Interventions for treating functional dysphonia in adults (Protocol)

Ruotsalainen JH, Sellman J, Lehto L, Jauhiainen M, Verbeek JH

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ABSTRACT

This is the protocol for a review and there is no abstract. The objectives are as follows:
1) To categorise interventions aimed at treating patients diagnosed with functional (non-organic) dysphonia.
2) To assess the effectiveness of interventions for treating functional (non-organic) dysphonia compared to no intervention or an alternative intervention.

BACKGROUND

Voice disorders are generally characterised by abnormalities in pitch, loudness and/or quality of the voice that can limit the effectiveness of oral communication (Ramig 1998). Recent definitions of a disordered voice stress the ability of the voice to fulfill the speaker’s social and occupational requirements (Stemple 1995; Aronson 1985; Sataloff 2000). Due to the difficulties of classifying voice disorders in a systematic way, there is no universally accepted classification system for voice problems (Oates 2004). Traditionally, two major classes of voice disorder have been identified: organic and functional (Fawcus 1986; Oates 2004; Titze 1994).

Functional disorders are characterised by an abnormal quality of voice in the absence of an identifiable lesion. Some clinicians label them as idiopathic, indicating that there is no known cause, while others view them as resulting from the individual’s improper use of his or her voice (Titze 1994). The improper use of voice (also known as vocal misuse) refers to functional voicing behaviours (e.g. excessive shouting or loud talking) and/or functional misuse of vocal components (respiration, phonation, resonance, pitch, loudness and rate) that can contribute to the development of laryngeal pathologies (Stemple 1995). When the classification into functional versus organic emphasises the aetiology of the problem (vocal strain or excessive muscular tension), minor tissue changes such as vocal-fold thickening and vocal nodules are often considered functional (Boone 1987) or behavioural (Fawcus 1986). Therefore, in a strict sense dichotomous classification is undeniably problematic and overly simplistic. In this review we define functional dysphonia as an impaired voice sound and/or reduced vocal capacity (Seifert 2005; Roy 2003) with a possible concomitant diagnosis of minor pathologies of vocal fold cover (nodules, polyps, oedema) that are direct results of either vocal misuse or result from trauma caused to vocal fold tissues by phonatory behaviour.

The prevalence of voice disorders in the general adult population has been suggested as between 3% and 9% in the USA and at about 4% in Australia (Verdolini 2001). In the UK up to 40,000 patients with dysphonia are referred to voice therapy every year (Wilson 1995). Professional voice users such as teachers and singers are at significantly higher risk of developing a voice disorder compared to the general population (Smith 1997; Russell 1998). It has been estimated that at least in developed countries, a well functioning voice is an essential tool for a third of the entire adult working population (Vilkman 2004). In Poland, occupational voice problems ranked highest among all occupational diseases in 2004 (Szeszenia-D. 2005). In a group of 1262 voice patients, the prevalence of vocal pathologies that could be considered as functional dysphonia (no visible pathology, and those deemed psychogenic) or as being direct results of traumatising phonatory behaviour (vocal nodules, oedema, polyps) was 57.6% (Herrington-Hall 1988).

The voice is a multidimensional function that, like physical strength, cannot be measured with any one single scale or test (Hi-
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OBJECTIVES

1) To categorise interventions aimed at treating patients diagnosed with functional (non-organic) dysphonia.

2) To assess the effectiveness of interventions for treating functional (non-organic) dysphonia compared to no intervention or an alternative intervention.

CRITERIA FOR CONSIDERING STUDIES FOR THIS REVIEW

Types of studies

We will consider for inclusion all randomised controlled studies or cluster-randomised trials evaluating the effectiveness of treatments targeted at individuals with functional dysphonia. For environmental or work-related treatment interventions, it is much more difficult to randomise when the intervention is applied at the group level. For this type of intervention we will, therefore, also consider for inclusion prospective cohort studies (otherwise known as controlled clinical trials, controlled before-after studies or quasi-experimental studies).

Types of participants

We will include studies in which the participants are adults (16 or over) who have been diagnosed as having functional / non-organic dysphonia which means that they are experiencing one of the following two symptoms:

1) an impaired voice sound;
2) reduced vocal capacity.

For practical reasons, we will include studies in which a minority of participants may have been diagnosed with minor tissue changes of vocal fold cover (nodules, polyps, oedema) that are regarded as a result of vocal misuse. The voice changes throughout life. In childhood the morphology of vocal fold tissues keeps changing and during puberty the larynx grows (Titze 1994). The three connective tissue layers of the lamina propria, despite being apparent already during puberty, continue to become more differentiated until the age of 16 or 17 (Colton 2006). In advanced age some age-related changes (e.g. ossification, atrophy, dys trophy and oedema) affect phonation (Jasper 2000). However, it is the physiological rather than the chronological age that has a strong impact on how well the larynx functions in phonation (Titze 1994). In this study we consider patients older than 16 years as adults.

