Does therapy with biofeedback improve swallowing in adults with dysphagia? A systematic review and meta-analysis

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Conflicts of Interests

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ABSTRACT

Objective: To describe and systematically review the current evidence on the effects of swallow therapy augmented by biofeedback in adults with dysphagia (PROSPERO 2016:CRD42016052942).

Data sources: Two independent reviewers conducted searches which included MEDLINE, EMBASE, trial registries and grey literature up to December 2016.

Study selection: Randomised controlled trials (RCTs) and non-RCTs were assessed, including for risk of bias and quality.

Data extraction: Data were extracted by one reviewer and verified by another on biofeedback type, measures of swallow function, physiology and clinical outcome, and analysed using Cochrane Review Manager (random effects models). Results are expressed as weighted mean difference (WMD) and odds ratio (OR).

Data Synthesis: Of 675 articles, we included 23 studies (n=448 participants). Three main types of biofeedback were used: accelerometry, surface electromyography and tongue manometry. Exercises included saliva swallows, manoeuvres and strength exercises. Dose varied between 6-72 sessions for 20-60 minutes. Five controlled studies (stroke n=95; head and neck cancer n=33; mixed aetiology n=10) were included in meta-analyses. Compared to control, biofeedback augmented dysphagia therapy significantly enhanced hyoid displacement (three studies, WMD=0.22cm; 95% CI [0.04, 0.40], p=0.02) but there was no significant difference in functional oral intake (WMD=1.10; 95%CI [-1.69, 3.89], p=0.44) or dependency on tube feeding (OR =3.19; 95%CI [0.16, 62.72], p=0.45). Risk of bias was high and there was significant statistical heterogeneity between trials in measures of swallow function and number tube fed (I^2 70-94%). Several non-validated outcome measures were used. Subgroup analyses were not possible due to a paucity of studies.

Conclusions: Dysphagia therapy augmented by biofeedback using surface electromyography and accelerometry enhances hyoid displacement but functional improvements in swallowing are not evident. However data are extremely limited and further larger well-designed RCTs are warranted.

Key words: Biofeedback, dysphagia, rehabilitation

Dysphagia is increasingly common in an aging population with reports of symptoms occurring in 40% of adults over 65.[1] Prevalence of dysphagia varies according to aetiology; 55 % of stroke patients,[2] 11–81 % in Parkinson's disease,[3] 11-93.5% in head and neck cancer [4] and more than 90% of patients with motor neurone disease.[5] Dysphagia can cause complications such as aspiration pneumonia, dehydration and malnutrition and lead to increased mortality.[6-8] Early assessment, tube feeding, texture modification and adaptive strategies can reduce these risks [9-11] but can impact on quality of life.[12]

Therapeutic interventions which aim to maintain or rehabilitate swallowing vary from muscle strengthening exercises, swallow skill exercises, sensory stimulation and emerging techniques such as peripheral and central stimulation.[13] Feedback is advocated for enhancing outcomes in rehabilitation.[14] Error-based learning where the learner has knowledge of the errors they need to correct, can aid learning or re-learning a skill.[15] When this information about performance is given based on kinematic measures it is called biofeedback.

In post stroke upper limb therapy, performance feedback enhances motor recovery.[16] Further, biofeedback has resulted in moderate to large treatment effects in gait when compared to usual therapy[17] and has been shown to be beneficial with other physical, psychological, cardiac and respiratory conditions;[18] for example, using a decibel meter has been widely used in speech therapy, for increasing voice volume and quality in patients with Parkinson's disease.[19] During swallowing, intrinsic feedback is acquired from sensation within the oral cavity and pharynx, but it may be suboptimal or impaired in patients with dysphagia. There is a progressive reduction in pharyngeal and laryngeal sensation with increasing age [20] and sensory impairments can be one of the characteristics of dysphagia in many aetiologies.[21-24] Making accurate judgements about subtle differences in the efficacy of the pharyngeal stage of swallowing is difficult without instrumental measurements.[25]

Biofeedback in swallowing therapy is not routinely used to augment dysphagia therapy [26] nor is there national recognition and guidance regarding its use. However, it is gaining more interest and several commercially available biofeedback instruments and software are on the market and so there is a need to evaluate its effectiveness. We performed a systematic review and meta-analysis to describe the current evidence on the effects of dysphagia therapy with all types of biofeedback in adults with dysphagia in order to discover the most superior methods.

METHODS

This review aimed to answer the following questions in adults with dysphagia: Does biofeedback paired with dysphagia therapy, as compared with no biofeedback, improve (1) Functional swallowing outcomes? (2) Clinical outcomes? (3) Swallow physiology?

