



Review

Neuromuscular Electrical Stimulation for Dysphonia and Dysarthria: A Systematic Review

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ABSTRACT:

Introduction: People with speech disorders such as dysarthria and dysphonia have difficulty producing speech, which is often related to speech muscle dysfunction. Neuromuscular electrical stimulation (NMES) can activate motor units and improve their tropism and functionality. Therefore, this systematic review aimed to evaluate the effects of NMES on the phonation, voice quality, intelligibility, speech muscle strength, and quality of life of patients with dysarthria and dysphonia.

Methods: This systematic review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. In September 2022, we searched the EMBASE, PubMed, and Cochrane databases for studies that evaluated the effect of NMES on dysarthria and dysphonia. Randomized controlled trials, observational studies, case series, and case reports were included in the search strategy. The risk of bias in each study was assessed using the ROBINS-I tool.

Results: This review included four studies designed as case studies or non-randomized trials with a low risk of bias. NMES showed a positive effect on voice quality and phonation in patients with dysarthria and/or dysphonia. Reported improvements included restored function of the muscles responsible for laryngeal elevation, a reduction in voice breathiness and strain, and enhanced articulation.

Discussion: NMES seems to be a promising tool for the rehabilitation of patients with speech disorders. Randomized clinical trials should be conducted to confirm these benefits.

Keywords: Neuromuscular stimulation; peripheral electrical stimulation; NMES; electrical stimulation; dysarthria; voice disorders; articulation disorders; dysphonia; phonation disorder; voice disturbance

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Introduction

People with speech disorders have difficulty producing speech, which is often related to speech muscle dysfunction. Dysarthria and dysphonia, among the most prevalent speech disorders, result from various pathologies, such as vocal fold paralysis, spasmodic dysphonia, traumatic brain injury, post-surgical meningioma, meningitis, stroke, and other neurologic diseases (Sellars et al., 2005; Almeida et al., 2022). Patients with these conditions face considerable barriers in their activities and participation in social and civil life (Wray et al., 2019; Brady et al., 2011). Therefore, improving dysarthria and dysphonia is important for rehabilitating affected patients.

Neuromuscular electrical stimulation (NMES) involves intermittent peripheral nerve stimulation that activates the motor units and improves their trophism and functionality. Furthermore, it promotes modulation of the sensorimotor cortex and spinal motor neurons (Maffiuletti, 2010). Since the early 1960s, NMES has been used to aid post-stroke recovery (Ijzerman et al., 2009), and several studies have proven its benefits in patients with post-stroke dysphagia (Oh et al., 2017; Oh et al., 2020; Alamer et al., 2020; Park et al., 2016). Since the muscles involved in dysphagia and those involved in dysarthria/dysphonia show an overlap, it seems reasonable to use NMES to improve speech disorders. However, to date, there are no evidence-based recommendations for NMES implementation in dysarthria/dysphonia rehabilitation.

This systematic review makes a novel contribution to the literature by evaluating the effects of NMES on the phonation, voice quality, intelligibility, speech muscle strength, and quality of life of patients with dysarthria and dysphonia and comparing these effects with those of other speech therapies, in the short and long term.

Materials and Methods

Study design

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines were followed in this study (Page et al., 2021).

Search strategy

This is a systematic review of NMES for treating patients with dysarthria and/or dysphonia. In September 2022, we systematically searched the electronic databases EMBASE, PubMed, and Cochrane for articles published in English, Spanish, Portuguese, and German using the search terms ("dysphonia"[MeSH

Terms]" OR "Dysphonia OR 'Hyperkinetic Dysphonia' OR 'Neurologic Adductor Spastic Dysphonia' OR 'Organic Tremor Dysphonia' OR 'Phonation Disorder*' OR 'Spastic Dysphonia'") OR ("dysarthria[MeSH Terms]" OR "(Dysarthria OR 'Dysarthrosis*' OR 'Dysarthrias' OR 'Flaccid Dysarthria*' OR 'Guttural Dysarthria*' OR 'Mixed Dysarthria*' OR 'Scanning Dysarthria*' OR 'Spastic Dysarthria*") AND ("electric stimulation therapy[MeSH Terms]" OR "'Electric Stimulation Therapy' OR 'Electric* Stimulation Therapy' OR 'Electrotherapy' OR 'Interferential Current Electrotherapy' OR 'Stimulation Therapy, Electrical' OR 'Therapeutic Electric* Stimulation' OR 'NMES' OR 'Electrical stimulation*' OR 'Neuromuscular stimulation'").

