Systematic review of techniques to enhance peak cough flow and maintain vital capacity in neuromuscular disease: the case for mechanical insufflation-exsufflation

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CRD summary
This review assessed the most effective physiotherapy method for enhancing peak cough flow and maintaining vital capacity in patients with neuromuscular disease. The authors concluded that mechanical insufflation-exsufflation appears the most effective technique. However, this is based on four small relatively poor-quality studies and these findings need to be confirmed by further research.

Authors' objectives
To assess which is the most effective physiotherapy method for enhancing peak cough flow (PCF) and maintaining vital capacity in patients with neuromuscular disease (NMD).

Searching
MEDLINE (from 1996), CINAHL, AMED, DARE, the Cochrane Controlled Trials Register, ACP Journal Club, the Cochrane Database of Systematic Reviews and PEDro were searched from inception to 2003. The search terms were selected through networking with other colleagues via the interactive CSP website; the terms were reported. The authors also searched for articles published by known researchers in the topic area and checked the reference lists of retrieved articles for further studies. The searches were limited to controlled trials, systematic reviews and research articles published in the English language.

Study selection
Study designs of evaluations included in the review
Randomised controlled trials and controlled studies were eligible for inclusion in the review. Systematic reviews were also eligible.

Specific interventions included in the review
Studies that compared different physiotherapy methods aimed at improving PCF and maximum insufflation capacity (MIC) were eligible for inclusion. The methods assessed in the review included: MIC manoeuvre; MIC and manual assist; mechanical insufflation-exsufflation (MI-E); breath stacking techniques; manually assisted cough, mechanical insufflation and manual assistance; noninvasive ventilation (NIV)-assisted cough; and exsufflation alone. In one study physiotherapy methods were compared with each other and unassisted ventilation. In two studies assisted cough techniques were compared with each other and unassisted cough.

Participants included in the review
Studies that assessed patients with NMD were eligible for inclusion. Patients with NMD could either be studied as a whole population or as a subgroup. The patients assessed in the included studies were reported to have bulbar muscle weakness in one study and scoliosis (a subgroup) in another study; one study included both children and adults (age range: 10 to 56 years). In three studies patients acted as their own controls, but one study compared findings with a group of age-matched controls. A further study also included a control group of normal non-smokers without a history of respirator, neuromuscular or cardiovascular disease. This study also included patients with chronic obstructive pulmonary disease, but only patients with NMD were included in the review.

Outcomes assessed in the review
Studies that reported numerical values for clinical outcome measures of PCF and vital capacity (i.e. MIC) were eligible for inclusion in the review.

How were decisions on the relevance of primary studies made?
The authors did not state how the papers were selected for the review, or how many reviewers performed the selection.
Assessment of study quality
The validity of the studies was assessed using a published critical appraisal tool (Bury et al., see Other Publications of Related Interest).

Three reviewers assessed the validity of the included studies. Agreement was reached between all reviewers as to the quality of the studies.

Data extraction
The authors did not state how the data were extracted for the review, or how many reviewers performed the data extraction. Details of the study design, patient eligibility, intervention, outcome data and overall findings were abstracted and tabulated.

Methods of synthesis
How were the studies combined?
Differences between the studies in terms of the study design, interventions, populations and methods of outcome measurement led to the use of a qualitative synthesis. The findings were summarised in a data table and in a narrative, where studies were discussed according to outcome (i.e. PCF recordings, PCF recordings after combination treatments, and the effect of increasing vital capacity on PCF); a short summary of the limitations of the included studies was also provided. The care of NMD patients was discussed using findings from a case study that was not included in the main review.

How were differences between studies investigated?
Some differences between the included studies were evident from the data tables and were also discussed in the text of the review.

Results of the review
Four controlled trials, including a total of 98 patients with NMD, were included in the review. In most cases patients acted as their own controls. Two studies randomised the order of the different interventions under comparison and two studies used 'normal' patients as controls.

One controlled study (21 NMD patients) compared unassisted inspiration with MIC manoeuvre, MIC plus manual assist, and MI-E. When compared with unassisted inspiration (1.81 litres per second, L/s), MI-E was found to produce the greatest PCF (7.47 L/s; P<0.001), followed by MIC plus manual assistance (4.27 L/s; P<0.001), and MIC manoeuvre (3.37 L/s; P<0.001). However, the manual techniques were not delivered by the same training individual and different machines were used for MIC, thus the consistency and quality of the findings is questionable.

