Interventions for oropharyngeal dysphagia in children with neurological impairment (Protocol)

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TABLE OF CONTENTS

[Intervention Protocol]

Interventions for oropharyngeal dysphagia in children with neurological impairment

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ABSTRACT

This is the protocol for a review and there is no abstract. The objectives are as follows:

To assess the efficacy of intervention targeting oropharyngeal dysphagia in children with neurological impairment.

BACKGROUND

Description of the condition

Dysphagia is a broad term that encompasses many subtypes of swallowing disorder. This review is focused on the efficacy of interventions for oropharyngeal swallowing impairments only, that is, difficulty with the oral preparatory phase of swallowing (chewing and preparing the food), the oral phase (moving the food or fluid posteriorly through the oral cavity with the tongue, into the back of the throat) and the pharyngeal phase (swallowing the food or fluid and moving it through the pharynx to the oesophagus). Dysphagia, which is caused by disorders of the oesophageal phase of the swallow (for example, problems such as lower oesophageal sphincter function or gastroesophageal reflux), is managed primarily by surgery and/or medication and not by behavioral interventions, which have been excluded from examination in the present review. Oropharyngeal dysphagia is most commonly diagnosed and managed by speech pathologists (SPs) (also known as speech and language therapists or speech language pathologists) working in a multidisciplinary team of health professionals including occupational therapists, physiotherapists, nurses, radiologists, gastroenterologists, and ear, nose and throat specialists.

Dysphagia is common in children who acquire their brain impairment early (for example, cerebral palsy) (Reilly 1996; Calis 2008) or later in life (for example, traumatic brain injury, stroke, encephalitis, brain tumour) (Cornwell 2003; Morgan 2010a). Another group of children experience dysphagia associated with genetic syndromes such as Down syndrome (Faulks 2007) or Rett syndrome (Morton 1997) or neurological degeneration (for example, muscular dystrophy) (Philpot 1999). Few rigorous epidemiological reports of dysphagia prevalence are available for populations of children with neurological impairment, with the exception of cerebral palsy and traumatic brain injury. For example, up to 99% of children with severe generalised cerebral palsy are reported to have dysphagia (Calis 2008), and between 68% and 72% of children with severe traumatic brain injury present with dysphagia during the acute phase of care (Morgan 2003; Morgan 2010a). An association is reported between neurological severity and dysphagia prevalence, with more severely affected children increasingly presenting with dysphagia (Morgan 2010b).

Diagnosis of oropharyngeal dysphagia

Dysphagia in childhood associated with neurological impairment is complex, with many interrelated factors contributing to its severity and nature of presentation.

A thorough diagnosis of oropharyngeal dysphagia in children with neurological disorder typically involves both a clinical swallowing evaluation (CSE), followed by the most appropriate instrumental assessment (Arvedson 2008). The CSE and instrumental examination are complementary procedures.

In brief, the CSE involves taking a detailed case history, observing the general presentation and cognitive-behavioral state of the patient, examining oromotor, laryngeal, and respiratory status, and determining aspiration risk during trials of foods and fluids. In childhood, a CSE must evaluate a child's feeding and swallowing function in the context of the skills expected during their particular transitional stage of feeding or developmental level. For example, children from birth to six months are predominantly breast or bottle fed, whereas children from six to 18 months are moving toward independent feeding where they are learning to drink from an open cup, to manipulate a spoon, and are moving towards handling increasingly varied textures. These developmental or transitional stages of feeding have important implications for the type of treatment approach used and its success. It is also important to note that the CSE does not allow objective diagnosis of impairment(s) in the pharyngeal phase of swallowing. Objective measurement requires instrumental diagnostic techniques that provide information on the anatomy and physiology of the swallowing process and in particular the functioning of the pharyngeal phase, including being able to determine the presence of prandial aspiration (aspiration of food or fluid into the trachea and lungs). These predominantly include the videofluoroscopic swallow examination (VFSE) and the endoscopic swallowing examination (ESE) (Arvedson 2008).

