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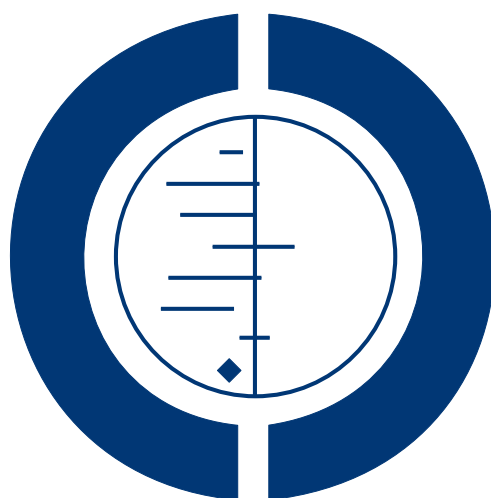


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# Non-speech oral motor treatment for developmental speech sound disorders in children (Protocol)

Lee ASY, Gibbon FE



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[Intervention Protocol]

# Non-speech oral motor treatment for developmental speech sound disorders in children

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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 non-speech oral motor treatment (NSOMT) for treating developmental speech sound disorders.

## BACKGROUND

### Description of the condition

Developmental speech sound disorder is a generic term that refers to clinically significant problems in producing the speech sounds of the language in children. This can be a secondary impact of a structural deficit of the articulators (for example, cleft palate), a sensory or motor disorder (for example, hearing impairment) or a neuromuscular disorder (for example, dysarthria, apraxia of speech). Alternatively, it may be a primary disorder where the cause is unknown (Ruscello 2008a; Flipsen 2009; Shriberg 2010). Speech problems can have damaging effect on many aspects of life. For example, they could hamper daily communication, causing difficulties in socialisation. It has been reported that about 50% to 70% of children with speech sound disorder show general difficulty with this through to secondary education (see Williams 2010). This could impair self-esteem, which in turn could affect quality of life.

It has been estimated that around 7.5% of children between the age of three and 11 years show clinically significant speech sound disorders (Shriberg 1994). Children with developmental speech sound disorders account for a large proportion of speech and language therapists' (SLTs) caseloads, nearly half of a typical caseload for the clinicians in the United Kingdom and Australia (see Joffe 2008). In the United States, it has been estimated that about 80% of children with speech sound disorders require treatment services and about 92% of school-based speech-language pathologists provide treatment services to children with speech sound disorders (see Ruscello 2008a).

### Description of the intervention

#### Phonetic and phonemic treatments

Different treatment approaches for managing speech sound disorders have been developed and they can be categorised broadly into phonetic (or sensory motor-based) treatments, phonemic (or conceptual-based) treatments, and hybrid treatments that incorporate both phonetic and phonemic components (Ruscello 2008b). Phonetic treatments focus on improving the accuracy of articulatory movements for the speech sounds through different levels of practice, from an isolated sound level to attain correct target sound production to nonsense syllables, words, phrases, and finally conversational speech (Ruscello 2008a; Ruscello 2008b). For phonemic treatments, the aim is to restructure or develop the children's phonological knowledge through different types of contrastive practice (for example, minimal pairs) or metalinguistic awareness tasks (for example, metaphon), or both (Ruscello 2008a; Ruscello 2008b). Hybrid treatments are a combination of phonetic and phonemic treatments in which both phonetic practice

and phonemic contrast are employed in the treatment (Ruscello 2008a).

The clinical decision of treatment approach is based mainly on the cause of the speech disorder. For example, phonetic approaches, which focus on developing the client's motor skills, are usually applied to those who have knowledge of phonological rules of the language but are unable to produce certain speech sounds correctly. Speech therapy using phonetic, phonemic, or a combination of these approaches is regarded as standard speech intervention.

#### Non-speech oral motor exercises

One alternate treatment approach for managing developmental speech sound disorder is non-speech oral motor treatments or exercises (NSOMTs). An operational definition of NSOMTs is provided by the American Speech-Language-Hearing Association's (ASHA) National Center for Evidence-Based Practice in Communication Disorders. They are "non-speech activities that involve sensory stimulation to or actions of the lips, jaw, tongue, soft palate, larynx, and respiratory muscles that are intended to influence the physiological underpinnings of the oropharyngeal mechanism to improve its function. They may include activities described as active muscle exercise, muscle stretching, passive exercise, or sensory stimulation" (McCauley 2009, p 344).