The protocol was registered with Prospero (2016:CRD42016052942) in December 2016. Studies were eligible for inclusion if they were full text, English language studies that involved dysphagia therapy using biofeedback in adults with any aetiology resulting in acquired oropharyngeal dysphagia and reported pre- and post-swallowing measures and/or clinical outcomes. Two independent reviewers conducted electronic searches from when records began until December 2016 of the following databases: Cochrane Stroke Group

Trials Register, MEDLINE, EMBASE, CINAHL, Conference Proceedings Citation Index-Science (CPCI-S) and Web of Science. Reviews of reference lists, conference abstracts and internet searches were conducted to ensure inclusion of unpublished or ongoing trials. Authors were contacted where partial or incomplete data were not available. An example of the search strategy for the MEDLINE search is included in Figure 1.

Study selection

Two reviewers (JB and LE) searched the title and abstracts of the studies and excluded those that were not relevant. If there were any doubts the full text was sought. Once the full text was obtained the same reviewers selected the relevant studies for (1) A descriptive analysis of the types and application of biofeedback used in dysphagia therapy, and (2) Those meeting criteria for inclusion in a meta-analysis. Any disagreements were resolved with a third reviewer TE. Only those with a non-confounded control group and outcome data were included in the meta-analysis.

Data acquisition

Data were extracted using a predesigned and piloted proforma by one reviewer, JB and then verified by a second reviewer, LE. Authors were contacted if data were not available. TE resolved any discrepancies.

Risk of bias

Randomised control trials (RCTs) were assessed for risk of bias and quality as recommended in the Cochrane Handbook.[27] This included assessing methods of randomisation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data and selective outcome reporting. Non RCTs were assessed using a combination of different tools for non RCTs and observational studies [28-30] and included assessing quality of study designs for small N and N=1 studies, data analysis, generalisability, replicability, blinding, incomplete and selective reporting.

Statistical analysis

Review Manager (version 5.3) was used to derive odds ratios (OR) and confidence intervals (CIs) for dichotomous data and mean difference (MD) and CIs for continuous data. Study data were combined if the outcome measures used were comparable. In the Aoki 2015 study the mean and standard deviation (SD) were estimated from the median and range using published formulae [31]. Heterogeneity was assessed between different studies for each measure. Sub-group analysis was planned to examine whether biofeedback type, dose, aetiology of dysphagia or setting made a difference to outcome.

RESULTS

Initial searches identified 669 articles, and a further 6 were found through searching grey literature. After screening titles and abstracts, full text was sought for 53 studies. One full text article could not be obtained but there was sufficient detail in the abstract to be included in our analysis. [32] Of those, 23 were suitable for inclusion in a qualitative synthesis and 5 met the criteria for inclusion in the meta-analysis (Table 1, Figure 2).

Study characteristics

Twenty-three studies (n=448 participants) described dysphagia interventions with biofeedback in adults with structural, neurological and psychological dysphagia (Table 1). The three main types of biofeedback used were surface electromyography (sEMG, n=164), accelerometry (n=150) and tongue manometry (n=67). Less frequent forms of biofeedback included videoendoscopy (n=33), respiratory plethysmography (n=30) and external laryngeal manometry (n=4). There was no type of biofeedback exclusive to a specific patient group. Dosing and frequency of therapy varied across studies and across types of biofeedback; from 4 to 72 sessions carried out twice daily to fortnightly.[33-36] Over 80% of studies reported 2 or more sessions per week. Overall, treatment sessions varied in length across study and type of biofeedback and lasted between 20-60 minutes with 45-60 minutes being the most common (50%).

Accelerometry

Five studies used accelerometry as a means of biofeedback. This consists of a small accelerometer being placed just above the thyroid cartilage. It measures the epidermal vibrations caused by the internal sounds and vibrations of the superior/inferior and or anterior/posterior movements of the hyoid and larynx during swallowing.[37] The vibrations are converted into a voltage signal, which the patient can use as visual feedback to facilitate their swallowing therapy. In three of the studies, feedback was presented as a graph on a computer screen with instruction to match the shape of a signal derived from a normal swallow.[38-40] In one study, the signal from the accelerometer was converted into an animation of a frog swallowing a mosquito at different locations on a screen. [41] The target was adjusted based on performance. Another study used signals from accelerometry and surface electromyography (sEMG) in a similar virtual reality game.[42] Only one of these studies had a control group, [41] which reported that accelerometry significantly improved functional intake (functional oral intake scale, FOIS, p=0.014) and hyoid displacement (p=0.07) compared to control which received the same intensity of exercise without biofeedback. The other four accelerometry studies were of lower quality and also reported functional improvements in swallowing following the therapy.