Consequences of dysphagia

The impact and human consequences of oropharyngeal dysphagia are widespread. The direct impacts of oropharyngeal dysphagia include physiological limitations to the oral phase (for example, poor lip closure, and poor oral transit due to reduced mobility of the tongue for propelling food posteriorly into the oropharynx to trigger a swallow) and pharyngeal phase of swallowing (for example, inadequate closure or paralysis of the vocal folds, resulting in aspiration of food and fluid into the trachea; inadequate pharyngeal peristalsis, resulting in excessive pooling of food or fluid in the valleculae or pyriform sinuses). In turn, this oropharyngeal impairment may disrupt the ingestion of food or fluid to result in two further significant consequences, namely *nutritional deficiencies* or *respiratory compromise*, both of which are potentially life threatening.

Nutritional deficiencies

Marked oropharyngeal dysphagia places children at risk of reduced energy and nutrient intake, and poor growth (Thommessan 1991; Stallings 1993; Arrowsmith 2006) leading to failure to thrive, or if left untreated, malnutrition. Prolonged poor energy and nutrient intake may have wide-ranging effects beyond physical growth, with potential impacts on psychomotor development and even neurodevelopment. There are also recognised effects of reduced nutrient intake due to dysphagia on the immune, skeletal, and

cardiovascular systems (Rosenbloom 1996). Micronutrient deficiencies have been reported in children with cerebral palsy (Patrick 1990), and specific deficits such as iron deficiency have been reported in children with neurodisability where their diets are limited to specific food sources that may be easier to ingest, but reduced in variety of nutrients (Rosenbloom 1996). Children with oropharyngeal dysphagia are unable to consume sufficient energy and nutrients and require supplemental non-oral feeding options, such as nasogastric tube feeding or, in severe cases, may require a gastrostomy.

Respiratory compromise

Oropharyngeal dysphagia puts a child at risk of prandial aspiration (where food or fluid is misdirected from the typical path from pharynx to oesophagus and rather enters the trachea and lungs), as well as choking and increased work of breathing during feeding. Respiratory complications, such as chest infection or pneumonia, may subsequently arise from oropharyngeal dysphagia due to the presence of aspiration (Loughlin 1989; Arvedson 1994). Children may be required to modify their diet in an effort to compensate for their feeding difficulties and avoid aspiration. In severe cases of aspiration (i.e., where children develop respiratory compromise such as chest infections or pneumonia associated with prandial aspiration), children may be required to use non-oral methods of feeding such as nasogastric tube feeding or gastrostomy.

Beyond the direct medical impacts of oropharyngeal dysphagia, there are other significant life impacts of the disorder. Dysphagia has impacts on a child's ability to participate in daily food-related activities. For example, in the case of a 15-year-old girl who returned to school with persistent dysphagia and risk of aspiration one year following a traumatic brain injury, the adolescent remained socially isolated from peers during her lunch break because she had to receive non-oral feeds (via gastrostomy) with the school nurse (Morgan 2004). Further social impacts can be seen in relation to mealtime interactions for children with dysphagia and their families. A recent study reported on the characteristics of mealtime communication between 20 mothers and their children with cerebral palsy (Veness 2008). In contrast to the positive communication behaviours typically seen for children without feeding impairments, mothers of children with dysphagia and cerebral palsy were found to dominate the mealtime interactions and used more directive communicative functions than their children. While mealtimes are typically an enjoyable time for socialisation within the family unit, they are often a stressful occasion for the child and family affected by dysphagia.

Description of the intervention

The focus of this review is to examine the efficacy of interventions targeting oropharyngeal dysphagia in children with neurological impairment. We will examine oropharyngeal dysphagia treatment in any setting at any frequency or duration. The comparison will typically be standard treatment. Standard treatment is the 'normal' care given to those children with oropharyngeal dysphagia. The nature of available interventions can be conceptualised as direct and indirect interventions. By targeting treatments at the impairment level, it is anticipated that participants will also experience associated improvements in activity and participation (WHO 2001) aspects associated with swallowing or oropharyngeal feeding success.