NSOMTs are different from the phonetic and phonemic treatments because they do not involve practice of speech sound articulation and auditory discrimination of the error sound and the target sound. Instead, they target non-speech motor movements and oral postures with the aim of developing motor skills for correct speech sound production (Ruscello 2008a). For example, by doing exercises with the lips in non-speech activities, such as blowing horns, straws, and bubbles, the motor skills developed could be transitioned to the production of bilabial speech sounds (for example, /p/ and /b/) (Rosenfeld-Johnson 2001). See the paper by Ruscello 2008b for details of the different types of NSOMTs.

NSOMTs are used in a variety of ways and for different client groups. A recent survey conducted in the United States revealed that most clinicians, 68% of 537 respondents, use NSOMTs as a 'warm-up' followed by speech intervention (Lof 2008). About 25% used NSOMTs in conjunction with speech intervention and 7% of the clinicians used NSOMTs exclusively to target speech productions (Lof 2008). The survey also showed that the clinicians often use NSOMTs with children presenting with motor speech disorders, structural anomalies (for example, cleft palate), and Down syndrome (Lof 2008). They also use NSOMTs, but less frequently, with children who are identified as late talkers, and those with phonological disorders, hearing impairment, and speech sound disorders with unknown origin (Lof 2008). A survey conducted in Canada showed slightly different findings. The clinicians there are most likely to use NSOMTs with children exhibiting phonological disorders, apraxia of speech, dysarthria, Down syndrome, and cerebral palsy (Hodge 2005).

## How the intervention might work

The use of NSOMTs for treating speech sound disorders in children is motivated by several assumptions (Bunton 2008; Ruscello 2008b; Clark 2010). First, it is assumed that a common set of control principles, such as force and timing, are used for controlling the same structure for conducting different motor activities (Bunton 2008; Ruscello 2008b). Hence, for example, the movement characteristics and task demands for the production of bilabial speech sounds and those for blowing bubbles or kissing are presumably similar.

Second, the principles of motor learning suggest that breaking down the complex movements in speech production into subcomponents could facilitate learning by allowing the motor system to plan simpler movement patterns and gradually develop skilled control of more complex movement patterns (Bunton 2008; Clark 2010). For example, practising tongue tip-to-alveolar ridge movement gestures for helping the production of alveolar stops (for example, /t/ and /d/) (Lof 2009).

Third, it is assumed that speech production and non-speech activities share common neural anatomical representation in the human nervous system (Chapman Bahr 2001; Bunton 2008). On the basis of these assumptions, it is believed that the training effect from the practice of non-speech oral motor exercises could be transferred to speech production (Powell 2008).

In theory, NSOMTs are most likely to benefit children who have speech sound disorders due to sensorimotor impairments (Clark 2010), which means that causation may be an important determinant in the selection and use of NSOMTs.

## Why it is important to do this review

NSOMTs are used extensively by clinicians when treating developmental speech sound disorders. Recent surveys reported that between 71.5% and 85% of speech and language therapists in the United Kingdom and speech-language pathologists in the United States and Canada use NSOMTs (Hodge 2005; Joffe 2008; Lof 2008). There had not been a systematic review on this issue until the recent work by McCauley and colleagues (McCauley 2009); however this systematic review was not conducted according to the standards set by The Cochrane Collaboration. There are a number of limitations as the types of studies were not limited to randomised clinical trials (RCTs) and quasi-RCTs; only studies published in English were included for review; and the literature search was limited to databases that encompass peer-reviewed journals. There are Cochrane systematic reviews on the efficacy of speech intervention for speech problems related to childhood apraxia of speech (Morgan 2008a), dysarthria associated with acquired brain injury (Morgan 2008b), and primary speech and language delay or disorder (Law 2003). These reviews planned to compare NSOMTs to standard speech intervention and to evaluate the treatment efficacy on three levels of outcomes. However, whether factors such as

frequency of therapy sessions and the presence or absence of intellectual disability could affect the treatment efficacy of NSOMTs is uncertain. Although there could be some overlap with these systematic reviews, this review will cover a broader spectrum of developmental conditions and subsequently provide a more in-depth evaluation of the treatment efficacy of NSOMTs.

