatment phases, 12 weeks after TCM withdrawn	Lack of experimental control of other factors. Cross-over design makes comparison of both treatments difficult as many changes only noted after withdrawal of MIT (accumulation effects). Limited outcome data. Lack of generalisation data No blinded assessors. No replication across participants
Martin 2016	12 children (sex unknown) aged 3 to 10 years with CAS (11 with co-occurring conditions)	Case series (pre and post design)	DuBard Association Method®. It is a multimodal, phonetic therapy which works from accurate sounds	Motor	Daily in small groups in a school programme for an 11-month period	Articulation, mean length of utterance (MLU), and intelligibility on Arizona Articulation Proficiency Scale-	Significant changes in articulation, intelligibility and MLU, and some resilience measures over 2-year period	Pre- and post- treatment	Lack of experimental control regarding maturation effects (despite using the Intervention Efficiency

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

			in isolation			Third Revision (AAPS-3) and perceptions of resilience judged by parents and SLPs			Index and Proportional Change Index) and lack of control of covariate, including other potential intervention over the same period. No control group. No follow-up or generalisation data
McCabe 2014	4 males aged 5.5 to 8.6 years with CAS. 2 children had additional auditory processing impairments	Not quasi-/ RCT (Single case (AB) design with 1 month follow-up)	Rapid Syllable Transition Treatment (ReST)	Motor	60-minute session, 4 × per week for 3 weeks (12 sessions in total). Minimum of 1200 trials per session	Articulation, prosodic and simultaneous articulation and prosodic accuracy on trained and untrained probe pseudo words; PCC, PVC and per cent lexical stress matches from connected speech; PPVT-4 as control	All 4 participants increased perceptual accuracy. 1/4 participants showed change in untreated items. All participants showed change in prosody (average prosody gain 58%, 3/4 in PVC and 2/4 in PCC; average gain 79%)	Baseline × 2, probes in treatment × 2, 1 month follow-up	There was no immediate post-treatment data taken to determine treatment effects, the follow-up data was 1 month post-treatment and included a withdrawal phase. There was no statistical analysis of connected speech

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

						data	. Control data (receptive vocabulary on PPVT-IV) changed minimally		data. 1 participant reached ceiling. No blinding of assessors. No stimulus generalisation measures
McNeill 2009a	12 children (9 males, 3 females) aged 4.2 to 7.6 years with CAS	Not quasi-/ RCT (Case series design)	Integrated Phonological Awareness Intervention	Linguistic	45-minute session; 2 × per week for 6 weeks in 2 blocks with 6-week withdrawal between blocks. Total of 245 sessions	Trained speech accuracy and phonological awareness accuracy on a probe. Generalisation- BTOPP and first trial of DEAP inconsistency subtest for PVC, PVC and inconsistency score. PIPA for 4-year-olds. TOPA for 5 to 7-year-olds. Burt Word Reading Test for non-word reading and informal non-word	Speech: 9/12 children improved on trained items. Phonological awareness: 8/12 children improved in 1 or both intervention blocks. Generalisation for 8/12 on all measures except Burt Word Reading Test	Pre- and post-treatment	Lack of experimental control, control group or control data. CAS diagnosis unclear regarding prosody. Limited information provided on each participant. Limited treatment phase data. No maintenance data. No blinding of assessors

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

						reading probe (Gillon 2000). Per cent grapheme correct score in spelling 10 words from DEAP inconsistency subtest			
McNeill 2009b	2 male identical twins aged 4.5 years with CAS (deletion at 10q21.2-22.1)	Not quasi-/ RCT (Single case design)	Integrated Phonological Awareness intervention	Linguistic	45-minute session; 2 × per week for 6 weeks in 2 blocks with 6-week withdrawal between blocks. Total of 245 sessions	PPC, PVC on BTOPP, and DEAP inconsistency percentage. PIPA, PhonRep, Burt Word Reading, Non-word Reading, Neale accuracy and comprehension	PCC and PVC improved at post-treatment and follow-up. Reduced inconsistency. Sound-letter knowledge increased from 0 to 7 at post-treatment. Reading WNL and spelling demonstrated use of strategies at final follow-up	Pre- and post- treatment, and 6-month follow-up	Lack of experimental control, control group or control data. CAS diagnosis unclear regarding prosody. Limited information provided on each participant. Limited treatment phase data. No maintenance data. No blinding of assessors. No stimulus generalisation measures
McNeill 2010	12 children (9 males, 3 females)	Not quasi-/RCT (12-month follow-up to	Integrated Phonological Awareness inter-	Linguistic	As per McNeill 2009a	BBTOP and 1st trial of	Significant difference for CAS	1-year follow-up to McNeill	7/ 12 of original partic-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	aged 4.2 to 7.6 years diagnosed with CAS	2009 case series)	vention			DEAP yielding PPC. PIPA for 4-year-olds & TOPA for 5 to 7-year-olds. Decoding measures (Burt Word Reading Test and Non-word Reading Task) and spelling measures (probe of 10 words from the DEAP inconsistency subtest) were completed for participants at least 6 years of age at the beginning of the study. The NARA was administered for participants aged 5 years, 6 months and up	group from pre- to post-treatment on letter knowledge, non-word reading probe, spelling, PCC, TOPA and Burt Non-Word Reading. 3/7 improved on NARA to age-appropriate level	2009a	ipants followed up. Whole group data - case series. No control group or control data for experimental control or maturation effects
Moriarty 2006	3 children (2 males, 1 female) aged 6.3, 6.	Not quasi-/RCT (Single case multiple base-	Integrated Phonological Awareness Intervention	Linguistic	45-minute sessions 3 × per week for 3 weeks	PPC on probe, phoneme segmentation probe,	2/3 significantly increased PPC, 2/3 signif-	Baseline and post-treatment (3 probes each)	Lack of control group and control data. CAS

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	10 and 7.3 years with CAS	line design across behaviours)				phoneme manipulation probes, initial sound identification probes, letter - sound knowledge subtest from the PIPA, non-word reading tasks	icantly improved phonological awareness skills on probes, letter- sound knowledge, and non-word reading. Limited transfer to untreated words		diagnosis unclear regarding prosody. Lack of multiple baseline data throughout treatment. No long-term follow-up. No blinding of assessors
Namasivayam 2013	12 children (9 males, 3 females) aged 3 to 6 years with speech sound disorders	Not quasi-/ RCT (Case series pre-post design)	Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT)	Motor	45-minute session 2 × per week for 8 weeks	GFTA2, HCAPP, VMPAC focal motor and sequencing subtests, Children's Speech Intelligibility Measure	Significant gains as a group for all speech measures	Baseline 1 week prior to treatment, and 1 week post-treatment	Lack of experimental control, control group, multiple baseline or control data. No blinding of assessors. No blinding of assessors. No long-term follow-up
Namasivayam 2015	37 children (28 males, 9 females) aged 2.6 to 4.5 years with CAS	Not quasi-/ RCT (pre-postgroup design)	Motor Speech Treatment Protocol (MSTP)	Motor	Intense treatment group: 45-minute session, 2 × per week × 10 weeks = 20 sessions. Less intense group: 45-minute	GFTA-2 sounds in words subtest; speech intelligibility using Children's Speech Intelligibility Measure	Intense group had greater changes in articulation and functional communication compared to the less	Pre- and post- treatment	No control group or control data. Participants were not directly randomised; however, no be-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