Direct interventions

Direct interventions use food or fluid during swallowing tasks to target the physiological limitations or impairments (WHO 2001) associated with oropharyngeal dysphagia across the oral (for example, poor lip closure or reduced mobility of the tongue for propelling food posteriorly into the oropharynx to trigger a swallow) and the pharyngeal phase of swallowing (for example, inadequate closure or paralysis of the vocal folds, resulting in aspiration of food and fluid into the trachea or inadequate pharyngeal peristalsis resulting in excessive pooling of food or fluid in the valleculae or pyriform sinuses). A range of impairment-level direct intervention methods are available, and we have provided examples of each below.

- Motor with swallow: for example, specific movement-based techniques such as the supraglottic swallow (for example, Logemann 1991; Logemann 1993; Ohmae 1996) or Mendelsohn manoeuvre (for example, Cook 1989; Huckabee 1999).
- Sensory with swallow: for example, altering bolus taste or flavour to make it sour or sweet to increase sensory input, increasing or decreasing the temperature of a food or fluid to increase sensory input and improve swallow physiology (Lazarus 1993; Logemann 1995).

Indirect interventions

- Motor without swallow: for example, exercises to increase oral motor function such as using the *Iowa Oral Pressure Instrument* to increase tongue strength and function with eventual improvements seen in swallow physiology (for example, Robbins 2007).
- Sensory without swallow: for example, techniques of applying thermal tactile stimulation such as icing the faucial arches in an attempt to increase sensation to this region with eventual improvements seen in swallow physiology (Lazzara 1986).
- Pharmacological/surgical: for example, interventions such as intrathecal baclofen or botox to increase or decrease tone to enable more functional oropharyngeal swallowing physiology.

Compensatory techniques

Compensatory techniques are based around improving activity limitations and participation restrictions, or removing environmental barriers to enhance oropharyngeal feeding success (WHO 2001). Examples of compensatory techniques include the following.

- Postural modifications (for example, altering the child's seating position to facilitate optimal trunk and body stability to effect improvement of function and control of the oropharyngeal musculature and hence improve swallow physiology) (Larnert 1995).
- Products and technology: altering feeding utensils or seating systems to facilitate swallowing function.
- Natural environment: experimentally altering the level of temperature, light or noise in the feeding environment to facilitate swallowing.
- Support networks: altering the level of external support required by children to facilitate swallowing.

How the intervention might work

See above.

Why it is important to do this review

As outlined earlier, oropharyngeal dysphagia may have deleterious impacts on health and quality of life for children with neurological impairment. Despite the high rate of dysphagia prevalence in children with marked neurological impairment, no study to date has systematically examined the literature in this field to summarise and report on the high quality evidence available. Speech pathologists most commonly manage children with dysphagia; however, the lack of evidence in this field has broader impacts across a range of health professionals, from medical officers to physiotherapists and dietitians. Further, the prevalence of dysphagia and lack of consensus on optimal interventions result in deleterious health economic impacts and places pressure on resources. It is, therefore, timely to undertake the present Cochrane review to systematically examine current literature and to encourage funding bodies and clinical researchers to address this striking evidence gap.

OBJECTIVES

To assess the efficacy of intervention targeting oropharyngeal dysphagia in children with neurological impairment.

METHODS

Criteria for considering studies for this review

Types of studies

Randomised controlled trials and quasi-randomised controlled trials (for example, where participants are allocated by date or birth, day of the week or alternate allocation).

Types of participants

Children aged 18 years or under, presenting with dysphagia of acquired (for example, following traumatic brain injury or stroke), developmental (for example, cerebral palsy), or degenerative neurological origin, including genetic syndromes (for example, Down syndrome, CHARGE syndrome, Rett syndrome), diagnosed by a health professional. We will include children with early structural deficits, including oesophageal atresia or tracheoesophageal fistulas, provided they have a coinciding oropharyngeal dysphagia. We chose the age limit because 18 years denotes the upper age limit of paediatric care for the majority of healthcare providers internationally, and as such it was anticipated that most paediatric-focused studies would include children of up to 18 years.