Given the high incidence of speech sound disorders and the abundance of commercial products and training workshops for NSOMTs (Kamhi 2008), there is a pressing need to examine the evidence regarding the efficacy of NSOMTs so that clinicians can make informed decisions in their treatment planning. Moreover, clients and their families also need to be made aware of the evidence relating to efficacy, or lack thereof, for treatments that involve NSOMTs.

## OBJECTIVES

To assess the efficacy of non-speech oral motor treatment (NSOMT) for treating developmental speech sound disorders.

## METHODS

### Criteria for considering studies for this review

#### Types of studies

All relevant randomised and quasi-randomised controlled trials (for example, studies in which participants were allocated to treatments by, for example, alternate allocation or allocation by date of birth). Trials using cross-over design will be excluded from the review as this is considered inappropriate for studying speech intervention.

#### Types of participants

Individuals aged three to 16 years with developmental speech sound disorders, as judged by speech and language therapist(s). Individuals with intellectual disability (for example, Down syndrome) and physical disability will not be excluded.

#### Types of interventions

Non-speech oral motor treatment versus treatment with placebo or control. We will also include NSMOTS as an adjunctive treatment if speech intervention with NSOMTs is compared with speech intervention alone. There will be no restriction on the frequency, intensity, and duration of intervention.

## Types of outcome measures

### Primary outcomes

1. Correct articulation of speech sounds targeted in the treatment, as measured by standardised tests (for example, Edinburgh Articulation Test (Anthony 1971)) or non-standardised articulation tests (for example, per cent correct speech sounds produced based on perceptual evaluation of articulation).
2. Speech intelligibility measured using a perceptual rating scale or percentage of words correctly transcribed by the investigator.
3. Speech physiology, as measured by instrumental techniques such as acoustic analysis (for example, the measure of formant frequencies for assessing vowel productions), kinematic analysis, and articulatory placement.
4. Adverse effect of an increase in articulation errors after treatment, which could be measured by the same standardised and non-standardised tests listed above. These articulation tests should be conducted by a speech and language therapist. We will include the first two primary outcomes, correct articulation of speech sounds targeted in the treatment and speech intelligibility, and adverse effects in the 'Summary of findings' tables.

### Secondary outcomes

1. Listener acceptability, speech naturalness or bizarreness (for example, judged by naive listener(s) using perceptual rating).
2. Self-perception of change in articulation or speech intelligibility (for example, using a rating scale).

### Time points for measuring the outcomes

- Immediately (within one month) after the end of the intervention.
- One to 12 months after the end of the intervention.
- One to two years after the end of the intervention.

## Search methods for identification of studies

### Electronic searches

We will search the following databases to identify relevant trials: Cochrane Central Register of Controlled Trials (CENTRAL) (*The Cochrane Library*), MEDLINE (or PubMed), EMBASE, ERIC, PsycINFO, CINAHL, ISI Web of Knowledge, LILACS, Academic Search Complete, The National Research Register Archive, UKCRN, ClinicalTrials.gov, metaRegister of Controlled Trials, WHO International Clinical Trials Registry Platform (ICTRP), and ProQuest Dissertations and Theses.

We will use the following search terms to search MEDLINE. We will modify terms and filters as appropriate for other databases. No language or date restriction will be applied to the searches. We will seek translation when necessary.

1. exp Speech Disorders/
2. dysarthri\$.tw.
3. (mute or mutism).tw.
4. ((speech or articulat\$ or phonetic\$ or phonological) adj5 (disorder\$ or delay\$ or impair\$ or problem\$)).tw.
5. or/1-4
6. oral motor.tw.
7. oromotor.tw.
8. oro-motor.tw.
9. (NSOM\$ or OME).tw.
10. Speech Disorders/rh, th [Rehabilitation, Therapy]
11. (non-speech or nonspeech\$).tw.
12. Speech Therapy/
13. or/6-12
14. randomized controlled trial.pt.
15. controlled clinical trial.pt.
16. randomi#ed.ab.
17. placebo\$.ab.
18. drug therapy.fs.
19. randomly.ab.
20. trial.ab.
21. groups.ab.
22. or/14-21
23. exp animals/ not humans.sh.
24. 22 not 23
25. 5 and 13 and 24

### Searching other resources

We will check reference lists of relevant journal papers, book chapters, and systematic reviews which are identified by the electronic searches. We will approach colleagues and researchers by email to identify other possible published and unpublished studies, such as technical or research reports, conference papers, and dissertations. We will use Google to find websites of relevant organisations, and will search these using appropriate search terms from the strategy above.