					session, 1 × per week × 10 weeks = 10 sessions	(CSIM) at word level, and Beginner's Intelligibility Test (BIT) at sentence level. Functional Outcomes for Children Under Six (FOCUS) scale	intense group with large effect sizes. Mixed results were found for intelligibility: at word-level (CSIM), both the less intense and 1/2 intense groups made a significant and large change. At sentence level, 1/2 intense groups made a significant change		tween-group differences were found at baseline. There were missing data (dealt with using intention-to-treat analysis) . No information on session trials was obtained, which is important for intensity calculations
Preston 2013	6 males aged 9 to 15 years with CAS. 1 child had additional ADHD and another child had additional dysarthria	Not quasi-/RCT (Single case multiple baseline across behaviours across participants)	Ultrasound biofeedback (targeting articulation on clusters and CV or VC sequences of inaccurate phones)	Motor (instrumentally based)	60 minute sessions, 2 × per week × 18 sessions (at least 150 trials per session)	Probe of whole-word accuracy of treated and untreated items	U002 and U007 had significant gains on 2/4 treated combinations, U005 for 3/4, and U008, U009 and U012 had significant gains on all treated combinations. All exhibited some gen-	Probes at baseline × 3, every treatment session, post-treatment, and 2 months post-treatment	No control group or comparison treatment. No blinding of assessors. Untreated items were not clearly selected as control or generalisation data with some showing change and others not

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							eralisation (target-dependant). U005, U007, U008, U009, U012 demonstrated maintenance above pre-treatment levels		
Preston 2016	3 male children aged 11 to 13 years diagnosed with CAS and poor expressive language and phonological processing. 1 participant had additional flaccid dysarthria, ADHD, language and learning difficulties	Not quasi-/RCT (Single case multiple baseline across behaviours (syllable positions))	Ultrasound biofeedback (using structured chaining and principles of motor learning)	Motor (instrumentally based)	1 hour sessions × 14 sessions. Sessions 1 to 7 addressed target 1 and sessions 8 to 14 addressed target 2 with randomly assigned prosody or no prosody conditions	Treatment acquisition data, generalisation probe of untreated words, maintenance to 2 months post-treatment	2/3 participants acquired accurate articulation. 0/3 demonstrated generalisation or maintenance	3 × baseline probes, midway therapy probe, post-therapy probe (within 1 week after treatment), and 2-month follow-up	No control group. Greater within-treatment probes and post-treatment probes would have allowed for greater statistical analysis. No control data. No blinding of assessors. No stimulus generalisation measures
Preston 2017	3 males aged 11 to 14 years with CAS	Not quasi/RCT (Single case (ABA) design)	Ultrasound biofeedback (using structured chaining and principles of mo-	Motor (Instrumentally based)	2 × 1-hour articulation treatment a day for 2 weeks. 16 hours of therapy in total. Over	Treatment acquisition of /ɹ/, /s/ or /ʧ/. Generalisation to untrained items using a probe	Case 1 had acquisition, generalisation, and maintenance of targets. Case 2 had some	Probe conducted 1 × before treatment, at the end of the first week, and at the end of the sec-	Lack of experimental control, multiple baselines or control data. No blinding of as-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

			tor learning.)		100 trials per session	and sentence imitation task, and maintenance 1 to 3 weeks post-treatment (audio-samples submitted)	acquisition in the 2nd week of therapy and no generalisation and maintenance. Case 3 showed acquisition, limited generalisation to words and not phrases, and no maintenance	ond week (post-treatment)	sessors. No long-term follow-up data. No stimulus generalisation measures
Ray 2003	1 adult with CAS and class III malocclusion. Another 5 adults aged 18 to 23 years with persistent articulation disorders	Not quasi-/ RCT (Case series)	Orofacial myofunctional therapy	Motor (Instrumentally based)	45-minute session, 1 × per week for 6 weeks	Dworkin-Culatta Oral Mechanism Examination for oral postures and intelligibility in single words, sentences, and spontaneous speech	All improved lips and tongue postures. 5/6 participants increased intelligibility. No improvement in intelligibility for person with DVD	Pre- and post-treatment	Lack of experimental control, multiple baselines or control data. No treatment data or follow-up reported. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of as-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									sessors. No immediate post-treatment data or generalisation data. No replication across participants
Rosenbek 1974	1 female aged 9 years with CAS	Not quasi-/ RCT (Case study)	Intensive, systematic drill motor therapy	Motor	22 sessions over 3 months	20-item probe of /r/ (target), ineligibility in spontaneous speech	/r/ improved from 0 to 20 correct in probe. Intelligibility judged by unfamiliar listeners improved	Treatment sessions	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No follow-up data. Only ancedotal generalisation data. No statistical analysis. No reliability of judgments reported. No replication across participants
Rosenthal 1994	4 children (3 males, 1 female) aged 10-14 years diagnosed with CAS	Not quasi-/ RCT (Single subject (ABAB) design)	Rate Control Therapy	Linguistic and motor	20-minute session per reading passage. No further information available	Articulation accuracy (words read correctly)	Improved to 85% accuracy at 50% habitual rate and maintained in therapy	Reading rate in 5-minute intervals	Lack of control and follow-up data. CAS diagnosis unclear

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							as rate was slowly increased. Limited generalisation to conversation - therapy implemented		and not replicable. No statistical analysis. No blinding of assessors. No stimulus generalisation measures. No report of data reliability
Skelton 2014	3 children (2 males, 1 female) aged 4 to 6 years diagnosed with CAS	Not quasi-/RCT (Single case multiple baseline design across participants)	Concurrent treatment (using randomised variable practice)	Motor	Therapy until target sounds reached 80% accuracy. P1 had 26, P2 had 12 and P3 had 28 sessions. 2 × per week, 30 minutes per session and on average 100 to 115 trials per session	Per cent correct productions on /s, z, f, v/ trained targets during baseline and treatment; generalisation probes to untrained words and 3-word phrases	All children reached 80% accuracy on target sounds. Moderate to large generalisation effects at word and 3-word phrases levels (70% to 100% accuracy)	3 × baseline probes, probes every 5 therapy sessions	No post-treatment or follow-up/maintenance data. No blinded assessors. No stimulus generalisation data. P3 continued regular school therapy during the study so could be a confounding factor. No stimulus generalisation measures
Stokes 2010	1 male aged 7 years with residual CAS	Not quasi-/RCT (Single case (ABA) design)	Articulation with facilitative vowel contexts	Linguistic	45- to 55-minute session, 3 × per week for 3 weeks. 60+	Accuracy on 'sh' sound in word initial probe, 'tr' as con-	Significant improvement in 'sh' articulation accuracy	Pre-treatment, mid-therapy × 2 (after sessions 3 and 6),	Participant did not meet current CAS criteria. Lack