Eligibility criteria

We included studies that evaluated the effects of NMES on dysarthria and dysphonia. The outcomes were changes in quantitative or qualitative measurements of phonation, voice quality, intelligibility, speech muscle strength, and quality of life (for instance: assessment of articulation, vowel distortion, hypernasality, loudness and pitch variation). We included randomized controlled trials (RCTs), observational studies, case series, and case reports, published in the previous 10 years. We excluded studies on patients with speech or language disorders other than dysarthria and dysphonia, such as aphasia and speech apraxia. Those involving transcutaneous electrical nerve stimulation or deep brain stimulation were also excluded.

Study selection

The electronic database search was conducted by one reviewer. After duplicates were removed, titles and abstracts were independently evaluated by three reviewers based on the eligibility criteria. Thereafter, eight reviewers screened the full text of each potential article. Disagreements were settled through group discussion among the eight reviewers to reach a consensus about each article.

Assessment of risk of bias in individual studies

Two reviewers independently assessed the risk of bias for each of the four papers that were finally included using the ROBINS-I tool for non-randomized trials (Sterne et al., 2016).

Data extraction and analysis

Articles found in the databases were exported to the Rayyan AI platform. We followed the PICOS

criteria for data extraction as follows: (P) Population, adult patients with dysphonia and/or dysarthria; (I) Intervention, NMES; (C) Comparison, any other treatment, including placebo; (O) Outcome, changes in quantitative or qualitative measurements of phonation, voice quality, intelligibility, speech muscle strength, and quality of life; (S) Studies, RCTs, observational studies, case series and case reports.

Results

The systematic search yielded 542 articles, with 81 duplicated studies. After screening the titles and abstracts, 15 papers remained. After reading the entire papers, we excluded 11 articles for the following reasons: patients with diagnoses other than dysarthria/dysphonia ($n=5$), interventions other than NMES ($n=1$), publication date outside the stipulated time frame ($n=2$), language other than those stipulated ($n=1$), and conference abstracts ($n=2$). Finally, four articles were included in this review (Figure 1). The details of the included studies are shown in Table 1. All the included studies were classified as having a low risk of bias according to the ROBINS-I guidelines (Sterne et al., 2016).

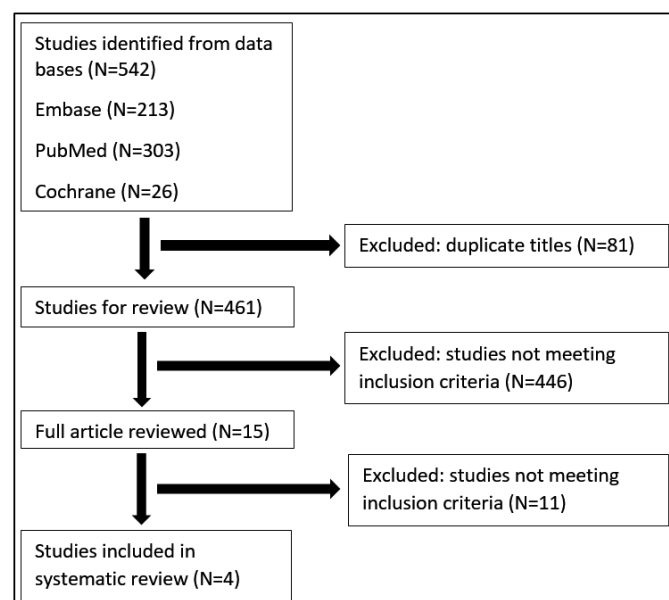


Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flow diagram

Effect of NMES on voice quality

Ko et al. (2016) examined the impact of laryngopharyngeal NMES therapy in patients with stroke or traumatic brain injury who had speech disorder and dysphagia. Eighteen patients were evaluated, of whom

only six received conventional swallowing training and twelve received this training plus NMES. The authors reported an improvement in vocal fold vibration/tension and restoration in the function of the muscles responsible for laryngeal elevation.

In a retrospective case study, Guzman et al. (2014) reported positive effects of NMES therapy in addition to traditional vocal exercises on the voice quality of two female patients with superior laryngeal nerve weakness. With the addition of NMES, a significant improvement in voice breaking and vocal range, a decrease in voice breathiness, and an increase in voice resonance could be attained.