Types of interventions

We will consider direct and indirect interventions for oropharyngeal dysphagia that target the level of impairment, activity, and participation, or environmental factors (WHO 2001) (see Description of the intervention for exemplar treatment approaches under each of these domains).

Types of outcome measures

Primary outcomes

Outcomes will include the following.

1. Measurement of the physiological function of the oropharyngeal mechanism for swallowing, including:
    - effectiveness of lip seal;
    - tongue movement;
    - jaw rhythmicity;
    - triggering of swallow;
    - adequacy of laryngeal closure, etc.
2. We will measure these outcomes based on CSE, ESE or VFSE.
3. Presence of chest infection and pneumonia (possible indicators of effectiveness of laryngeal closure/airway protection during swallowing).
4. Diet consistency a patient is managing (a possible indicator of oral and pharyngeal skills, i.e. whether the patient can manage a developmentally appropriate oral diet, or if the texture/consistency of foods and fluids must be modified.

Secondary outcomes

Further outcomes will include the following.

1. Changes in growth: weight and height percentiles; growth velocity.
2. Level of reliance on supplementary feeding (for example, nasogastric or gastrostomy tube feeding).
3. Child's level of participation in mealtime routine with family, peers, or strangers.
4. Level of parent or carer stress association with feeding.

We will measure primary and secondary outcomes using medical chart review, questionnaires, rating scales, checklists or interviews by a relevant caregiver or health professional, including parent, carer, speech pathologist, medical officer, or teacher. Due to likely variance in quality of reporting, we will consider all measures but will discuss evidence of their reliability and validity. If studies have retrospectively used medical records to determine outcomes, we will also consider these studies individually.

We will group outcome time points for primary and secondary outcome measures as follows: immediately post-intervention, up to six months post-intervention, and more than six months post-intervention. It is difficult to anticipate the length of time to follow up post-intervention across studies and hence we will alter time points accordingly to best represent follow-up periods across studies.

Search methods for identification of studies

Electronic searches

We will search the following electronic databases:

Cochrane Central Register of Controlled Trials (CENTRAL), part of the Cochrane Library

MEDLINE

EMBASE

CINAHL

ERIC

PsycINFO

Science Citation Index

Social Science Citation Index

Cochrane Database of Systematic Reviews, part of the Cochrane Library

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

The search strategy will employ sensitivity rather than specificity to avoid missing any potential studies. We will use the following search strategy to search MEDLINE:

1 Deglutition Disorders/
2 (deglut$ adj5 (abnormal$ or disorder$ or dysfunc$ or impair$)).tw.
3 (swallow$ adj5 (abnormal$ or difficult$ or disorder$ or dysfunc$ or function$ or impair$)).tw.
4 ((oropharynx$ or trachea$ or lung$ or pulmon$) adj5 aspirat$).tw.
5 nasal regurgit$.tw.
6 or/1-5
7 oropharyn$.tw.
8 Oropharynx/
9 7 or 8
10 (dysphag$ or disorder$ or dysfunc$ or impair$).tw.
11 9 and 10
12 (pharyng$ adj5 (dysphag$ or dysfunct$ or disorder$ or impair$)).tw.
13 6 or 11 or 12
14 exp Infant/
15 exp Child/
16 (baby or babies or newborn$ or neonat$ or toddler$ or child$ or preschool$ or pre-school$ or schoolchild$ or child$ or adolescen$ or teen$ or juvenil$ or young people or young person$).tw.
17 Adolescent/
18 or/14-17
19 randomized controlled trial.pt.
20 controlled clinical trial.pt.
21 randomi#ed.ab.
22 placebo$.ab.
23 drug therapy.fs.
24 randomly.ab.
25 trial.ab.
26 groups.ab.
27 or/19-26
28 exp animals/ not humans.sh.
29 27 not 28
30 13 and 18 and 29

We will modify search terms as necessary when searching other databases. No date or language limits will be applied.