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

					trials per session. Home practice provided	trol	in trained and untrained words. No change in control words with 'tr' initial	post-treatment, and maintenance (2 weeks post-treatment)	of generalisation data beyond 'sh' sound. No blinded assessors. No replication across participants. No long-term follow-up data. No reliability of data reported
Strand 2000	1 female aged 5 years with "severe motor planning deficits but no dysarthria" (CAS)	Not quasi-/RCT (Single case multiple baseline design)	Integral stimulation	Motor	30- to 50-minute session, 3 to 5 × per week (1 to 2 × per day) for 10 to 16 sessions. No home practice	Articulation accuracy ratings on a 2-point scale	Improvement from 0.25 to 0.80 on 2-point scale. 4/5 treatment stimuli achieved rating of 2/2 by end of therapy	Treated stimuli at start of each session, control stimuli twice a week	No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up data or generalisation data. No replication across participants
Strand 2006	4 males aged 5.5 to 6.1 years with CAS (2 with dysarthria and 1 with mild intellectual disability)	Not quasi-/RCT (Single case multiple baseline across participants)	Dynamic Temporal and Tactile Cueing	Motor	30-minute sessions, 2 × per day for 5 days a week for 38 to 50 sessions	Articulation accuracy on a 3-point scale	Treatment gains for 3/4 participants maintained by 2/4	Baseline × 4 (or more, staggered baseline), 20+ treatment probes	No follow-up or generalisation data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

Thomas 2014	4 children (2 males, 2 females) aged 4.8 to 8 years with CAS	Not quasi-/RCT (Single case multiple baseline across participants and behaviours)	Rapid Syllable Transition Treatment (ReST)	Motor	50 minute sessions 2 × per week for 6 weeks. 100 trials per session	Accuracy on imitated (a) treated words, (b) untreated pseudo words, (c) untreated real words and control words	Significant improvement on treated words and untreated real words. Significant improvement for 2/4 participants on untreated pseudo words. No change in control items	Baseline × 3 to 6, treatment × 3, and 1 day, 1 month and 4 months post-treatment	Use of GFTA2 for control items. No stimulus generalisation data
Thomas 2016	5 children (4 males, 1 female) aged 5 to 11 years with CAS (3 with mild or moderate receptive language disorder)	Not quasi-/RCT (Single case multiple baseline across participants)	Rapid Syllable Transition Treatment (ReST)	Motor (instrumentally based - telehealth)	60-minute session, 4 times a week for 3 weeks (12 sessions in total). Minimum of 1200 trials per session	Accuracy on treated pseudword items, generalisation to untreated non-words and real words, and control items (articulation of rhotics) on a probe; client/family satisfaction with telehealth treatment	5/5 participants demonstrated significant change in treated items. 4/5 maintained gains to 4 months post-treatment. 4/5 had significant generalisation to untrained non-words and real words, and 1/5 demonstrated change in control	At least 3 baseline probes, 3 therapy probes (sessions 5, 9 and 1 day post-treatment). Follow-up at 1 week, 4 weeks & 4 months post-treatment	Missing data for some participants at certain time points in Table 3. Problems with change in control data. Some internet issues (dropouts, port sound quality, etc.) were observed in 61% of sessions; however, significant outcomes were

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							data (articulation errors of rhotics or /s/). Families very satisfied and motivated by telehealth treatment		found. No stimulus generalisation data
Tierney 2016	1 male aged 3 years with CAS and fine motor delay	Not quasi-/RCT (Single case design; descriptive)	Multimodal therapy: Signed Exact English sign language, Sarah Rosenfeld Johnson's oromotor programme and Kaufman Speech Praxis Program	Augmentative and alternative communication	Clinic-based sessions 45 minutes 1 to 2 × per week and home-based sessions for 60 minutes 1 × per week	Language assessment; observations and Kaufman Speech Praxis Test; Verbal Motor Production Assessment for Children (VMPAC)	Receptive and expressive language consistently in average range but receptive relatively better than expressive language. By 3.6 years of age receptive and expressive language same level. Marked drooling and limited inventory and sequencing at 18 months, yet skills on Kaufman & VMPAC in average range at	Language assessment at 1.1 year, 3 years and 3.6 years. Kaufman test or observations at 1. 6, 3 and 3.9 years. VMPAC at 3 years, 9 months	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable regarding prosody and drooling. No statistical analysis. No blinding of assessors. No replication across participants. Limited repeated measures on same instrument. Participant had multiple ther-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

							3 years, 9 months. Discharged from therapy		apies concurrently
Vashdi 2013	1 male aged 14 years with severe CAS and limb/motor apraxia and obsessive compulsive disorder	Not quasi-/ RCT (Case study)	Verbal Motor Learning (with Dynamic Distal Stabilization Technique (DDST))	Motor	1 × 30-minute clinic session and 6 × home practice sessions a week for 4 weeks	(1) Producing highest pitch using /I/ sound with and without DDST, to determine minimum and maximum frequency and length using Speech Analyser 1.5 (2) Imitation of 18 words to analyse word length, maximum loudness, maximum and minimum frequency	Significant t-test results for (1) increase in maximum frequency and length of pitch after DDST, no change in minimum frequency, and (2) decrease in word length (word said faster), maximum loudness, and maximum frequency	Pre- and post- treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up or generalisation data. No replication across participants. Unclear data analysis procedures (unclear if they used visual analysis or perceptual analy-

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

									sis, and if they tested assumptions for the statistical analysis completed)
Vashdi 2014	1 female aged 10 years with CAS and ASD	Not quasi-/ RCT (Case study)	Verbal Motor Learning (Initial Phoneme Cue (IPC) technique)	Motor	2 × 1 hour sessions, 2 weeks apart (participant had initial therapy: 1-hour session weekly for 1 year prior to this study)	Imitation accuracy of CVCV treated words either (a) with IPC or (b) without IPC	Imitation of CVCV was 0% to 22% accuracy and imitation with IPC was 96% to 100% accuracy	Pre- and post- treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No statistical analysis. No blinding of assessors. No follow-up or generalisation data. No replication across participants
Yoss 1974	10 children (no information on gender reported) aged 6 to 11 years with moderate to se-	Not quasi-/RCT (Case descriptions/ file audit)	School-based intervention	Motor	25 to 307 hours of therapy	Articulation, polysyllable words and connected speech in speech samples. Intelligi-	Significant improvement on articulation. Minimal generalisation to polysyllable words	Pre- and post- treatment	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear

Table 1. Excluded, low-quality evidence from observational studies (case-series, case control) (*Continued*)

	vere DAS					bility rated on a 9-point scale	and connected speech. Intelligibility improved by at least 0.5 points		and not replicable. No statistical analysis. No blinding of assessors. No follow-up data
Zaretsky 2010	1 female aged 11.6 years with CAS, intellectual disability and language disorder	Not quasi-/ RCT (Single case design)	Phonological awareness (phoneme grapheme mapping, reading comprehension, 'Basics' programme). Speech - PROMPT and Moving Across Syllables	Linguistic	Between 6.0 and 11.6 ongoing weekly treatments - 1 hour × 1:1 sessions and PROMPT institute over summer	Per cent accuracy on phonological awareness and decoding	Improvement seen in phoneme grapheme mapping, segmentation and short vowel identification. Some improvement in decoding	Ongoing 1 × per week sessions from 6.0 to 11.6 years	Lack of experimental control, multiple baselines or control data. CAS diagnosis unclear and not replicable. No statistical analysis. Limited outcome measures. No blinding of assessors. No follow-up or generalisation data. No replication across participants. Difficult to replicate measures and treatment used

Participants: All participants are English speakers unless otherwise reported.
AOS: apraxia of speech; **BBTOP**: Bankson-Bernthal Test of Phonology; **CAS**: childhood apraxia of speech; **CSIP**: consonant substitute inconsistency percentage; **DAS**: developmental apraxia of speech; **DEAP**: Diagnostic Evaluation of Articulation and Phonology; **DVD**: developmental verbal dyspraxia; **GDD**: global developmental delay; **GFTA-2**: Goldman Fristoe Test of Articulation 2; **HCAPP**: Hodson Computerized Analysis of Phonological Patterns; **ISP**: inconsistency severity percentage; **KLPA-2**: Khan-Lewis Phonological Analysis,

Second Edition; **NARA**: Neale Analysis of Reading Ability; **PCC**: percentage consonants correct; **PDD-NOS**: pervasive developmental disorder - not otherwise specified; **PMLU**: phonological mean length of utterance; **PVC**: percentage vowels correct; **PWC**: percentage words correct; **PWP**: proportion of whole-word proximity; **PIPA**: Preschool and Primary Inventory of Phonological Awareness; **RCT**: randomised control trial; **SSD**: speech sound disorder; **TOCS+**: Test of Children's Speech Plus; **TOPA**: Test of Phonological Awareness; **VMPAC**: Verbal Motor Production Assessment for Children

APPENDICES

Appendix 1. Search strategies 2007 onwards

Cochrane Central Register of Controlled Trials (CENTRAL), in the Cochrane Library, and which includes the Cochrane Developmental, Psychosocial and Learning Problems Specialised Register

Searched 6 April 2017 (172 records)
Searched 6 June 2014 (103 records)
Searched 4 August 2011 (62 records)
1MeSH descriptor: [Apraxias] explode all trees
#2MeSH descriptor: [Speech Disorders] this term only
#3dysprax*
#4aprax*
#5prax*
#6(speech near/3 disorder*)
#7(speech near/3 impair*)
#8(speech near/3 problem*)
#9(speech near/3 difficult*)
#10voice near/3 disorder*
#11voice near/3 impair*
#12voice near/3 problem*
#13voice near/3 difficult*
#14vocal near/3 disorder*
#15vocal near/3 impair*
#16vocal near/3 problem*
#17vocal near/3 difficult*
#18communication near/3 disorder*
#19communication near/3 impair*
#20communication near/3 problem*
#21communication near/3 difficult*
#22#1 or #2 or #3 or #4 or #5 or #6 or #7 or #8 or #9 or #10 or #11 or #12 or #13 or #14 or #15 or #16 or #17 or #18 or #19 or #20 or #21
#23MeSH descriptor: [Adolescent] this term only
#24MeSH descriptor: [Child] 1 tree(s) exploded
#25(child* or girl* or boy* or pre next school* or pre-school*)
#26#23 or #24 or #25
#27#22 and #26 in Trials

MEDLINE Ovid

Searched 6 April 2017 (960 records)
Searched 6 June 2014 (896 records)
Searched 4 August 2011 (759 records)
1 exp Apraxias/
2 Speech disorders/
3 dysprax$.tw.
4 aprax$.tw.
5 prax$.tw.
6 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
8 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
9 or/1-8
10 adolescent/
11 exp Child/
12 (adolescen$ or child$ or girl$ or boy$ or pre school$ or pre-school$ or teen$).tw.
13 or/10-12
14 speech therapy/
15 language therapy/
16 (therap$ or train$ or measur$ or assess$ or habilitat$ or rehabilitat$ or manage$ or assist$ or treat$ or remedia$ or augment$ or recover$ or intervent$).tw.
17 or/14-16
18 9 and 13 and 17
19 limit 18 to yr="2007 -Current"20 limit 18 to ed=20110401-20140529
21 limit 18 to ed=20140501-20170324

MEDLINE Epub Ahead of Print Ovid

Searched 6 April 2017 (10 records)
1 dysprax$.tw.
2 aprax$.tw.
3 prax$.tw.
4 1 or 2 or 3
5 (speech$ or language$).tw.
6 4 and 5
7 (child$ or boy$ or girl$ or preschool$ or preschool$ or teen$ or adolesc$).tw.
8 6 and 7

MEDLINE In-Process and Other Non-Indexed Citations Ovid

Searched 6 April 2017 (30 records)
1 dysprax$.tw.
2 aprax$.tw.
3 prax$.tw.
4 1 or 2 or 3
5 (speech$ or language$).tw.
6 4 and 5
7 (child$ or boy$ or girl$ or preschool$ or preschool$ or teen$ or adolesc$).tw.
8 6 and 7

Embase Ovid

Searched 10 April 2017 (1237 records)

Searched 6 June 2014 (1356 records)
Searched 4 August 2011 (1011 records)
1 exp Apraxias/
2 "apraxia of speech"/
3 dysprax$.tw.
4 aprax$.tw.
5 prax$.tw.
6 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw. .
8 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
9 or/1-8
10 adolescent/
11 child/ or preschool child/
12 (adolescen$ or child$ or girl$ or boy$ or pre school$ or pre-school$ or teen$).tw.
13 or/10-12
14 speech rehabilitation/
15 speech therapy/
16 (therap$ or train$ or manage$ or assist$ or measure$ or treat$ or assess$ or remedia$ or augment$ or recover$ or intervent$).tw.
17 or/14-16
18 9 and 13 and 17
19 limit 18 to yr="2007 -Current"
20 limit 18 to yr="2011 -Current"
21 limit 18 to yr="2014 -Current"

CINAHL Plus EBSCOhost (Cumulative Index to Nursing and Allied Health Literature)