Safi et al. (2018) included two patients with mildly to moderately dysarthric speech after a cerebrovascular accident in a study with a single-subject design that involved a combination of oral motor exercise with NMES. Submental NMES showed no effect on lingual strength, but facial NMES led to an improvement of labial strength in both patients; this can ultimately contribute to an improvement in voice quality, considering the complexity and involvement of different muscles for speech production.

Furthermore, NMES showed promising results in one patient with severe dysarthria. Berenati et al. (2021) describe the strengthening of muscle contractions and improvement in vocal strain in an 18-year-old with severe speech impairment after post-anoxic brain injury.

Effects of NMES on phonation

Ko et al. (2016) report positive effects on phonation after 2 and 4 weeks of NMES therapy in patients with stroke. Additionally, Guzman et al. (2014) describe an improvement in soft phonation index values after 17 and 8 sessions with NMES, respectively.

Furthermore, as reported by Berenati et al. (2021), NMES can improve articulation and hypernasality, contributing to improved phonation. The patient presented in the case study by Berenati et al. (2021) was unable to perform conventional speech therapy due to his impaired cognitive abilities. Consequently, his dysarthria severity was reduced solely through NMES therapy. These findings are especially remarkable in light of the reported increased amplitude of the patient's trigeminal-facial reflex responses, which can be attributed to the induction of neuroplastic changes in the sensorimotor areas responsible for articulatory functions.

Table 1. Included studies.

Author (Year)	Study Design	Objective	Outcome Measurements	Changes in the Outcome	Population	Intervention	Comparison
Berenati et al. (2021)	Case study	Investigate the role of NMES in improving severe dysarthria	Evaluation of the a) Aachen Aphasia Test, b) Aphasia Quotient, c) Robertson Dysarthria Profile	Patient, changes after treatment: a) spontaneous speech: 2 points increased, auditory comprehension: 1 point increased, repetition: 1 point increased, naming: 1 point increased b) 8 points increased c) 25 points increased	One 18-year-old patient with severe dysarthria following post anoxic brain injury	Conventional rehabilitation alone, then NMES alone for 4 weeks (two daily sessions of 30 mins, six days a week); NMES parameter: stimulation chronaxie 300/260 ms	No comparison group
Guzman et al. (2014)	Retrospective case study	Demonstrate the effectiveness of NMES combined with traditional voice therapy for dysphonia after superior laryngeal nerve SLN weakness	Evaluation of the a) SPI, b) NHR, c) jitter, d) shimmer, e) peak-to-peak amplitude variation, f) fundamental frequency variation	Patient 1, changes after treatment: a) 5.32 points decreased b) 0.01 points increased c) 0.17 % decreased d) 1.33 % increased e) 7 % decreased f) 0.09 % increased Patient 2, changes after treatment: a) 10.65 points decreased b) 0.05 points decreased c) 0.24 % decreased d) 4.2 % decreased e) 5.45 % decreased f) 1.48 % decreased	Two female patients, 48 and 63 years old, with dysphonia secondary to SLN weakness	Traditional voice therapy, then NMES for 17 and 8 sessions, respectively; NMES parameter: biphasic wave and pulse duration 80 Hz and 700 µs, maximum intensity of stimulation 12 and 13 mV, respectively	No comparison group
Ko et al. (2016)	Non-randomized trial	Investigate the effect of NMES on dysphonia in patients with dysphagia post-stroke or traumatic brain injury	Evaluation of the a) perceptual score (GRBAS), b) acoustic parameters (jitter, shimmer, NHR, SPT), c) aerodynamic score (SPL), d) swallowing functions (PAS, FDS, ASHA NOMS)	Intervention group, changes after 2 and 4 weeks of treatment: a) improvement from baseline to 2 and to 4 weeks b) improvement in jitter from baseline to 2 and to 4 weeks, no statistically significant differences in other parameters c) aerodynamic score in other parameters d) improvement from baseline to 2 weeks and to 4 weeks e) improvements over time like comparison group Comparison group, changes after 2 and 4 weeks of treatment: a) no improvement b) no improvement in jitter over time, no statistically significant differences in other parameters c) improving trend, but no significant improvement over time d) improvements over time like intervention group	18 participants, NMES group (n=12) and group receiving conventional swallowing training CST (n=6)	NMES (one daily session of 60 mins, five days a week) combined with CST for 2 weeks, then CST without NMES for 2 weeks; NMES parameter: biphasic wave and pulse duration 80 Hz and 300 µs	CST for 4 weeks
Saf et al. (2018)	Single-subject ABC/ACB design	Evaluate the effect of facial and submental NMES in patients with lingual, labial and buccal weakness	Evaluation of the a) labial muscular strength, b) lingual muscular strength with the Iowa Oral Performance Instrument	Patient 1, changes after treatment: a) phase A: 1.67 kPa decreased, phase B: 19.33 kPa increased, phase C: 2.67 kPa increased b) phase A: 1 kPa increased, phase B: 4.67 kPa increased, phase C: 7 kPa increased Patient 2, changes after treatment: a) phase A: 0.67 kPa decreased, phase B: 15.67 kPa increased, phase C: 7.67 kPa increased b) phase A: 1 kPa increased, phase B: 3.67 kPa increased, phase C: 2.67 kPa increased	Two male patients, 62 and 58 years old, with dysarthria after cerebrovascular accident	Phase A: baseline measurement Phase B: NMES (one daily session of 30 mins, three times a week) and oral motor exercise for 4 weeks Phase C: oral motor exercise only for 4 weeks; NMES parameter: 50 µs phase duration, frequency of 30 Hz	No comparison group