## Data collection and analysis

### Selection of studies

One review author (AL) will conduct the literature search. We will manage all references generated from the search strategy using a reference management programme (EndNote). The two review authors (AL and FG) will independently conduct an initial screening of titles and abstracts to eliminate any references that



are apparently irrelevant to the review (for example, single case studies). In cases where an abstract contains insufficient information for judging whether a study meets the inclusion criteria, we will obtain the full paper. AL and FG will then independently evaluate each paper against the inclusion criteria. In the event of disagreement over inclusion of a particular paper, AL and FG will seek to reach a consensus by assessing the paper together. We will report the disagreement, including the title(s) and the reason(s) for different judgements between the two review authors, and the consensus obtained after discussion.

### Data extraction and management

We will develop and pilot a data extraction form and subsequent versions of the form will include revision dates. AL and FG will independently extract the following information from each paper:

1. participants - number; age; sex; inclusion and exclusion criteria; severity level of developmental speech sound disorders; and other baseline characteristics reported (for example, hearing ability, intellectual disability, etc);
2. methods - speech assessment(s) and outcome measure(s) used and assessment results (for example, number and types of articulation errors).
3. interventions - type of interventions; number of therapy sessions given, duration of each therapy session, frequency of therapy and length of intervention; date and location; and whether compliance was evaluated;
4. intervention integrity - using the categories proposed by Dane and Schneider (Dane 1998), we will record the presence or absence of features of fidelity verification and promotion (for example, training manual developed for training intervention provider).

### Assessment of risk of bias in included studies

AL and FG will independently assess the risk of bias in each included study in the following six domains according to the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). The assessment will consist of two parts: (1) a succinct description, which will include verbatim quotes from the paper or correspondence with the trial author(s) or a comment from the review author about the procedures used to avoid bias, or both; and (2) an assessment of the risk of bias (by assigning a judgement of 'low risk', 'high risk', or 'unclear risk' of bias). We will not include studies that are judged as 'high risk' for each area of potential risk of bias in subsequent analyses. The judgement criteria for each domain are described below.

### Sequence generation

We will describe the method used to generate the allocation sequence using quotes wherever possible. We will add a comment, such as 'probably done' or 'probably not done', to supplement any

ambiguous quote. We will assign each included study to one of the following categories:

- 'low risk', which indicates an adequate method was used for randomisation (for example, computer generated or table of random numbers) or quasi-randomisation;
- 'high risk', which indicates that an inadequate method of randomisation was used (for example, case file number, date of birth, or alternate numbers);
- 'unclear risk', which indicates uncertainty about whether an appropriate method of randomisation was used.

### Allocation concealment

We will assign the included studies to one of the following quality criteria whereby:

- 'low risk' indicates adequate concealment of allocation (for example, pre-numbered or coded identical containers administered serially to participants);
- 'high risk' indicates that the allocation was not adequately concealed (for example, alternate assignment);
- 'unclear risk' indicates uncertainty about whether the allocation was adequately concealed (for example, the authors did not describe the allocation methods).

### Blinding

Blinding of participants and intervention providers (that is, the SLTs) is not possible but blinding of outcome assessor(s) and data analyst(s) from knowledge of which intervention a participant had received should be ensured. We will describe and evaluate the measures used to ensure blinding. We will grade this domain as 'low risk', 'high risk', and 'unclear risk'. Assessment will be made for each main outcome (for example, outcome measured at six months post-therapy, outcome measured at 12 months post-therapy).

### Incomplete outcome data

Incomplete outcome data refer to those that were due to attrition (dropout) during the study, or exclusions from the analysis. We will extract the number of and reason(s) for attrition or exclusions, and note whether attrition was analysed appropriately (for example, intention-to-treat analysis). We will grade this domain as 'low risk', 'high risk', and 'unclear risk' according to the criteria stated in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011).