Searched 10 April 2017 (376 records)
Searched 6 June 2014 (571 records)
Searched 4 August 2011 (866 records)
S23 S17 AND S22
S22 EM 20140601-
S21 S17 AND S20
S20 EM 20110401-
S19 S17 and S18
S18 EM >=20070101
S17 S13 and S16
S16 S14 or S15
S15 (MH "Rehabilitation, Speech and Language") OR (MH "Speech Therapy+") OR (MH "Language Therapy") OR (MH "Voice Therapy")
S14 (therap* or train* or rehabilitat* or manage* or assist* or measure* or treat* or assess* or remedia* or augment* or recover* or intervent*)
S13 S9 and S12
S12 S10 or S11
S11 child* or girl* or boy* or pre school* or pre-school*
S10 (MH "Child") OR (MH "Child, Preschool")
S9 S1 or S2 or S3 or S4 or S5 or S6 or S7 or S8
S8 (communication N3 disorder*) or (communication N3 impair*) or (communication N3 problem*) or (communication N3 difficult*)
S7 (vocal N3 disorder*) or (vocal N3 impair*) or (vocal N3 problem*) or (vocal N3 difficult*)
S6 (voice N3 disorder*) or (voice N3 impair*) or (voice N3 problem*) or (voice N3 difficult*)
S5 (speech N3 disorder*) or (speech N3 impair) or (speech N3 problem*) or (speech N3 difficult*)
S4 prax*
S3 aprax*
S2 dysprax*

S1 (MH “Apraxia+”)

PsycINFO Ovid

Searched 10 April 2017 (600 records)
Searched 6 June 2014 (902 records)
1 apraxia/
2 speech disorders/
3 dysprax$.tw.
4 aprax$.tw.
5 prax$.tw.
6 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
8 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
9 or/1-8
10 (adolescen$ or child$ or girl$ or boy$ or pre school$ or pre-school$ or teen$).tw.
11 (adolescence 13 17 yrs or childhood birth 12 yrs or preschool age 2 5 yrs or school age 6 12 yrs).ag.
12 10 or 11
13 Speech Therapy/
14 Language Therapy/
15 Speech Language Pathology/
16 intervention/
17 Rehabilitation/
18 (therap$ or train$ or measur$ or assess$ or rehabilitat$ or manage$ or assist$ or treat$ or remedia$ or augment$ or recover$ or intervent$).tw.
19 or/13-18
20 9 and 12 and 19

PsycINFO EBSCOhost

Searched 4 August 2011 (2409 records)
S31 S11 and S15 and S30
S30 S17 or S18 or S19 or S20 or S21 or S22 or S23 or S24 or S25 or S26 or S27 or S28 or S29
S29 (evaluation N3 stud* or evaluation N3 research*)
S28 (effectiveness N3 stud* or effectiveness N3 research*)
S27 DE “Placebo” or DE “Evaluation” or DE “Program Evaluation” OR DE “Educational Program Evaluation” OR DE “Mental Health Program Evaluation”
S26 (DE “Random Sampling” or DE “Clinical Trials”) or (DE “Experiment Controls”)
S25 “cross over*”
S24 crossover*
S23 (tripl* N3 mask*) or (tripl* N3 blind*)
S22 (trebl* N3 mask*) or (trebl* N3 blind*)
S21 (doubl* N3 mask*) or (doubl* N3 blind*)
S20 (singl* N3 mask*) or (singl* N3 blind*) S
S19 (clinic* N3 trial*) or (control* N3 trial*)
S18 (random* N3 allocat*) or (random* N3 assign*)
S17 randomis* or randomiz*
S16 S12 and S15
S15 S13 or S14
S14 AG childhood Limiters - Age Groups: Childhood (birth-12 yrs)
S13 (child* or girl* or boy* or pre school* or pre-school*)
S12 S10 and S11
S11 S1 or S2 or S3 or S4 or S5 or S6 or S7 or S8 or S9

S10 therap* or train* or rehabilitat* or manage* or assist* or measure* or treat* or assess* or remedia* or augment* or recover* S
S9 (communication N3 disorder*) or (communication N3 impair*) or (communication N3 problem*) or (communication N3 difficult*)
S8 (vocal N3 disorder*) or (vocal N3 impair*) or (vocal N3 problem*) or (vocal N3 difficult*)
S7 (voice N3 disorder*) or (voice N3 impair*) or (voice N3 problem*) or (voice N3 difficult*)
S6 (speech N3 disorder*) or (speech N3 impair*) or (speech N3 problem*) or (speech N3 difficult*)
S5 prax*
S4 aprax*
S3 dysprax*
S2 DE "Speech Disorders"
S1 DE "Apraxia"

ERIC EBSCOhost (Education Resources Information Center)

Searched 10 April 2017 (293 records)
S1 DE "Speech Impairments" OR DE "Articulation Impairments" OR DE "Voice Disorders"
S2 verbal apraxia of speech
S3 aprax*
S4 dysprax*
S5 prax* N10 speech*
S6 (speech n3 disorder*)
S7 (speech n3 impair*)
S8 (speech n3 problem*)
S9 (speech n3 difficult*)
S10 voice n3 disorder*
S11 voice n3 impair*
S12 voice n3 problem*
S13 voice n3 difficult*
S14 vocal n3 disorder*
S15 vocal n3 impair*
S16 vocal n3 problem*
S17 vocal n3 difficult*
S18 communication n3 disorder*
S19 communication n3 impair*
S20 communication n3 problem*
S21 ommunication n3 problem* [Note: Input error. Correct in line 20]
S22 communication n3 difficult*
S23 S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22
S24 DE "Speech Improvement" OR DE "Speech Therapy"
S25 (therap* or train* or rehabilitat* or manage* or assist* or measure* or treat* or assess* or remedia* or augment* or recover* or rehab*)
S26 S24 OR S25
S27 S23 AND S26
S28 DE "Adolescents" OR DE "Early Adolescents" OR DE "Late Adolescents"
S29 DE "Children" OR DE "Preadolescents" OR DE "Young Children"
S30 (adolescen* or child* or girl* or boy* or pre school* or pre-school* or teen*)
S31 S28 OR S29 OR S30
S32 S27 AND S31
S33 YR 2014-
S34 S32 AND S33
S35 YR 2017-
S36 S32 AND S35

ERIC Proquest

Searched 6 June 2014 limited to publication year =2011-2014 (379 records)
Searched 4 August 2011 limited to publication year =2007-2011 (321 records)
"((((APRAX$.TI,AB.) OR (DYSPRAX$.TI,AB.) OR (PRAX$.TI,AB.) OR ((SPEECH NEAR (DISORDER$1 OR IMPAIR$4 OR PROBLEM$1 OR DIFFICULT$3)) .TI,AB.) OR (((VOICE OR VOCAL) NEAR (DISORDER$1 OR IMPAIR$4 OR PROBLEM$1 OR DIFFICULT$3)) .TI,AB.) OR (COMMUNICATION NEAR (DISORDER$1 OR IMPAIR$4 OR PROBLEM$1 OR DIFFICULT$3))) .TI,AB.) AND ((CHILD$3 OR GIRL$1 OR BOY$1 OR PRE ADJ SCHOOL$ OR ADOLESCEN$3 OR TEEN$5) .TI,AB.)) AND ((SPEECH-THERAPY.DE.) OR (INTERVENTION#.W..DE.) OR ((THERAP$4 OR TRAIN$3 OR REHABILITAT$3 OR assess$5 OR measur$4 OR MANAGE$4 OR ASSIST$3 OR TREAT$5 OR REMEDIA$4 OR AUGMENT$2 OR RECOVER$1 OR INTERVENTION$1) .TI,AB.))