Tongue manometry

Five studies used tongue manometry for biofeedback.[33, 43-46] This intervention consists of using a 2cm x 1cm x 0.5 cm air filled pressure bulb which acts as a pneumatic pressure sensor and measures isometric tongue strength. The bulb is placed on the tongue and the participant is instructed to push the tongue against the hard palate. The pressure generated is measured by a manometer and the signal can be displayed graphically on a screen to give patients biofeedback. Four studies used the Iowa Oral Performance Instrument (IOPI)

[33, 43, 45, 46] and one used a Japanese version manufactured by Japan Medical Supply Ltd (JMS).[44] Robbins and colleagues used isometric anterior and posterior tongue strength exercises with the aim of increasing muscle strength and mass to lead to improvements in functional swallow.[33] The other four studies used isometric tongue strengthening, tongue strength accuracy exercises and tongue strength during saliva swallow exercises.[43-46] One study used a control group which received tongue exercises without biofeedback at the same intensity.[44] They described significant differences in mean change between treatment and control groups on maximum isometric pressure (p=0.03), swallowing tongue pressures (p=0.014) and motor function of swallowing structures – Mann Assessment of Swallowing Ability (MASA) (p=0.04), but no significant differences between groups on swallow function. Four other studies of poor design reported positive outcomes in tongue strength[33, 43, 45, 46] . Moreover, reductions in vallecular[45] and pharyngeal wall residue[33] were observed on videofluoroscopy but the findings are contradicted in other studies where residue scores were neutral[33] or worse[43]. Only one of the studies described a positive functional swallowing outcome, [33] but no recognisable or specific outcome measures were presented.

Surface Laryngeal Manometry

One study used an air-filled balloon fixed externally to the cervical region to measure changes in pressure during swallowing.[47] Participants practised an effortful swallow and were given numerical feedback about their performance. It was a small study and there was no control group but the 4 patients with dysphagia secondary to Parkinson's reported improvements in swallow function following the intervention.

Surface Electromyography (sEMG)

Ten studies used sEMG as a means of providing biofeedback. sEMG measures the spatial and temporal properties of muscle action potentials. The amplitude of the signal increases with increased force of muscle contraction.[48] In 9 of 10 studies, sEMG was used to measure the activity of the muscles which elevate and tilt the larynx during the pharyngeal swallow (the remaining study utilised sEMG in a patient with psychogenic dysphagia).[49] Two small electrodes are placed on the submental muscles (mylohyoid, geniohyoid, anterior belly of digastric and genioglossus) and a third reference electrode is usually placed to one side.[50] The sEMG signal represents the timing and force of the muscle contraction and is displayed graphically on a screen. sEMG has been employed using a variety of strategies, such as providing progressively more challenging targets based on strength and timing;[51] and enhancing the design of a swallow protocol helping the participant with timing of muscle contraction and respiratory patterns.[52] The remaining studies used biofeedback to teach and practice either or both effortful swallow and the Mendelsohn manoeuvre (holding the larynx elevated for a target number of seconds).[34, 36, 50, 53-56] Two studies met the criteria to be included in a meta-analysis. McCullough et al used sEMG biofeedback to teach and practice the Mendelsohn manoeuvre to patients who had dysphagia secondary to stroke. The data was reported in two papers,[34, 57] demonstrating significant improvements in duration of hyoid elevation (p=0.011) and anterior hyoid movement (p=0.009) but no other physiological or functional changes were found. Huimin et al provided swallow function training with biofeedback compared to swallow function training without biofeedback and reported significant changes post intervention in the biofeedback group in upper oesophageal sphincter (UES) opening (p=0.001), pharyngeal transit time (PTT) (p=0.038) and maximum hyoid displacement (p=0.033).[32] Although in the remaining 8 studies design quality was poor, significant improvements were reported in functional and physiological swallowing measures.

Videoendoscopy

One study used videoendoscopy as a means of biofeedback.[58] This involves the insertion of a flexible nasoendoscope to the level of the soft palate so that the pharynx and larynx can be visualised. The timing, safety and efficiency of the swallow can also be visualised and used for biofeedback. Denk et al taught patients to employ swallowing manoeuvres and changes in posture using videoendoscopy for direct visual biofeedback. The manoeuvres included effortful swallow, Mendelsohn manoeuvre, supraglottic swallow and supra-supra glottic swallow depending on the nature of each participant's dysphagia. This study met the criteria for inclusion in a meta-analysis. The control group received the same intensity of therapy and exercise type without the biofeedback. All participants were tube fed initially and 73% of patients achieved therapeutic success, defined as tube removal and full and unrestricted oral intake. At 40 days, significantly more of the biofeedback group had achieved therapeutic success (p=0.041) however there was no significant difference between the intervention and control groups at 6 months.

Respiratory plethysmography

One study used respiratory inductance plethysmography and nasal airflow as a method of biofeedback to train participants to adopt a natural respiration/swallow pattern.[35] Nasal airflow is measured by a nasal cannula and respiratory inductance plethysmography measures movements of the ribcage and abdomen. These devices were attached to a Kay Pentax Digital Swallowing Workstation via Swallow Signals Lab which processed the signals and presented the respiration patterns on a screen for the patients to use as feedback. They went through identification, acquisition and mastery stages to learn to swallow mid expiration with a mid to low lung volume and exhale post swallow. Significant improvements were reported with swallow physiological measures and swallow respiratory patterns but the there was no control group to compare outcomes.

