

# Non-pharmacological treatments for stuttering in children and adults: a systematic review and evaluation of clinical effectiveness, and exploration of barriers to successful outcomes

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## Scientific summary

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# Scientific summary

## Background

Treatments for stuttering (which is more often known as stammering in the UK) have been available for children and adults since the 1950s. These treatments have encompassed diverse techniques from the use of carbon dioxide, or pharmacological interventions, to those that are non-pharmacological and behavioural or cognitive based. Although there has been a considerable growth in the range of interventions available for people who stutter, much of the review evidence to date has evaluated only behavioural programmes. There has been less examination of treatments that use outcome measures other than stuttering frequency.

The growing range of available treatment options for children and adults who stutter presents a challenge for clinicians, service managers and commissioners, who need to have access to the best available treatment evidence to guide them in providing the most appropriate interventions. Although a number of reviews of interventions for specific populations or a specific type of intervention have been carried out, a broad-based systematic review across all forms of intervention for adults and children was needed to provide evidence to underpin future guidelines, inform the implementation of effective treatments and identify future research priorities. The development of systematic review methods provides the opportunity for investigating not only the clinical effectiveness of interventions reported via a wider range of study designs, but also to use qualitative evidence to provide better understanding of why interventions may or may not lead to successful long-term outcomes.

## Aims and objectives

The aims of this study were to systematically identify, appraise and synthesise the international evidence on interventions to treat stuttering (and cluttering) in pre-school children, school-aged children, adolescents and adults; and to determine how applicable this evidence might be to the UK context including identifying patient and staff perceptions of potential obstacles to successful outcomes following intervention.

## Methods

A systematic review of the literature relating to the clinical effectiveness of interventions for stuttering and views and perceptions regarding interventions for stuttering was carried out. The population under consideration was children, adolescents and adults who have a stutter. Any intervention that was described as being a treatment for stuttering that is non-pharmacological and delivered in any setting, by any agent, was within the scope of the work. Studies reporting any outcome relating to an effect on stuttering or the emotional well-being of people who stutter were eligible for inclusion.

Comprehensive literature searches were undertaken in August 2013 to April 2014 to retrieve studies that met the review inclusion criteria. Databases searched included MEDLINE (via OvidSP); KysInfo (via OvidSP); EMBASE (via OvidSP); The Cochrane Library (Wiley), including the Cochrane Systematic Reviews Database; Applied Social Sciences Index and Abstracts (ProQuest); and Linguistics and Language Behavior Abstracts (ProQuest). Searches were not limited by language or location, but were restricted by date to studies published from 1990 onwards. Methods for identification of relevant studies included electronic database searching, reference list checking, citation searching and hand-searching of key journals.

Data were extracted by two reviewers using a data extraction form devised for the purpose. Extracted data were checked by the team and disagreement resolved by discussion. Appraisal of study quality was performed using tools based on established criteria for considering risk of bias, with a separate tool for the intervention studies and the qualitative papers.

Results are presented via narrative synthesis of the effectiveness studies, thematic synthesis of the qualitative data, and by a metasynthesis of the two review components in the form of a conceptual diagram, which illustrates elements of the pathway from interventions to long-term impact described in the literature. Meta-analysis of intervention clinical effectiveness across the body of literature was not possible owing to heterogeneity of intervention content and outcome measurement.

## Results

This wide-ranging review of the literature on interventions for people who stutter identified a sizeable body of work and included 137 papers in the evidence synthesis (111 papers contributed evidence to the review of effectiveness, 25 were qualitative studies, and one mixed-methods paper contributed to both reviews). The review identified seven typologies of intervention studies and found evidence of clinical effectiveness across the range of intervention types. Virtually all the work reported at least some positive outcome for most participants. There was evidence from all types of intervention that effects could be maintained following intervention (although this was weakest with regard to feedback and technology interventions). The review classified around one-third of the included work as providing stronger evidence that these health technologies are able to produce positive outcomes; however, around two-thirds of studies were considered to be at higher risk of bias.

However, the individual variability in response was notable, with little evidence that any intervention would be successful for all who received it. In the generally positive reporting of study findings, there was – in many cases – a sizeable number who did not achieve benefit and in the lower-quality studies, the potential for participants reported to differ from those not recruited and/or reported cannot be ruled out in the lower-quality studies. In relation to interventions for children who stutter, the natural recovery rate remains an issue for demonstrating levels of clinical effectiveness; however, research (although suggesting possible predictors) is unable to differentiate with absolute certainty those individuals who will spontaneously recover and those who will have long-term stuttering requiring intervention.

Variation in systems of measurement and variation in intervention contact hours has an adverse impact on the comparison of stuttering interventions with each other. There is little available research that compares the effectiveness of different interventions and, thus, a very limited pool of evidence for clinicians to draw on in selecting an optimal intervention, and also for people who stutter to use in order to make an informed choice. The qualitative literature suggested that important elements of successful interventions were attending to emotional and psychological needs; tailoring interventions to client needs; including maintenance sessions; therapists being client-centred; and having external support networks.