Cochrane Database of Systematic Reviews (CDSR), part of the Cochrane Library

Searched 10 April 2017 (5 records)
#1MeSH descriptor: [Apraxias] explode all trees
#2MeSH descriptor: [Speech Disorders] this term only
#3dysprax*:ti
#4aprax*:ti
#5prax*:ti
#6(speech near/3 disorder*):ti,ab
#7(speech near/3 impair*):ti,ab
#8(speech near/3 problem*):ti,ab
#9(speech near/3 difficult*):ti,ab
#10{or #1-#9}
#11MeSH descriptor: [Adolescent] this term only
#12MeSH descriptor: [Child] 1 tree(s) exploded
#13(child* or girl* or boy* or pre next school* or pre-school*):ti,ab
#14#11 or #12 or #13
#15#10 and #14 in Cochrane Reviews (Reviews and Protocols)

Database of Reviews of Effect (DARE), part of the Cochrane Library

Searched 10 April 2017 (8 records)
#1MeSH descriptor: [Apraxias] explode all trees
#2MeSH descriptor: [Speech Disorders] this term only
#3dysprax*:ti
#4aprax*:ti
#5prax*:ti
#6(speech near/3 disorder*):ti,ab
#7(speech near/3 impair*):ti,ab
#8(speech near/3 problem*):ti,ab
#9(speech near/3 difficult*):ti,ab
#10{or #1-#9}
#11MeSH descriptor: [Adolescent] this term only
#12MeSH descriptor: [Child] 1 tree(s) exploded
#13(child* or girl* or boy* or pre next school* or pre-school*):ti,ab
#14#11 or #12 or #13
#15#10 and #14 in Other Reviews

SpeechBITE (speechbite.com)

Searched 10 April 2017 (27 records)
Basic search: "childhood apraxia"

Advanced search:
Practice Area: Apraxia / Dyspraxia
Research Design: Randomised Controlled Trial

Australian New Zealand Clinical Trials Registry (ANZCR; anzctr.org.au/BasicSearch.aspx)

Searched 10 April 2017 [5 records]
Searched 20 June 2014 [2 records]
Advanced search
speech AND apraxia limited to children

Chinese Clinical Trial Registry (ChiCTR; www.chictr.org.cn/index.aspx)

Searched 10 April 2017 (0 records)
(childhood apraxia of speech) or (dyspraxia) or (apraxia), (child) AND (speech)

ClinicalTrials.gov (clinicaltrials.gov)

Searched 10 April 2017 (3 records)
Searched 20 June 2014 (12 records)
Condition: apraxia OR dyspraxia Limited to children 0-17

EU Clinical Trials Register (clinicaltrialsregister.eu)

Searched 10 April 2017 (0 records)
(childhood apraxia of speech) or (dyspraxia) or (apraxia), (child) AND (speech)

ISRCTN Registry (www.isrctn.com)

Searched 10 April 2017 (0 records)
(childhood apraxia of speech) or (dyspraxia) or (apraxia), (child) AND (speech)

Nederlands Trial Register (www.trialregister.nl/trialreg/index.asp)

Searched 10 April 2017 (0 records)
(childhood apraxia of speech) or (dyspraxia) or (apraxia), (child) AND (speech)

World Health Organization International Clinical Trials Registry Platform (WHO ICTRP; apps.who.int/trialsearch)

Searched 10 April 2017 (8 records)
Searched 20 June 2014 (35 records)
Searched 10 August 2011 (1 record)
Basic search: apraxia OR dyspraxia. Limited to clinical trials in children

Appendix 2. Search strategies up to 2007

Cochrane Central Register of Controlled Trials (CENTRAL), in the Cochrane Library, and which includes the Cochrane Developmental, Psychosocial and Learning Problems Specialised Register

Searched 2016, Issue 4
#1 MeSH descriptor Apraxias explode all trees
#2 dysprax*
#3 aprax*
#4 prax* 1007
#5 (speech near/3 disorder*)
#6 (speech near/3 impair*)
#7 (speech near/3 problem*)
#8 (speech near/3 difficult*)
#9 voice near/3 disorder*
#10 voice near/3 impair*
#11 voice near/3 problem*
#12 voice near/3 difficult*
#13 vocal near/3 disorder*
#14 vocal near/3 impair*
#15 vocal near/3 problem*
#16 vocal near/3 difficult*
#17 communication near/3 disorder*
#18 communication near/3 impair*
#19 communication near/3 problem*
#20 communication near/3 difficult*
#21 (#1 OR #2 OR #3 OR #4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10 OR #11 OR #12 OR #13 OR #14OR #15 OR #16 OR #17 OR #18 OR #19 OR #20)
#22 (therap* or train* or rehabilitat* or manage* or assist* or measure* or treat* or assess* or remedia* or augment* or recover* or rehab*)
#23 child near "MESH check words"
#24 (child* or girl* or boy* or pre school* or pre-school*)
#25 (#23 OR #24)
#26 (#21 AND #22 AND #25)

MEDLINE Ovid

Searched 1966 to January 2007
1 exp Apraxias/
2 dysprax$.tw.
3 aprax$.tw.
4 prax$.tw.
5 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
6 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
8 or/1-7
9 (therap$ or train$ or rehabilitat$ or manage$ assist$ or measure$ or treat$ or assess$ or remedia$ or augment$ or recover$ or rehab$).tw.
10 8 and 9
11 Child/
12 (child$ or girl$ or boy$ or pre school$ or pre-school$).tw.
13 or/11-12
14 8 and 10 and 13

15 randomized controlled trial.pt.
16 controlled clinical trial.pt.
17 randomized controlled trials.sh.
18 random allocation.sh.
19 double blind method.sh.
20 single-blind method.sh.
21 or/15-20
22 (animals not human).sh.
23 21 not 22 (362564)
24 clinical trial.pt.
25 exp Clinical Trials/
26 (clin$ adj25 trial$).ti,ab.
27 ((singl$ or doubl$ or trebl$ or tripl$) adj25 (blind$ or mask$)).ti,ab.
28 placebos.sh.
29 placebo$.ti,ab.
30 random$.ti,ab.
31 research design.sh.
32 or/24-31
33 32 not 22
34 33 not 23
35 comparative study.sh.
36 exp Evaluation Studies/
37 follow up studies.sh.
38 prospective studies.sh.
39 (control$ or prospectiv$ or volunteer$).ti,ab.
40 or/35-39
41 40 not 22
42 41 not (23 or 34)
43 23 or 34 or 42
44 14 and 43

Embase Ovid

Searched 1980 to January 2007
1 exp Apraxias/
2 dysprax$.tw.
3 aprax$.tw.
4 prax$.tw.
5 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
6 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
8 or/1-7
9 (therap$ or train$ or rehabilitat$ or manage$ assist$ or measure$ or treat$ or assess$ or remedia$ or augment$ or recover$ or rehab$).tw.
10 Child/
11 (child$ or girl$ or boy$ or pre school$ or pre-school$).tw.
12 or/10-11
13 clin$.tw.
14 trial$.tw.
15 (clin$ adj3 trial$).tw.
16 singl$.tw.
17 doubl$.tw.
18 trebl$.tw.