Quantitative synthesis

Five studies had a non-confounded control group and thus met the criteria for inclusion in the meta-analysis (N=138).[32, 34, 41, 44, 58] Two were excluded because two different interventions were compared.[45, 55] The remaining 18 were excluded because they did not include a control group nor did they demonstrate an observational study design of sufficient quality. Study quality was variable (Table 2) with at least one element of bias evident in all of the studies.

Due to the range of outcome measures used, data from only three outcomes could be synthesized. Biofeedback did not improve swallow function (FOIS, t=2, n=51, MD=1.10; 95% CI [-1.69, 3.89], Figure 3A); or clinical outcome (feeding tube removal, t=2, n=53, OR =3.19; 95% CI [0.16, 62.72], Figure 3C). Biofeedback intervention had a significant positive effect on swallow physiology, specifically hyoid displacement (t=3, n=90, MD=0.22; 95% CI [0.04, 0.40], Figure 3B); two of these studies used sEMG and one used accelerometry (Table 1). There was significant statistical heterogeneity between trials in measures of swallow function and number tube fed (I^2 = 70-94%) and low in physiological measures (I^2 = 8%). Sub-group analyses were planned to explore effects of biofeedback type, aetiology of dysphagia, setting and dose, including assessment for publication bias, but this could not be performed due to the paucity of studies.

DISCUSSION

There is an absence of good quality, large-scale RCTs assessing biofeedback as an adjunct to therapy for dysphagia in adults. Meta-analysis of controlled studies showed a positive effect of biofeedback on one swallow physiology outcome; maximum displacement of the hyoid bone. No conclusions can be drawn from other positive results in functional, physiological and clinical outcome measures reported in several small, non-randomised controlled trials.

Three controlled trials found that biofeedback augmented dysphagia therapy resulted in increased hyoid displacement [32, 41, 57] when compared to a control. Two of these studies used sEMG and the other used accelerometry for biofeedback, both of which show patients a representation of hyolaryngeal elevation. Studies with healthy subjects have demonstrated that increases in sEMG amplitude correlate with onset and offset of hyoid [59] and laryngeal elevation.[60] The sEMG signal represents activity predominantly from mylohyoid, anterior belly of the digastric, and the geniohyoid muscles, confirmed using intra-muscular EMG.[61] sEMG amplitude increases with effortful swallowing [62] and the peak accelerometry signal correlates with peak laryngeal elevation.[63] Biofeedback is used with the aim of improving timing, strength and duration of hyolaryngeal elevation. Therefore, it stands to reason that therapy targeting hyolaryngeal elevation results in corresponding physiological changes in hyoid displacement. Li et al reported functional changes in swallowing in their accelerometry study but unfortunately the other two studies did not report any data on functional outcome. Whether physiological change results in improvements in functional swallowing remains unclear. Three trials (using tongue manometry, [44] accelerometry [41] and videoendoscopy [58]) reported improvement in swallow function [41, 44] and tube removal post biofeedback intervention.[41, 58] However, when pooled in the meta-analysis these became neutral and non-significant.

These results need to be interpreted with caution since different types of biofeedback were used across studies and so heterogeneity was high. Included studies were also limited by both trial design and small sample size. For example McCullough et al used a cross over design in a heterogeneous population, a mix of subacute and chronic stroke participants, which will naturally recover at different rates.[57] In addition, they did not report the time allowed for treatment wash-out (if one exists) or any data in the crossover period, hence both treatment and 'control' groups received the intervention. Aoki and colleagues also had unmatched groups at baseline with more severe dysphagia in the intervention group, further confounding interpretation.[44] The causes of dysphagia in this trial were also mixed, hence understanding the results must be put into context of aetiology and the potential variation in response to treatment.

Biofeedback might enhance recovery and improve aspiration risk in the short-term but may not lead to significant gains in the long-term. In patients with head and neck cancer, Denk reported a significant difference in means between groups at 40 days but not at the end of the study (6 months).[58] The authors suggest that biofeedback helps patients learn manoeuvres and exercises but once learnt, the biofeedback has no benefit. If so, these early gains could be beneficial for those with dysphagia secondary to multiple causes – it may mean quicker return to full normal intake, improve a patient's quality of life, reduce morbidity, length of stay in hospital and health costs. Whether biofeedback for dysphagia is beneficial or not in both the short and long term needs further investigation.