### Selective reporting bias

This is also known as within-study publication bias. It may arise in several ways (Higgins 2011).

- Only some of the analysed outcomes were included in the study.

- Different time points at which the outcome was measured, or different instruments or assessors employed to measure the outcome at the same time point.
- Selective reporting of analysis using the same data.
- Selectively reporting the results of subscales of full measurement scale or a subset of events.
- Some outcomes were reported but with inadequate detail for the data to be included in a meta-analysis.

We will assign the included studies to one of the following quality criteria whereby:

- ‘low risk’ indicates that the studies have reported all pre-specified outcomes;
- ‘high risk’ indicates that any of the above-mentioned selective reporting is evident in the study; and
- ‘unclear risk’ indicates that it is uncertain whether selective reporting bias is avoided.

#### Other sources of bias

Other sources of bias may include baseline imbalance, early stopping, and co-intervention. This domain will be graded as ‘low risk’, ‘high risk’, and ‘unclear risk’.

#### Measures of treatment effect

##### Binary and categorical data

Binary data (for example, articulation improved versus no change) is likely. We will analyse the data by calculating the risk ratio.

##### Continuous data

Most data from the expected outcome measures, such as standardised articulation test results, articulation accuracy based on perceptual evaluation, judgement of speech intelligibility, and listener acceptability are likely to be continuous data. We will calculate the mean difference (MD, or the ‘difference in means’) if the outcome measurements in all studies are made on the same scale. Otherwise, we will use standardised mean differences (SMD) to combine studies that measured the same outcome using different methods.

##### Unit of analysis issues

##### Cluster-randomised trials

It is possible that we will include cluster-randomised trials in this review. In this case, appropriate statistical approaches should be used; for example, using a two-sample t-test to compare the means of the clusters in the intervention group to those in the control group at cluster level, or a mixed effects linear regression approach

at individual level (Donner 2000). We will contact the trial author(s) in case it is unclear if appropriate adjustments have been made (Donner 2000). If individual level data cannot be secured, we will control the data for the clustering effect using the procedures described in the *Cochrane Handbook for Systematic reviews of Interventions* (Higgins 2011). For dichotomous data, we will divide the number of participants and the number experiencing the event by the design effect,  $1 + (M-1) * ICC$ , where M is the average cluster size and ICC is the intra-cluster correlation coefficient. For continuous data, we will divide the number of participants by the design effect, with the means and standard deviations remaining unchanged. We will combine the results with those from individually randomised trials for meta-analysis using the generic inverse-variance method in RevMan, if the clinical heterogeneity between studies is small (Donner 2000; Higgins 2011).

##### Multi-arm studies

For studies with more than two intervention groups, that is, multi-arm studies, we will combine groups to create a single pair-wise comparison (Higgins 2011). We will combine all relevant experimental intervention groups to form a single group, and we will combine all relevant control groups and placebo groups to form a single control group. To avoid any confusion over the nature of each study, we will mention all intervention groups of a multi-arm study in the ‘Notes’ section of the table ‘Characteristics of included studies’. We will give detailed descriptions of the intervention groups relevant to the review in the ‘Interventions’ section of the table.

##### Cross-over trials

Cross-over trials are not appropriate for an intervention that can have a lasting effect (Higgins 2011). Therefore, this design is not suitable for studying speech intervention and we will not include studies that employed this design in this review.

##### Dealing with missing data

We will assess missing data and dropouts for each included study and report the reasons, numbers, and characteristics of dropouts. Whenever possible, we will contact the trial author(s) for supplying the missing data and any relevant information. If the missing data appear to be missing at random (for example, data lost due to computer problems), we will conduct analysis on the available data (Higgins 2011).

However, if the data are not missing at random, we will conduct the analysis by imputing the missing data with replacement values. For dichotomous data, we will use a sensitivity analysis based on consideration of ‘best-case’ and ‘worst-case’ scenarios to assess the extent to which the results of the review could be altered by the missing data (Gamble 2005). The ‘best-case’ scenario means that all participants with missing outcomes in the intervention group

had good outcomes (for example, improvement in articulation) and those with missing outcomes in the control group had poor outcomes (for example, no improvement in articulation); and the 'worst-case' scenario is the reverse.