19 tripl$.tw.
20 blind$.tw.
21 mask$.tw.
22 ((singl$ or doubl$ or trebl$ or tripl$) adj3 (blind$ or mask$)).tw.
23 randomi$.tw.
24 random$.tw.
25 allocat$.tw.
26 assign$.tw.
27 (random$ adj3 (allocat$ or assign$)).tw.
28 crossover.tw.
29 28 or 27 or 23 or 22 or 15
30 exp Randomized Controlled Trial/
31 exp Double Blind Procedure/
32 exp Crossover Procedure/
33 exp Single Blind Procedure/
34 exp RANDOMIZATION/
35 30 or 31 or 32 or 33 or 34 or 29
36 8 and 9 and 12 and 35

CINAHL Ovid

Searched 1982 to December 2006
1 exp Apraxias/
2 dysprax$.tw.
3 aprax$.tw.
4 prax$.tw.
5 (speech adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
6 ((voice or vocal) adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
7 (communication adj3 (disorder$ or impair$ or problem$ or difficult$)).tw.
8 or/1-7
9 (therap$ or train$ or rehabilitat$ or manage$ assist$ or measure$ or treat$ or assess$ or remedia$ or augment$ or recover$ or rehab$).tw.
10 Child/
11 (child$ or girl$ or boy$ or pre school$ or pre-school$).tw.
12 or/10-11
13 randomi$.mp. [mp=title, subject heading word, abstract, instrumentation]
14 clin$.mp. [mp=title, subject heading word, abstract, instrumentation]
15 trial$.mp. [mp=title, subject heading word, abstract, instrumentation]
16 (clin$ adj3 trial$).mp. [mp=title, subject heading word, abstract, instrumentation]
17 singl$.mp. [mp=title, subject heading word, abstract, instrumentation]
18 doubl$.mp. [mp=title, subject heading word, abstract, instrumentation]
19 tripl$.mp. [mp=title, subject heading word, abstract, instrumentation]
20 trebl$.mp. [mp=title, subject heading word, abstract, instrumentation]
21 mask$.mp. [mp=title, subject heading word, abstract, instrumentation]
22 blind$.mp. [mp=title, subject heading word, abstract, instrumentation]
23 (17 or 18 or 19 or 20) and (21 or 22)
24 crossover.mp. [mp=title, subject heading word, abstract, instrumentation]
25 random$.mp. [mp=title, subject heading word, abstract, instrumentation]
26 allocate$.mp. [mp=title, subject heading word, abstract, instrumentation]
27 assign$.mp. [mp=title, subject heading word, abstract, instrumentation]
28 (random$ adj3 (allocate$ or assign$)).mp.
29 Random Assignment/
30 exp Clinical Trials/

31 exp Meta Analysis/
32 28 or 24 or 23 or 16 or 13 or 29 or 30 or 31
33 8 and 9 and 12 and 32

PsycINFO SilverPlatter

Searched up to January 2007
#28 (((trial*) in TI) or ((randomly) in AB) or ((placebo) in AB) or ((randomized or randomised) in AB) or ("Clinical-Trials" in MJ,MN)) and ((child* or girl* or boy* or pre school* or pre-school*) and ((therap* or train* or rehabilitat* or manage* or assist* or measure* or treat* or assess* or remedia* or augment* or recover*) and ((communication near 3 difficult*) or (communication near 3 problem*) or (communication near 3 impair*) or (communication near 3 disorder*) or ((voice or vocal) near 3 (difficult*)) or ((voice or vocal) near 3 (problem*)) or ((voice or vocal) near 3 (impair*)) or ((voice or vocal) near 3 (disorder*)) or (speech near 3 difficult*) or (speech near 3 problem*) or (speech near 3 impair*) or (speech near 3 disorder*) or (prax*) or (aprax*) or (dysprax*) or ("Apraxia-" in MJ,MN))))

ERIC Dialog Datastar (Education Resources Information Center)

Searched 1966 to January 2007
1 APRAX$.TI,AB.
2 DYSPRAX$.TI,AB.
3 PRAX$.TI,AB.
4 (SPEECH NEAR (DISORDER$ OR IMPAIR$ OR PROBLEM$ OR DIFFICULT$)).TI,AB.
5 ((VOICE OR VOCAL) NEAR (DISORDER$ OR IMPAIR$ OR PROBLEM$ OR DIFFICULT$)).TI,AB.
6 (COMMUNICATION NEAR (DISORDER$ OR IMPAIR$ OR PROBLEM$ OR DIFFICULT$)).TI,AB.
7 (1 OR 2 OR 3 OR 4 OR 5 OR 6).TI,AB.
8 (THERAP$ OR TRAIN$ OR REHABILITAT$ OR MANAGE$ OR ASSIST$ OR MEASURE$ OR TREAT$ OR ASSESS$OR REMEDIA$ OR AUGMENT$ ADJ RECOVER$).TI,AB.
9 (CHILD$ OR GIRL$ OR BOY$ OR PRE ADJ SCHOOL$ OR PRE-SCHOOL$).TI,AB.
10 7.TI,AB. AND 8.TI,AB. AND 9.TI,AB.
11 (RANDOMISED OR RANDOMIZED).AB.
12 PLACEBO.AB.
13 RANDOMLY.AB.
14 TRIAL$.TI,AB.
15 11 OR 12 OR 13 OR 14
16 10 AND 15

Linguistics Abstracts Online

Searched 1985 to January 2007
Terms used:
dyspraxia AND child or children
OR
apraxia AND child or children

Appendix 3. Methods for future updates

Electronic searches

We will include non-English language abstracts in any future updates of this review.

Measures of treatment effects

Binary data

We will analyse binary outcomes by calculating the risk ratio (RR) with 95% confidence intervals (CIs). Wherever necessary, we will contact original study authors for raw data.

Continuous data

To enable the combination of studies measuring the same outcome using different methods, we will report standardised mean difference (SMD) effect sizes with 95% CIs. For studies measuring the same outcome using the same measure, we will report mean difference (MD) effect sizes with 95% CIs. Wherever necessary, we will contact original study authors for raw data (e.g. where authors have only reported change from baseline data). We will transform and include skewed data where appropriate.