Currently, core outcomes for stuttering have not been established and studies that we identified used a range of outcomes including clinician-measured counts, independent listener counts and rating by the people who stutter. The challenge in establishing what a 'good outcome' following intervention should be is a key issue for the field. Although a sizeable body of studies included in this review reported effectiveness in terms of percentage reduction in dysfluency, it is debatable how significant a reduction of, for example, 2–3 syllables per 100 syllables might be for the everyday functioning of a person who stutters. Although there is some evidence of increasing involvement of people who stutter in the determination of outcomes, the field remains dominated by measures of overt stuttering behaviours, in particular the percentage of syllables that are stuttered. The qualitative literature highlighted the different views of people who stutter regarding their stutter and their differing needs at different stages of the life course, with reduction in overt stuttering being only one aspect. Further understanding regarding how and to what degree intervention outcomes relate to the everyday lives of people who stutter is needed. Few of the interventions considered any potential adverse impact.

This systematic review did not include consideration of the economic aspects of these health technologies. If questions regarding the cost-effectiveness of interventions for stuttering are to be investigated, further understanding of the short- and long-term outcomes is needed. The conceptual model that we developed which summarises the pathway from interventions to impacts highlights both the complexity of outcome measurement and the need for greater understanding regarding how and why these interventions may lead to positive impacts.

## Limitations

The review findings are based on data from a substantial number of published studies and considered both quantitative and qualitative evidence. We had hoped to include evidence from studies of professional views; however, we were unable to identify any qualitative papers exploring professional perceptions that met our inclusion criteria. The work included a range of study designs encompassing both controlled and non-comparator studies; however, the body of work reporting single cases and multiple case studies was excluded, together with surveys. Although case studies are able to contribute potentially useful data, their inherent propensity for bias, limited generalisability and the availability of a large volume of higher-quality designs underpinned our decision to exclude them from this review. The body of work that we included encompassed both studies that we categorised as being at higher risk of bias as well as those at lower risk. We considered whether or not to use quality criterion as a basis for rejection; however, this would have precluded analysis and reporting of a large quantity of literature and we intended to produce a comprehensive 'state of the art' review of the area. Therefore, in reporting of the results we have detailed and fully considered the quality of study design.

We had intended to carry out a meta-analysis of the clinical effectiveness data, but the heterogeneous nature of the literature and variability in outcome reporting meant that a narrative synthesis was most appropriate. In addition, the lack of mixed-method designs and qualitative papers that described specific interventions precluded our planned metasynthesis approach, which juxtaposes quantitative and qualitative results. Instead, we used the two sets of data to develop a conceptual model that sets out components of the pathway from interventions to impacts, and which we believe provides a useful tool to aid understanding of the review findings.

## Conclusions

The review indicates that a variety of interventions can produce positive outcomes for people who stutter. The evidence does not permit identification of programmes that are more effective versus those that are less effective, with all intervention types seeming able to lead to some benefit for some participants. The heterogeneity in outcomes measures and limited quality of the interventions meant that we were only able to compare intervention efficacy at a narrative level. We were unable to demonstrate any clear dose–response relationship, meaning that currently interventions with many hours of contact did not seem to offer substantially different outcomes to those with fewer, with variation in outcomes at the level of the individual rather than the intervention. The qualitative literature provides insight into factors that are perceived to facilitate successful outcomes, including ensuring that interventions encompass emotional/psychological/social aspects; incorporating 'real-world' elements; having follow-up sessions; and interacting with other people who stutter. This literature highlighted factors that may lead to variation in outcome relating to the individual, the intervention and interpersonal/social processes.

## Recommendations for research

1. The field has a large body of small-sample baseline follow-up investigations suggesting that alternative study designs are required in the future, such as research comparing interventions. Around two-thirds of the intervention studies were classified as being at potential higher risk of bias with more robust study designs needed.
2. There seems to be a research gap around aspects of process evaluation such as intervention fidelity, practitioner-specific effects, acceptability and feasibility. Little of the literature included consideration of resource and training implications of interventions, which is information that is needed in order to inform commissioning as well as clinical decisions.
3. Although the literature currently has a tendency for focusing on demonstrating that a particular intervention is effective, the evidence base suggests a need instead to explain how and why therapy works and, in particular, a need to further investigate individual variation in response. The use of more mixed-methods research could help to address these evidence gaps by exploring in-depth participant experiences and factors underpinning outcomes.
4. The measurement of outcomes in the field is a considerable obstacle to the evaluation of clinical effectiveness. Although different studies continue to use varied measures of stuttering, comparison between them remains challenging. Although measures of overt stuttering behaviours continue to dominate evaluation, the establishment of core outcomes (which are of importance and relevance to people who stutter) seems to be an urgent priority.
5. A gap in the qualitative literature concerns the views of children receiving therapy. Although the issues relating to young people taking part in research are not insubstantial, a reliance on retrospective recall of adults regarding their childhood means that views will inevitably be of historic approaches and potentially affected by later experiences.
6. Another recommendation for future studies concerns the recruitment of less heterogeneous participants. Although it is recognised that investigators have a limited pool to recruit from, many studies had variation in baseline characteristics of participants, which adds to the challenge of investigating why and for whom interventions are most successful.
7. An element described as facilitating successful outcomes for people who stutter was a client-centred approach and an individually tailored intervention. This is at odds with some of the programmes evaluated in the included literature, which offer a carefully structured and planned product. If 'real-world' interventions in clinical practice are bespoke and tailored for each individual client drawing on a variety of approaches and techniques, research should ensure that studies that are able to contribute evidence that is applicable to practice.
8. We were able to identify only one study that specifically reported participants who were cluttering. Research on interventions for this disorder seems to be very underdeveloped.
9. A further gap concerns the lack of qualitative studies regarding professional views and experiences of interventions.

## Study registration

This study is registered as PROSPERO CRD42013004861.

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