Across all the biofeedback intervention studies included in the qualitative analysis, heterogeneity in method and therapy exercise was observed, hence it is important to use appropriate outcome measures depending on the mechanism targeted. Accelerometry and sEMG biofeedback enables a representation of the strength and duration of hyolaryngeal elevation; 6 of 15 studies aimed to increase hyolaryngeal elevation[32, 38, 39, 41, 50, 57] but only four measured this as an outcome.[32, 41, 50, 57] The remaining studies aimed to improve swallowing skill and measured function or overall severity. Tongue manometry aims to improve lingual strength and timing; 4 of 5 studies [43, 45, 46, 64] measured this and oral control appropriately as an outcome. The study utilising respiratory plethysmography measured coordination of breathing and swallowing which is the mechanism it was targeting in therapy.[35] Videoendoscopy enabled feedback should measure changes in swallow safety and efficiency and physiological changes dependent on the strategies learnt i.e. Mendelson manoeuvre targets hyolaryngeal elevation. However, in the included study only 'therapeutic success' (defined as tube removal and return to full oral diet) was measured.[58]

Biofeedback is often used in physiotherapy to augment skill based therapy and skill training results in better functional outcomes than non-specific strength training in adults post stroke [65]. All but one of the studies included in the qualitative synthesis used the task of swallowing as either the target exercise or one of the exercises within the therapy sessions. This involved exercises and strategies to improve the strength, timing and/or duration of the swallow. Further work is needed to determine whether biofeedback paired with swallow skill vs strength training results in better outcomes.

It is not known if biofeedback may be better focussed on specific types of dysphagia, or whether it can be applied more generally. In the present review, only four studies included patients with a specific type of impairment that the biofeedback targeted, none of which were included in the meta-analysis. Three tongue manometry studies included patients if they had poor oral control and/or reduced lingual strength.[45, 46, 64] One of the sEMG studies included patients only if they had evidence of reduced hyolaryngeal excursion.[50] The remainder included patients with any type of swallowing impairment or any type of pharyngeal dysphagia. The diverse range of methods used with biofeedback provides a challenge in selecting the most appropriate technique for future studies. This will also depend on the expected natural progression of the underlying cause of dysphagia in the population studied. Defining the nature of the swallowing impairment in future studies will help to identify which patients might benefit from specific forms of biofeedback.

Due to the paucity of studies, sub group analysis was not possible to investigate whether one type of biofeedback was more efficacious over others, whether specific impairments respond better to biofeedback, or the optimal dose of therapy relative to outcomes, and timing of intervention. Therefore there is insufficient evidence to guide clinicians in the use of biofeedback and its use will be dependent on the local resource.

Study Limitations

Several limitations should be considered when interpreting our results. Selection bias may be present but this risk was minimised by searching a range of databases and grey literature, and using two reviewers to search and select appropriate publications. Authors were contacted when information was not available in the text, although there was a limited response to these requests. Only English language studies were included which increases a risk of bias towards publications in larger English language international journals, which possibly tend towards studies with positive results. One Chinese article with sufficient detail in an English abstract was included despite no access to the full text.[32] However, there were limited methodological details available such as the means of measuring hyoid elevation and thus it was impossible to assess its full risk of bias and quality. A second limitation in interpreting this review is the paucity of good quality RCTs with blinding and transparent reporting of data. Most of the studies identified were single case studies or small studies with no control groups. There is also an absence of good quality observational or longitudinal studies which that use pre-interventional measures as a comparator. We have purposely been broad on the inclusion of studies in the meta-analysis because there are so few. It would be easy to exclude all of them on the basis of quality. Therefore, the outcomes must be interpreted with caution. For example two of the five studies in the meta- analysis had a control group that did not receive exactly the same intervention [44, 57]. The control groups in the remaining three studies received the same type and intensity of exercise – the only difference being biofeedback [32, 41, 58]. Thus, the meta-analysis may not solely tell us about the augmentative effects of biofeedback per se but the effects of biofeedback paired with a variety of exercises. Third, the variety of outcome measures limited the amount of data that could be pooled in meta-analyses and some studies reported only outcomes in swallow physiology or performance on a target exercise but these do not necessarily signify meaningful change for patients.

CONCLUSIONS

Dysphagia therapy augmented by biofeedback seems to improve physiological outcome, specifically hyoid displacement, but whether this translates to functional improvements is not clear. However, data were obtained from small studies at high risk of bias and conclusions must be interpreted with caution. Further good quality research is required to guide whether biofeedback augmented dysphagia therapy leads to better outcomes for patients with dysphagia. Particular attention should address specific populations (aetiology and dysphagia type) with clearly defined timing of administration relative to the onset of dysphagia. Further, the dose of swallow therapy (number, length and intensity of sessions) paired with biofeedback is unknown and should be assessed using well-designed, randomised controlled trials. Further research is also needed establishing validated and meaningful outcome measures following swallow therapy.

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Figure Legends

Figure 1. Search strategy for MEDLINE

Figure 2: Study flow diagram

Figure 3. Results from Meta-analysis (Review Manager 5.3) showing changes in A) function, B) physiology and C) clinical outcome in patients receiving swallowing therapy with biofeedback compared to usual care.