We will exclude studies in which the majority of participants have been diagnosed as having any of the following:
• a voice disorder associated with nervous system involvement (e.g. spasmodic dysphonia, essential laryngeal tremor, vocal fold paralysis);
• neurological disorders (e.g. Parkinson’s, Alzheimer’s, ALS, Tourette’s, essential tremor, paralysis);
• organic disease or trauma (e.g. keratosis, contact ulcers, papillomas, laryngeal granulomas and inhalation, thermal etc. trauma);
• the paediatric (e.g. with congenital anomalies) or the geriatric voice;
• carcinoma or other tumours;
• gastro-oesophageal reflux disease.

We will (also) exclude studies in which participants have been diagnosed with a hearing impairment which may affect auditory discrimination.

Types of intervention
We will include studies with any intervention aiming to treat patients diagnosed with functional (non-organic) dysphonia. Possible categories of interventions could be:
1) direct voice therapy;
2) indirect voice therapy and education;
3) other treatment methods.

In the second category we will discern individual and environmental interventions. We will compare interventions with no intervention and, when possible, with alternative interventions.

Types of outcome measures
As primary outcomes we will include patient-reported measures of voice handicap, voice symptoms or voice-related quality of life.

As secondary outcomes we will include all other measurement techniques for establishing the state of vocal or laryngeal performance including:
1) aerodynamic measurements;
2) fundamental frequency and/or intensity;
3) perceptual (visual or auditory) measurements (e.g. stroboscopy, GRBAS, etc.);
4) physiological measurements;
5) acoustic voice analysis (Voice Range Profile or Phonetography), as well as sickness absence and return to work.

SEARCH METHODS FOR IDENTIFICATION OF STUDIES

See: methods used in reviews.

We will search the literature for evaluation studies of interventions for functional voice disorders without restrictions on language or publication. Systematic search strategies have been developed together with the Cochrane ENT Trials Search Co-ordinator and the Cochrane Occupational Health Field Information Specialist for use in MEDLINE, EMBASE and The Cochrane Library. They will be adapted for CINAHL, Psychinfo, the Science Citation Index and the occupational database OSH-ROM. The search string for randomised controlled trials is based on Robinson 2002 and the string for non-randomised studies on Verbeek et al 2005. Since the opportunities for naming and classifying voice disorders and their various treatments are so abundant, the searches will be developed with the aim of maximum sensitivity at the expense of specificity.

Search strategy for MEDLINE through PubMed
#6 (#1 OR #2 OR #3) AND (#4 OR #5)

Search strategy for EMBASE
#1 dysphoni* OR hoarseness OR phonastheni* OR trachyphoni* OR “functional voice disorder” OR “psychogenic voice disorder” OR “ventricular phonation” OR “conversion voice disorder” OR “functional aphonias” OR “conversion aphonias” OR “conversion dysphonias” OR “phonation break” OR “functional falsetto” OR “mutation falsetto” OR puberty phonias OR juvenile voice OR “laryngeal myasthenias”
#2 phonation AND (disease* OR disorder*)
was conducted, the type of study design used, characteristics of each of the included trials regarding the country where the study

Two authors (JR and JS) will independently extract data from for reaching a decision on eligibility.

We will seek to obtain further information from the inclusion criteria. A third author (LL) will resolve any disagreements. We will however assess if allocation was concealed for those assessing the outcome. Two authors (JR and JS) will independently assess trial quality using the quality criteria mentioned in the Cochrane Handbook for the Systematic Review of Interventions. For the appraisal of cohort studies, we will use a validated instrument (Slim 2003). This methodological checklist for non-randomised studies (MINORS) contains 12 items, the first eight of which are specifically for non-comparative studies. MINORS has been shown to have good reliability, internal consistency and validity. The items are scored 0 (not reported), 1 (reported but inadequate) or 2 (reported and adequate). The overall ideal score is 24. Disagreements will be settled through discussion.

References from articles will also be carefully reviewed. Authors of studies and other experts in the field will be contacted for advice on further studies.

**METHODS OF THE REVIEW**

**Selection of trials**

After employing the search strategies outlined above, two authors (JR and JS) will undertake study selection. Both authors will independently assess whether the studies thus found meet the inclusion criteria. A third author (LL) will resolve any disagreements. We will seek to obtain further information from the authors if a paper is found to contain insufficient information for reaching a decision on eligibility.