Searching other resources

Grey literature

We will identify unpublished and ongoing trials by searching Current Controlled Trials (ISRCTN Register), ClinicalTrials.gov and WHO ICTRP. In addition we will search for dissertations and theses using the following open access portals: Networked Digital Library of Theses and Dissertations, Australasian Digital Theses Program and DART-Europe E-theses Portal

Handsearching

We will handsearch the following journals for relevant trials:

- Developmental Medicine and Child Neurology
- Dysphagia
- Journal of Speech, Language and Hearing Research

We will search the reference lists of studies included in this review and relevant papers to identify additional studies in the published or unpublished literature.

Data collection and analysis

Selection of studies

Three review authors (AM, PD, and EW) will independently screen titles and abstracts for inclusion. In cases of uncertainly over whether an abstract meets the inclusion criterion by the review authors, we will obtain the full text article. Three authors (AM, PD, and EW) will then independently evaluate each paper for inclusion. In the event of disagreement over inclusion of a paper, we will form a consensus reassessing the inclusion criterion together. We will seek additional information from the authors of the studies where required to resolve questions about study methodology. We will record reasons for excluding studies. No review author will be blind to the authors, institutions, or the journals of publication of the articles.

Data extraction and management

Three review authors (AM, PD, and EW) will independently extract data for each study using a data extraction form to collect information about the population (including aetiology, severity of dysphagia, comorbid conditions), the intervention (including the length and frequency of intervention, professions involved), randomisation methods, blinding, sample size, outcome measures, follow-up duration, setting (for example, community clinic, school, hospital, home), attrition and handling of missing data, and methods of analysis. We will resolve disagreements through consensus discussion and reassessing the inclusion criteria together.

Assessment of risk of bias in included studies

Three review authors (AM, PD, and EW) will independently assess risk of bias in the studies by using The Cochrane Collaboration's tool for assessing risk of bias (Higgins 2008). We will resolve any disagreements by discussion until we reach consensus. We will use the tool to assess the following domains: sequence generation, allocation concealment, blinding, incomplete outcome data, selective outcome reporting, and other sources of bias (for example ceasing the trial early, changing methods during the trial, etc.).
We will present the quality of the trials in a 'Risk of bias' table where, for each question-based entry, we will make the judgement 'low risk', 'high risk' or 'unclear risk' of bias, followed by a text box providing details on the available information that lead to each judgement. We will assess the following sources of bias.

Random sequence generation

We will judge randomisation as follows.
'Low risk' when participants were allocated to treatment conditions using randomisation such as computer-generated random numbers, a random numbers table, or coin-tossing.
'Unclear risk' when randomisation method was not clearly stated or unknown.
'High risk' when randomisation did not use any of the above methods.

Allocation concealment

We will judge allocation concealment as follows.
'Low risk' when participants and researchers were unaware of participants' future allocation to treatment condition until after decisions about eligibility were made and informed consent was obtained.
'Unclear risk' when allocation concealment was not clearly stated or unknown.
'High risk' when allocation was not concealed from either participants before informed consent or from researchers before decisions about inclusion were made or allocation concealment was not used.

Blinding of participants, personnel, and outcome assessors

We will determine quality of participant, personnel, and outcome assessor blinding by whether knowledge of the allocated interventions was adequately concealed from these people during the study, by using the following judgements.
'Low risk' when participants, personnel and outcome assessors were blind to the treatment conditions and it was unlikely that the blinding could have been broken; where either participants or some key study personal were not blinded but outcome assessment was blinded and the non-blinding of others was unlikely to introduce bias.
'Unclear risk' when blinding of assessors was not reported and information was not available from researchers.
'High risk' when no blinding or incomplete blinding occurred and the outcome or outcome measurement was likely to be influenced by lack of blinding, or where blinding was attempted but could have been broken.