Table Legends

Table 1: Summary of included studies

Table 2: Risk of bias in the studies included in the meta-analysis

Table 1: Summary of included studies

Author	Biofeedback device	N	Exercise	Aetiology	Intensity	Frequency	Duration	Outcomes
Aoki 2015* [44]	Tongue manometry - JMS	34	TS and ES	23 stroke 11 mixed aetiology	45 mins	5 days/ week	3 weeks	Improvement in tongue strength and swallow physiology (MASA) post therapy, (but no significant difference between groups). Control group received the tongue exercises at the same intensity.
Athukorala 2014 [51]	sEMG	10	SS	Parkinson's Disease (PD)	60 mins	5 days/ week	2 weeks	Improvement in swallow physiology (timed swallow test and VFS) post therapy
Bogaardt 2009 [36]	sEMG	11	MM	Stroke	20 mins	1-2 x fortnight	4-24 weeks	Improvement in swallow function (FOIS) and tube status post therapy
Bryant 1991 [53]	sEMG	1	MM and ES	Head & Neck Cancer (H&N Ca)	no info	3 x week	10 weeks	Subjective improvement in swallow severity and tube status
Carnaby-Mann 2009 & 2010 [a] [55, 56]	sEMG	24	MM and ES	Mixed	60 mins	5 x week	up to 3 weeks	Improvement in swallow function (FOIS) and tube status post therapy (less improvement than case group)
Crary 2004 [52]	sEMG	45	Fixed swallow protocol	Mixed	50 mins	5 days/week	3-4 weeks	Improvement in swallow function (FOIS) and tube status post therapy
Denk, 1997* [58]	Videoendoscopy	33	MM, ES, SGS, SSGS	H&N Ca	45 mins	2-5 days per week	up to 6 months	Improvement in tube status post therapy - no significant difference between groups at the end of the study (6 months). The control group received the same intensity and type of intervention without biofeedback.
Felix 2008 [47]	External laryngeal manometry	4	ES	PD	no info	5 days/week	2 weeks	Subjective improvement in swallow function post therapy
Hageman DASI web [40]	Accelerometry	103	SS	Mixed	no info	Unknown	3 months	Improvement in Swallow Function and Pneumonia Risk scale - 92% made average of 2-point improvement post therapy
Haynes 1976 [49]	sEMG	1	Relaxation	Psychogenic dysphagia	30 mins	1-2 x week	11 weeks	Subjective improvement in swallow function post therapy
Huckabee 1999 [54]	sEMG	10	MM and ES, Shaker, Masako	Brainstem injury	60 mins	2 x day	5 days	Improvement in swallow function (own scale) and tube status post therapy
Huimin 2015* [†] [32]	sEMG	36	Functional swallow training	Stroke	Unknown	6 days/week	4 weeks	Improvement in swallow physiology (pharyngeal transit time, UES opening and maximum hyoid displacement compared to control group (same intervention with no biofeedback)
Krishnan 2013 [39]	Accelerometry	1	SS with target	PD	30 mins	3 x week	2 weeks	Subjective improvement in oral intake post therapy
Li 2016* [41]	Accelerometry	20	SS, ES & MM with targets	Stroke	60 mins	3 x week	5-6 weeks	Significant improvement in hyoid displacement, function (FOIS) and tube status compared to control group (same intervention with no biofeedback)

Li 2016 [42]	Accelerometry & sEMG	21	SS with target	Mixed	60 mins	3 x week	5 weeks	Improvement in swallow function (FOIS) and tube removal post therapy
Martin-Harris 2015 [35]	Airflow and inductance plethysmography	30	Swallows on expiration	H&N Ca	60 mins	2 x week	up to 4 weeks	Improvement in swallow breathing coordination, aspiration (PAS) and MBS Imp sub scores post therapy (no meaningful difference in swallow function/QOL (MD Anderson Dysphagia Inventory))
McCullough 2012 & 2013* [a] [34, 57]	sEMG	18	MM	Stroke	45-60mins	2 x day	2 weeks	Improvement in hyoid displacement post therapy, no improvement in other physiological or functional measures. Cross over design – intervention vs no intervention
Reddy 2000 [38]	Accelerometry	5	SS, MM - with target	Mixed	30 mins	1-3 x week	3-9 weeks	Subjective improvement in dysphagia severity on VFS pre therapy
Robbins 2007 [33]	Tongue manometry - IOPI	10	TS	Stroke	no info	3 x day/3 days per week	8 weeks	Improvement in tongue strength and aspiration (PAS) post therapy but no or variable improvement in other physiological measures.
Steele 2012 [50]	sEMG	8	SS, ES & MM with targets	Mixed	Unknown	Unknown	Unknown	Improvement on swallow strength (sEMG) post therapy variable improvement on physiological measures
Steele 2013 [43]	Tongue manometry - IOPI	6	TS and ES	Traumatic Brain Injury	no info	2 x week	11-12 weeks	Improvement in tongue strength and aspiration (PAS) post therapy but no or variable improvement in other subjective and physiological measures. Worsening of residue.
Steele 2016 [45]	Tongue manometry - IOPI	14	TS and ES	Stroke	no info	2-3 x week	8-12 weeks	Improvement in tongue strength post therapy but no or variable improvement in other physiological measures pre and post therapy
Yeates 2008 [46]	Tongue manometry - IOPI	3	TS and ES	Mixed	45 mins	2-3 x week	8-12 weeks	Improvement in tongue strength post therapy but variable improvement in other subjective and physiological measures

* included in meta-analysis; [a] same data presented in both studies; [†] abstract data only

MM = Mendelsohn manoeuvre; SS = saliva swallow; ES = effortful swallow; SGS = supraglottic swallow; SSGS = super supraglottic swallow; TS = tongue strength.