For missing continuous data, we will conduct the analysis by imputing the missing data with replacement values (for example, last observation carried forward, mean of the treatment group) and treating these as if they were observed (Higgins 2011). We will address the potential impact of missing data on the findings of the review in the 'Discussion' section.

### Assessment of heterogeneity

Variability in the participants, interventions, and outcomes between the different included studies is known as clinical heterogeneity, and variability in the intervention effects being evaluated in the included studies is known as statistical heterogeneity, or simply as heterogeneity (Higgins 2011). Clinical heterogeneity will lead to statistical heterogeneity if the intervention effect is affected by factors such as patient characteristics (Higgins 2011). We will assess statistical heterogeneity by using the  $\text{Chi}^2$  test for heterogeneity, through visual inspection of forest plots, and by using the  $I^2$  statistic (Higgins 2002; Higgins 2003). The  $\text{Chi}^2$  test assesses whether observed differences in results are compatible with chance alone (Higgins 2011). However, it has low power if the meta-analysis includes only a small number of studies, or the studies included have small sample sizes. In this case, a P-value of 0.10 (rather than the conventional level of 0.05) will be used to determine statistical significance (Higgins 2011).  $I^2$  is a statistic for assessing the impact of inconsistency across studies on the meta-analysis. We will follow the rough guide to interpretation of the  $I^2$  statistic stated in the *Cochrane Handbook for Systematic Reviews of Interventions* (Higgins 2011). However, thresholds for the interpretation of the  $I^2$  statistic may be misleading. We will take into account other issues, such as the magnitude and direction of effects, and the strength of evidence for heterogeneity (for example, the P value from the  $\text{Chi}^2$  test), when determining the importance of the observed value of the  $I^2$  statistic (Higgins 2011).

### Assessment of reporting biases

Funnel plots (effect size against error) will be drawn if sufficient studies are found. An asymmetric funnel plot indicates a relationship between effect size and study size, which suggests the possibility of either publication bias or a systematic difference between smaller and larger studies. If a relationship is identified, the clinical diversity of the studies will also be examined (Egger 1997).

### Data synthesis

We will carry out meta-analysis using *Review Manager* 5.1 (RevMan), if there are sufficient data and where the interventions

are similar in terms of the characteristics of the participants, the types of NSOMTs used, the schedule (for example, frequency and duration) of the treatment, and outcome measures. We will apply both a fixed-effect and random-effects model and compare the results to assess the impact of statistical heterogeneity. We will present the results from the random-effects model unless contra-indicated (for example, if there is funnel plot asymmetry). In the case of serious funnel plot asymmetry, we will present both fixed-effect and random-effects model analyses, under the assumption that asymmetry suggests that neither model is appropriate. If the same outcome is presented as dichotomous data in some studies and as continuous data in other studies, we will convert odds ratios for the dichotomous data to standardised mean differences (SMD) if it can be assumed that the underlying continuous measurements follow a normal or logistic distribution. Otherwise, we will conduct separate analyses.

### Multiple time points

For studies where outcomes were measured at different time points, we will calculate the combined effect size across the different time points (Borenstein 2009).

### Subgroup analysis and investigation of heterogeneity

If there are sufficient homogenous studies, we will conduct subgroup analyses to assess the impact of the cause of speech sound disorders (for example, structural anomalies, neuromuscular impairment, unknown origin), intensity of therapy (to be determined by the frequency of therapy sessions), the presence or absence of intellectual disability, and the use of NSOMTs as an adjunctive to speech intervention.

### Sensitivity analysis

We will examine the impact of study quality on the robustness of conclusions by performing sensitivity analyses. Factors that are considered as important in judging study quality include randomisation, blinding to outcome assessment, and attrition (Juni 2001). We will include studies that we categorised as low or unclear risk of bias for these factors in the analysis.

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\* Indicates the major publication for the study

## HISTORY

Protocol first published: Issue 10, 2011

## CONTRIBUTIONS OF AUTHORS

AL and FG planned the review. AL wrote the protocol and developed the search strategies.

## DECLARATIONS OF INTEREST

- Alice S-Y Lee - none known.
- Fiona E Gibbon - none known.