Abbreviations. ASHA NOMS American speech-language-hearing association national outcome measurement system swallowing scale; CST, conventional speech therapy; FDS functional dysphagia scale; GRBAS, grade, roughness, breathiness, asthenia, strain scale; NHR noise-to-harmonic ratio; NMES neuromuscular electrical stimulation; PAS penetration-aspiration scale; SPL, superior laryngeal nerve; SPT soft phonation index; SPL sound pressure level.

Effect of NMES on quality of life

Dysarthria and dysphonia impact patients' social life and well-being, and affect patients face restrictions in daily activities that require the use of their voice. In addition to the impact on patients' communicative quality of life, other aspects, such as psychological or social domains, may be impacted (Ma & Yiu, 2001). Despite this, Guzman et al. (2014), Ko et al. (2016), and Safi et al. (2018) acknowledge that this patient group can have a significantly decreased quality of life; however, none of the studies included in this review investigated this knowledge gap further.

Discussion

Laryngeal muscle weakness can result in vocal fold instability. Acoustic evaluations reveal that patients with stroke-related dysarthria have significant differences in frequency, jitter, shimmer, and noise-to-harmonic ratio compared with those of neurologically healthy controls (Wang et al., 2010). As a result, treatments that strengthen these muscles seem to be promising for improving speech disorders.

Laryngopharyngeal NMES can restore the function of muscles controlling vocal fold vibration, which can strengthen the muscles attached to the hyoid bone (namely, the mylohyoid and thyrohyoid muscles) and increase laryngeal elevation. Consequently, excessive quavering during speech can be reduced. Furthermore, NMES can improve the sound pressure level by strengthening vocal ligament and thyroarytenoid muscle tension, thereby increasing glottal resistance (McHenry & Reich, 1985).

Despite the possible benefits, we would like to highlight some limitations of the present studies. None of them was a RCT, thus the patients were not blinded, and the sample size was small. The maximum duration of NMES treatment was 4 weeks and maybe this might be too short to generate a significant and lasting effect (especially if we consider that an improvement in muscle function or neuromodulation is proportional to the frequency of the stimuli). In addition, 3 of the 4 studies did not include a comparison group.

Conclusions

In conclusion, NMES may be an effective and promising tool for the rehabilitation of patients with speech disorders. The limited number of studies regarding its use for treating dysphonia and dysarthria in adults did not allow us to perform a meta-analysis and obtain statistically significant results supporting the

benefit of the use of electrical stimulation in these patients.

To the best of our knowledge, no RCTs investigated this topic up to date. Nevertheless, to confirm the benefits of NMES in the treatment of dysphonia and dysarthria and to further explore the findings presented in this systematic review, conducting an RCT would be highly beneficial.

None of the present studies has evaluated the patients in the medium to long term, and thus the duration of the effects of NMES is not yet clear. We suggest this assessment in future studies. Furthermore, we encourage the evaluation of quality of life to highlight the benefits of NMES therapy. This could broaden the understanding of the effects of NMES on dysphonia and dysarthria and its impact on the patient's social life and well-being.

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Conflicts of Interest: Cristina Stephan is a medical affairs consultant for different pharmaceutical companies. All other authors declare no conflict of interest.

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