Table 2: Risk of bias in the studies included in the meta-analysis.

Study	Suitable control	Random sequence generation	Allocation concealment	Blinding of participants/ therapist	Blinding of assessors	Incomplete data	Selective reporting
Aoki 2015 [44]	+	-	Unknown	+	+	+	+
Denk 1997 [58]	+	Unknown	Unknown	-	-	-	-
Li 2016 [41]	+	-	-	-	-	+	+
McCullough 2012 & 2013 [34]	+	+	-	-	+	+	-
Huimin 2015 [32]	+	+	Unknown	Unknown	Unknown	Unknown	Unknown

+ low risk of bias/good quality; **-** high risk of bias/poor quality

1. exp Deglutition Disorders/
2. oropharyngeal dysphagia.mp.
3. oro-pharyngeal dysphagia.mp.
4. dysphagia.mp.
5. 'swallowing impairment'.mp.
6. deglutition disorder.mp.
7. 1 or 2 or 3 or 4 or 5 or 6
8. exp Biofeedback, Psychology/
9. biofeedback.mp.
10. Feedback, Physiological/ or Feedback/ or Feedback, Sensory/ or Feedback, Psychological/
11. feedback.mp.
12. 'skill therapy'.mp.
13. (swallow* adj3 (therap* or exercise* or intervention* or rehabilitat* or train*)).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
14. 8 or 9 or 10 or 11 or 12 or 13
15. exp Deglutition/
16. deglutition.mp.
17. swallow*.mp.
18. 15 or 16 or 17
19. 7 and 14 and 18
20. limit 19 to (english language and humans)

Figure 1. Search strategy for MEDLINE

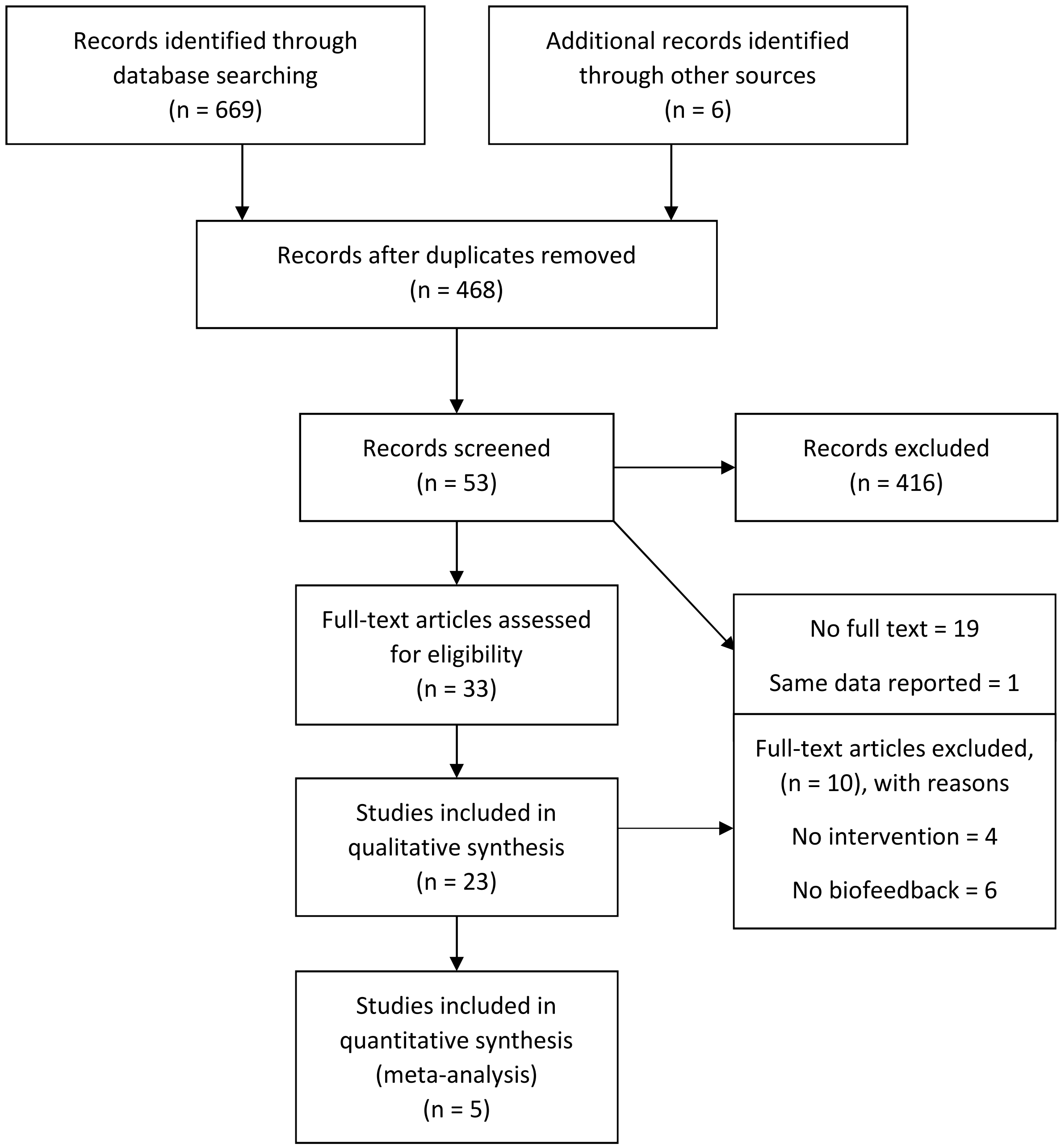
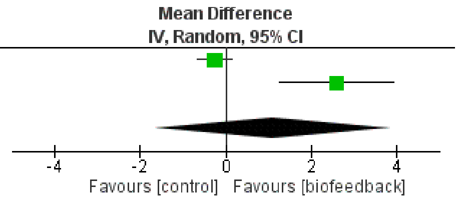