**Data extraction and management**

Two authors (JR and JS) will independently extract data from each of the included trials regarding the country where the study was conducted, the type of study design used, characteristics of the study participants (as per study inclusion criteria) and types of interventions and outcomes. Results data (means and standard deviations) will also be extracted for possible meta-analyses. Where possible, we will seek missing data from authors. A third reviewer (LL) will resolve any disagreements.

**Quality assessment**

For this review, it will be clear that allocation concealment is not an issue since the nature of treatments for voice disorders renders it impossible for the patients to be unaware of whether or not they are receiving active treatment. We will however assess if allocation was concealed for those assessing the outcome. Two authors (JR and JS) will independently assess trial quality using the quality criteria mentioned in the Cochrane Handbook for the Systematic Review of Interventions. For the appraisal of cohort studies, we will use a validated instrument (Slim 2003). This methodological checklist for non-randomised studies (MINORS) contains 12 items, the first eight of which are specifically for non-comparative studies. MINORS has been shown to have good reliability, internal consistency and validity. The items are scored 0 (not reported), 1 (reported but inadequate) or 2 (reported and adequate). The overall ideal score is 24. Disagreements will be settled through discussion.

**Data analysis**

For interventions directed at individuals, we will only use randomised controlled trials to draw conclusions, but for work or environment directed interventions that are applied at the group level we will also include controlled clinical trials as defined before.

The choice between qualitative and quantitative pooling will be based first on clinical homogeneity. Clinically homogeneous studies will be defined as those with similar populations, interventions and outcomes measured at the same follow-up point.

We will pool studies with sufficient data, judged to be clinically homogeneous, with RevMan 4.2 software. We will also test for statistical heterogeneity. When studies are statistically heterogeneous, a random-effects model will be used, otherwise a fixed-effect model will be used. All estimates will include a 95% confidence interval (CI).

The results of each trial will be plotted as point estimates, such as relative risks (RR) for dichotomous outcomes, mean and standard deviation (SD) for continuous outcomes, or other data types as reported by the authors of the studies. When the results cannot be plotted, they will be described in the table of included studies, or the data will be entered into the 'Additional Tables' section of the review. For continuous measures, preference will be given to analysing the results with weighted mean differences (WMD) because these results are easier to interpret for clinicians and other readers. If this is not possible, then standardised mean differences (SMD) or effect sizes will be used.

A qualitative analysis will be completed if the studies are found to be clinically heterogeneous, or if the relevant data to complete
statistical pooling are unavailable. A rating system, based on the Levels of Evidence, will be used to summarise the strength of scientific evidence of the effects of the treatment. The rating system will be based on both the quality and the outcome of the studies (Van Tulder 2003):

I. Strong evidence - consistent evidence in multiple high quality randomised controlled trials
II. Moderate evidence - consistent findings in multiple low quality randomised controlled trials and/or controlled clinical trials and/or one high quality randomised controlled trial
III. Limited evidence - one low quality randomised controlled trial or controlled clinical trial
IV. Conflicting evidence - inconsistent findings in multiple randomised controlled trials and/or controlled clinical trials
V. No evidence - no randomised controlled trials or controlled clinical trials.

The outcome of the studies will be considered 'consistent' if at least 75% of the trials report statistically significant results in the same direction.

For a sensitivity analysis, the results will be analysed again, including only high quality studies, to find out if the quality level leads to changes.

We expect to complete this review within one year of the publication of this protocol in The Cochrane Library. The authors also intend to perform a new search for trials every two years and to update the review accordingly.

POTENTIAL CONFLICT OF INTEREST

None known.

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Jacobson 1997

Jasper 2000

Ma 2001

Oates 2004

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Robinson 2002

Roy 2003

Russell 1998

Sataloff 2000

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Slim 2003

Smith 1997

Stemple 1995

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Titze 1994

van Tulder 2003

Verbeek et al 2005

Verdolini 2001

Vilkman 2004

Wilson 1987

Wilson 1995

COVER SHEET

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Interventions for treating functional dysphonia in adults

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Contribution of author(s)

Jani Ruotsalainen conceptualised the review jointly with JV and took the lead in writing the protocol.
Jos Verbeek conceptualised the review jointly with JR and wrote the methods section of the protocol.
Merja Jauhiainen designed the systematic search strategies in collaboration with the Cochrane ENT Group's Trials Search Co-ordinator.
Jaana Sellman and Laura Lehto wrote the second version of the protocol.

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