Incomplete outcome data

Assessment will take into account whether incomplete outcome data were adequately addressed by the researchers. We will contact corresponding authors of included studies where necessary, to provide any data that has not been reported (for example, group means and standard deviations (SDs), details of those who do not complete the trial, and details of interventions received by the control group). We will contact other authors for this data if the corresponding author fails to respond. If a study reports outcomes only

for participants completing the trial or only for participants who followed the protocol, we will contact the authors and ask them to provide additional information to permit analyses accordingly. We will describe missing data and dropouts/attrition for each included study in a 'Risk of bias' table and interpret what effect the missing data may have had on the results and conclusions of the review. We will conduct sensitivity assessment of any primary meta-analyses to missing data using the methodology outlined by Higgins 2008.

We will assess the adequacy of the way trials dealt with missing data using the following judgements.

'Low risk' when there were no missing outcome data, or reasons for missing outcome data were unlikely to be related to true outcome, or where missing outcome data are balanced in numbers across groups with similar reasons for missing data across groups, or where missing data have been imputed using appropriate methods.

'Unclear risk' when information about missing data was not available and cannot be acquired by contacting the researchers of the study.

'High risk' when the reason for missing outcome data is likely to be related to the true outcome, with either imbalance in numbers or reasons for missing data across intervention groups.

Selective reporting

We will assess the possibility of selective outcome reporting by review authors' judgement on whether reports of the study are free of the suggestion of selective outcome reporting; for example, whether it is clear that other data were collected and not reported.

Other bias

Assessment will determine whether any other bias is present in the trial, such as stopping the trial early, changing methods during the trial, or other anomalies.

Measures of treatment effect

Dichotomous data

Where dichotomous data are present, we will calculate a risk ratio (RR) with a 95% confidence interval (CI) for each outcome in each trial (Higgins 2008).

Continuous data

We will analyse continuous data when means and standard deviations are presented in the study papers, are made available by the authors of the trials, or are calculable from the available data. Where outcomes are measured using the same scale, we will calculate a mean difference (MD) to determine the differences in mean scores between groups. Where similar outcomes are measured using different scales, we will calculate a standardised mean difference (SMD) using Hedges *g*.

Time-to-event data

We will present the treatment effects of time-to-event data or survival data (for example child maltreatment incidence data) as a hazard ratio with 95% confidence intervals.

Unit of analysis issues

Cluster-randomised trials

It is possible that participants will be randomised to groups in clusters (for example, when participants are randomised by treatment locality or clinic). For trials that use clustered randomisation, we will present results with proper controls for clustering (robust standard errors or hierarchical linear models). If appropriate controls are not used and it is impossible to obtain the full set of individual participant data, we will control the data for clustering using the procedures outlined in Higgins 2008. That is, when outcome measures are dichotomous, the number of events and number of participants per trial arm will be divided by the design effect (1 + (1 - m) * r), where m is the average cluster size and r is the intra-cluster correlation coefficient (ICC). When outcome measures are continuous, we will divide the number of participants per trial arm by the design effect, while leaving the mean values unchanged. To determine the ICC, the review authors will use estimates in the primary trials on a study-by-study basis. However, where these values are not reported, the review authors will use external estimates of the ICC that are appropriate to each trial context and average cluster size by contacting the trialists and if they are not available, the reviewers will seek statistical assistance from the Cochrane Methods Group (Higgins 2008).

Multiple time points

When results are measured at multiple time points, we will analyse each outcome at each point in a separate meta-analysis with other comparable studies taking measures at a similar time point post-intervention, as described in the outcomes i.e. immediately post-intervention, up to six months post-intervention, more than six months post-intervention. If this is not possible we will define time frames to reflect short-term (up to two months), medium-term (two to six months) and long-term follow-up (more than six months).

Studies with multiple treatment groups

For trials where there are multiple treatment groups, we will not analyse data from the same group twice. We will select the treatment condition for meta-analysis according to which ones best match the inclusion criteria. The comparison condition will be treatment-as-usual or the least active treatment offered.