Unit-of-analysis issues

In future reviews, we will continue to consider the level at which randomisation occurred (i.e. in simple parallel-group designs, as encountered in the included study here (Murray 2015), where participants were individually randomised to one of two intervention groups, and a measurement for each outcome from each participant was collected and analysed). However, if we encounter cluster-randomised trials (i.e. where groups of individuals are randomised together to the same intervention), cross-over trials or multiple observations of the same outcome (e.g. repeated measurements, recurring events. etc.), we will consult the *Cochrane Handbook for Systematic Reviews of Interventions* for the latest recommendations on best management of unit-of-analysis issues (Higgins 2011b).

Dealing with missing data

If studies do not report intention-to-treat (ITT) analyses, we will contact the study authors and request the missing data. We will initially seek missing data via contact with the corresponding author. In regard to participant dropout, if the rate of attrition reaches a 30% threshold in an included study, we will conduct a sensitivity analysis and assess the impact of this attrition. If the impact is not significant, we will include the data. The maximum allowed difference in the dropout rate between the two groups that we will allow before we exclude an included study from a meta-analysis is 10%.

Assessment of reporting biases

Where appropriate, we will use funnel plots to assess the possibility that study selection might be affected by bias, by investigating any relationship between effect size and study precision (closely related to sample size) (Morgan 2008). Such a relationship may be due to publication or related biases, to systematic differences between small and large studies, or to a statistical artefact of the chosen effect measure. We will use Egger's test to examine potential bias (Egger 1997).

Assessment of heterogeneity

We will estimate between-study variance (τ^2) using a random-effects model and the inverse-variance approach. We will use the random-effects model because it is more conservative than the fixed-effect model.

Data synthesis

We will only perform a meta-analysis when studies employ similar interventions across the three intervention types (motor-based, linguistic, multi-modal communication). We will use a network meta-analysis with a random-effects model.

WHAT'S NEW

Last assessed as up-to-date: 6 April 2017.

Date	Event	Description
29 August 2017	New citation required and conclusions have changed	One new study included in review.
29 August 2017	New search has been performed	The review was updated following a new search on 6 April 2017

HISTORY

Protocol first published: Issue 4, 2006

Review first published: Issue 3, 2008

Date	Event	Description
4 September 2015	Amended	Duplicate paragraph removed from the description of the intervention and reference error corrected in background section
13 May 2008	Amended	Converted to new review format.
12 May 2008	Amended	Change of title from protocol stage ('developmental apraxia of speech') to 'childhood apraxia of speech'

CONTRIBUTIONS OF AUTHORS

Angela Morgan (AM; guarantor of the review), Frederique Liégeois (FL) and Elizabeth Murray (EM) contributed to drafts of the review. The authors developed the search strategy in concert with CDPLPG. AM and FL conducted study selection, study assessment, data extraction, data entry, and analysis. EM tabulated further detail on excluded studies in Table 1 and contributed to the Characteristics of included studies and Characteristics of excluded studies tables. AM and FL completed the first draft of the review. AM, FL and EM contributed to further drafts of the review. EM did not contribute to the study selection, risk of bias assessment, or extraction of data from this study due to potential for conflict of interest, given that EM was lead author of the included study.

DECLARATIONS OF INTEREST

Angela T Morgan (AM) - none known.

Elizabeth Murray (EM) is an author of the included study, Murray 2015, and was not involved in selecting this study for inclusion, or extracting or reviewing data from this study. Study selection as well as data extraction and review was conducted by two independent authors - AM and FL.

Frederique J Liégeois (FL) - none known.

SOURCES OF SUPPORT

Internal sources

- None, Other.

External sources

- National Health and Medical Research Council (NHMRC), Australia.

NHMRC Practitioner Fellowship (APP1105008) awarded to AM.

- National Health and Medical Research Council (NHMRC), Australia.

NHMRC Centre of Research Excellence in Speech and Language Neurobiology (CRE-SLANG) (APP1116976) awarded to AM and FL.

- National Health and Medical Research Council (NHMRC), Australia.

NHMRC Project Grant (APP1127144) awarded to AM and FL.

DIFFERENCES BETWEEN PROTOCOL AND REVIEW

Changes between 2006 protocol and 2008 review

The title was changed from 'Intervention for developmental apraxia of speech' to 'Intervention for childhood apraxia of speech' to reflect current terminology (ASHA 2007).

Changes between 2006 protocol and 2017 review

1. Description of the intervention. We reclassified the types of interventions from 'perceptually-based therapy' and 'instrumentally-based biofeedback approaches' to 'motor-based', 'linguistic-based' and 'multi-modal communication', to reflect more contemporaneous approaches in the field.
2. Criteria for considering studies for this review. We rewrote the inclusion criteria for studies to provide greater clarity around the specific types of interventions being targeted (i.e. interventions targeting speech and language); to specify that we would include studies comparing intervention to either no treatment (e.g. wait-list) control as well as other interventions; and to specify that the CAS diagnosis had to have been made by an SLP/SLT
3. Types of outcome measures. We updated our outcome measures to reflect those used in current literature.
4. Electronic searches.
    i) We increased the sensitivity of our search by adding additional search terms for the condition and intervention.
    ii) We added the following databases and trial registers to our electronic searches, to ensure our search was as comprehensive as possible:
        a) *Cochrane Database of Systematic Reviews;*
        b) MEDLINE E-Pub Ahead of Print and MEDLINE In-Process and Other Non-Indexed Citations, both of which are updated daily.

c) Database of Abstracts of Reviews of Effect (DARE); however, this was not searched in 2017, as DARE was last updated in 2015;

d) SpeechBITE;

e) Chinese Clinical Trial Registry (ChiCTR);

f) EU Clinical Trials Register;

g) ISRCTN Registry; and

h) Nederlands Trial Registry.

iii) We did not search Linguistic Abstracts Online and Dissertation Abstracts because we judged these would not identify any unique studies not found in other databases.

5. Data collection and analysis. Some methodological sections involving meta-analysis as reported in the original protocol, Morgan 2006, were not relevant in this review because only a single RCT was identified for inclusion. See Appendix 3 for further detail.

6. Dealing with missing data. Whilst not used in this version of the review, we have specified that in future updates of the review, if the rate of attrition reaches a 30% threshold in an included study, we will include the study in the review but not in the meta-analysis. The maximum allowed difference in the dropout rate between the two groups will be 10% before a study included in the review is excluded from meta-analysis. See Appendix 3.

7. Data synthesis > **Summary of findings.** We used the GRADE approach in this updated review to rate the quality of the evidence (Schünemann 2017). The GRADE system was not available when the original 2006 protocol (Morgan 2006), or 2008 review (Morgan 2008), were published.

INDEX TERMS

Medical Subject Headings (MeSH)

*Speech Therapy; *Speech-Language Pathology; Apraxias [*therapy]; Speech Disorders [*therapy]

MeSH check words

Adolescent; Child; Humans