A second controlled trial (8 patients) randomly assigned each of the following treatments with patients acting as their own controls: unassisted cough, manually assisted cough, mechanical insufflation, mechanical insufflation with manual assistance. In comparison with unassisted cough (1.7 L/s), the greatest increase in PCF in non-scoliosis patients was observed with mechanical insufflation with manual assistance (4.1 L/s; P<0.01), followed by manually assisted cough (3.1 L/s; P<0.01) and mechanical insufflation (2.6 L/s; P not significant). There were no significant differences for scoliosis patients.

The third controlled trial (22 NMD adults and children, compared with 19 'normal' age-matched controls) randomly assigned each of the following treatments with patients acting as their own controls: unassisted cough, manually assisted cough, NIV-assisted cough, MI-E and exsufflation alone. The only significant changes in PCF were found for MI-E in paediatric patients, which was more effective than unassisted cough (P<0.001), and MI-E (P<0.001) and exsufflation alone (P<0.01), which were more effective than unassisted cough in adult patients.

Relationship between vital capacity and PCF.

One controlled trial (43 NMD patients with intact bulbar function) trained patients with a vital capacity below 2,000
mL to use breath-stacking techniques and investigated the relationship between vital capacity and PCF. A blinded assessor was used to assess pulmonary function but the study was devised after the data were recorded. The results showed that the lower the vital capacity, the greater the percentage increases in MIC and PCF. MIC increased from 1.4 to 1.7 L (P<0.01) and PCF from 3.7 to 4.3 L/s (P<0.01) in 30 patients; MIC and PCF decreased in the remaining 13 patients. This suggests a relationship between improved MIC and improved PCF, and indicates that breath-stacking techniques may be useful in NMD patients with intact bulbar function.

Authors' conclusions
The use of MI-E for the management of airway secretions improves PCF above the level necessary for effective secretion removal without the need for invasive airway suction. Techniques such as breath-stacking, glossopharyngeal breathing and insufflation alone can also produce an effective PCF in patients with adequate bulbar function. PCF can be further improved by thoracoabdominal thrusts and anterior chest wall compressions, where feasible. Treatment options are more limited for patients with compromised bulbar function, but MI-E may still help delay the necessity for more invasive and hazardous airway clearance techniques. However, further research is required to fully investigate the place of MI-E in the management of patients with NMD.

CRD commentary
This review was based on an adequately defined question in terms of the study design, intervention, population and outcome assessment. The authors searched a number of electronic databases using a list of search terms derived through consensus with their colleagues in the field. However, some studies might have been missed by the inclusion of only English language publications and by not making more specific attempts to locate unpublished material. The authors also failed to report on how studies were selected for the review and how the study data were abstracted, therefore it is not possible to comment on the reliability of the review methods or to assess the risk of bias and error. The authors assessed the quality of the studies using a published tool, but details of their findings were not systematically reported.

The few included studies appeared to be very small and to vary significantly in terms of their study populations, study methods and interventions; the authors were right to present a qualitative summary of their data. Although the findings appear promising, the reliability of the effect sizes is questionable given the limitations and paucity of data. The authors were therefore correct to recommend that large well-designed trials are required to confirm the usefulness of such techniques in the care of patients with NMD.

Implications of the review for practice and research
Practice: The authors did not state any implications for practice.

Research: The authors stated that further research is required to fully investigate the place of techniques to improve PCF. Future studies should optimise treatment settings such as insufflation and exsufflation times, pressures, and the selection of manual or automatic controls. In addition, the frequency of treatments remains to be determined where the techniques are used to maintain vital capacity. Future studies should also ensure that the outcome measures are properly recorded and that there is some qualitative assessment of the patient's experience.

Bibliographic details

Other publications of related interest
Subject indexing assigned by CRD

MeSH
Amyotrophic Lateral Sclerosis /complications; Cough; Muscle Weakness; Muscular Dystrophy, Duchenne /complications; Pulmonary Ventilation; Respiration Disorders; Respiratory Muscles

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Record Status
This is a critical abstract of a systematic review that meets the criteria for inclusion on DARE. Each critical abstract contains a brief summary of the review methods, results and conclusions followed by a detailed critical assessment on the reliability of the review and the conclusions drawn.