Figure 2: Study flow diagram

A) Functional Oral Intake Scale (FOIS)

Study or Subgroup	Experimental Mean	Experimental SD	Experimental Total	Control Mean	Control SD	Control Total	Weight	Mean Difference IV, Random, 95% CI
Aoki 2015	4.75	0.77	17	5	0.33	14	52.6%	-0.25 [-0.65, 0.15]
Li 2016	5.1	1.2	10	2.5	1.78	10	47.4%	2.60 [1.27, 3.93]
Total (95% CI)			**27**			**24**	**100.0%**	**1.10 [-1.69, 3.89]**

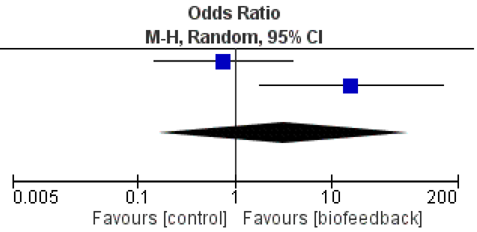
Heterogeneity: Tau² = 3.81; Chi² = 16.13, df = 1 (P < 0.0001); I² = 94%
Test for overall effect: Z = 0.77 (P = 0.44)



B) Numbers with feeding tube removed

Study or Subgroup	Experimental Events	Experimental Total	Control Events	Control Total	Weight	Odds Ratio M-H, Random, 95% CI
Denk 1997	14	19	11	14	53.0%	0.76 [0.15, 3.92]
Li 2016	8	10	2	10	47.0%	16.00 [1.79, 143.15]
Total (95% CI)		**29**		**24**	**100.0%**	**3.19 [0.16, 62.72]**
Total events	22		13			

Heterogeneity: Tau² = 3.66; Chi² = 4.76, df = 1 (P = 0.03); I² = 79%
Test for overall effect: Z = 0.76 (P = 0.45)



C) Hyoid displacement (cm)

Study or Subgroup	Experimental Mean	Experimental SD	Experimental Total	Control Mean	Control SD	Control Total	Weight	Mean Difference IV, Random, 95% CI
Huimin 2015	1.3	0.6	17	0.9	0.4	19	27.5%	0.40 [0.06, 0.74]
Li 2016	1.45	0.26	10	1.34	0.25	10	57.1%	0.11 [-0.11, 0.33]
McCullough 2012 & 2013	1.99	0.72	17	1.68	0.64	17	15.4%	0.31 [-0.15, 0.77]
Total (95% CI)			**44**			**46**	**100.0%**	**0.22 [0.04, 0.40]**

Heterogeneity: Tau² = 0.00; Chi² = 2.17, df = 2 (P = 0.34); I² = 8%
Test for overall effect: Z = 2.35 (P = 0.02)

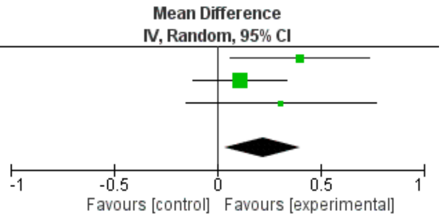


Figure 3. Results from Meta-analysis (Review Manager 5.3) showing changes in A) function, B) clinical outcome and C) physiology in patients receiving swallowing therapy with biofeedback compared to usual care.