Dealing with missing data

We will assess missing data and dropouts in the included studies. We will investigate and report reasons, numbers, and characteristics of dropouts. We will attempt to contact the authors when further information or data are necessary. Any meta-analyses will use data from all original participants when possible, and will report when that is not the case. For studies in which the missing data are not available, we will use a sensitivity analysis to assess potential bias in the analysis and discuss the extent to which the results might be biased by missing data.

Assessment of heterogeneity

We will examine heterogeneity among included studies through the use of the Chi^2 test, where a low P value indicates heterogeneity of treatment effects. We will use the I^2 statistic (Higgins 2008) to determine the percentage of variability that is due to heterogeneity rather than sampling error or chance. We will discuss possible reasons for heterogeneity and conduct sensitivity analyses accordingly, where data permit. We may also use subgroup analyses to investigate this further, as described below.

Assessment of reporting biases

We will attempt to assess the possibility of selective outcome reporting by investigators; specifically, by including the review authors' judgement of whether reports of the study are free of the suggestion of selective outcome reporting.

Data synthesis

Where the interventions are similar in 1) type of intervention, 2) type of participants, and 3) intensity, frequency and duration of the intervention, we plan to synthesise results in a meta-analysis. We will use both a fixed-effect and a random-effects model and compare to assess the impact of statistical heterogeneity. Unless the model is contraindicated (for example, if there is funnel plot asymmetry), we plan to present the results from the random-effects model. In the presence of severe funnel plot asymmetry, we will present both fixed-effect and random-effects analyses, under the assumption that asymmetry suggests that neither model is appropriate. If both indicate a presence (or absence) of effect we will be reassured; if they do not agree we will report this. We will calculate all overall effects using inverse variance methods. If some primary studies report an outcome as a dichotomous measure and others use a continuous measure of the same construct, we will convert results for the former from an odds ratio to a SMD, provided that we can assume the underlying continuous measure has approximately a normal or logistic distribution (otherwise we will carry out two separate analyses).

Subgroup analysis and investigation of heterogeneity

We may conduct further investigation of the causes of heterogeneity using subgroup analyses. We will consider developmental levels for all children. For children born preterm, we will use the corrected age to two years. We will stratify children by dependent age at onset of treatment, and according to neurological group status, as follows.

- Age at onset of treatment, i.e. feeding for babies and infants is different to children who are able to self-feed, etc. We will group children into transitional stages of feeding (for example, less than six months: breast/bottle feeding; six to 18 months: moving towards independent feeding, cup drinking, eating textures; between two and five years when establishing oral motor skills; six to 12 years as the child begins to refine their oral motor skills; children who are 13 to 16 years).
- Neurological group status, i.e. genetic syndrome; acquired neurological lesion such as stroke or traumatic brain injury; developmental or early acquired brain lesion such as cerebral palsy, or a degenerative neurological condition such as myotonic dystrophy.

Sensitivity analysis

If the methodology or analyses in the trials might conceivably have affected the robustness of the results of the review, we will conduct sensitivity analyses by removing studies with particular characteristics and reanalysing the remaining studies to determine whether the relevant factors affect the results. We will restrict analyses to studies judged to be at low risk of bias. Specifically, we will restrict the analysis to: (a) studies with low risk of selection bias (for example, associated with sequence generation or allocation concealment); (b) studies with low risk of performance bias (for example, associated with issues of blinding); (c) studies with low risk of attrition bias (for example, associated with completeness of data). We will also assess the sensitivity of findings to any imputed data.

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* *Indicates the major publication for the study*

HISTORY

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CONTRIBUTIONS OF AUTHORS

All authors contributed to the development of this protocol. AM and EW drafted the original version of the protocol, with input from PD. AM and EW devised the original search strategy with support from Margaret Anderson. AM, PD, and EW will screen the abstracts and titles and retrieve potentially eligible papers. AM, PD, and EW will review the papers and make decisions about eligibility. AM and PD will extract data. AM will draft the full review with regular input from PD and EW at every stage.

DECLARATIONS OF INTEREST

- Elizabeth C Ward - none known.
- Angela T Morgan - none known.
- Pamela Dodrill - none known.

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