Speech-Language Therapy for Treatment of Dysphagia and Dysphonia in Systemic Granulomatous Diseases. Granulomatous Diseases: Dysphagia and Dysphonia Treatments

Terapia fonoaudiológica para tratamento da disfagia e disfonia nas doenças granulomatosas sistêmicas.
Doenças granulomatosas: tratamento da disfagia e disfonia

Terapia del habla para la disfagia y disfonía tratamiento la enfermedades granulomatosas sistémicas.
Enfermedades granulomatosas: el tratamiento de la disfagia y disfonía

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The granulomatous lesions are frequently founded in infectious diseases and can involve the larynx and pharynx and can cause varying degrees of dysphonia and dysphagia. There is still no systematic review that analyzes effectiveness of speech therapy in systemic granulomatous diseases. Research strategy: A systematic review was performed according to Cochrane guideline considering the inclusion of RCTs and quasi-RCTs about the effectiveness of speech-language therapy to treat dysphagia and dysphonia symptoms in systemic granulomatous diseases of the larynx and pharynx. Selection criteria: The outcome planned to be measured in this review were: swallowing impairment, frequency of chest infections and voice and swallowing symptoms. Data analysis: We identified 1,140 citations from all electronic databases. After an initial shift we only selected 9 titles to be retrieved in full-text. After full reading, there was no RCT found in this review and therefore, we only described the existing 2 case series studies. Results: There were no randomized controlled trials found in the literature. Therefore, two studies were selected to be included only for narratively analysis as they were case series. Conclusion: There is no evidence from high quality studies about the effectiveness of speech-language therapy in patients with granulomatous diseases of the larynx and pharynx. The investigators could rely in the outcomes suggested in this review to design their own clinical trials.

Keywords: Language and Hearing Sciences; Pharynx; Laringe; Deglutition.

INTRODUCTION

Systemic granulomatous diseases are characterized by the presence of ulcerated lesions, vegetating and granuloma, which is a result of process of protecting and healing[1,2]. The granulomas can be classified into two types: non-specific (eosinophilic granuloma, lethal midline granuloma, Wegener) and

specific (tuberculosis, leprosy, syphilis, systemic mycosis and leishmaniosis). Head and neck signs and symptoms are common in patients with leishmaniasis and paracoccidioidomycosis. Nasal manifestations prevail in leishmaniasis and oropharyngeal ones in paracoccidioidomycosis[3].

Some authors described dysphonia as the main symptom of laryngeal tuberculosis, being present in 96.6% of cases[4]. It is possible to find diffuse injuries in granulomatous lesions of the larynx and pharynx, which can involve the whole larynx or even cause more restricted injuries in the anterior or posterior commissure, vocal folds, vestibular folds, arytenoids, epiglottis and infraglottic[5,6].

Any injury located in the upper airways may cause odynophagia and dysphagia symptoms, leading to emaciation, malnutrition and worsening in the patient's general condition[7-10].

In systemic granulomatous diseases, many patients report difficulty in swallowing[6,11] however, the symptoms of swallowing disorders, such as difficulty in initiating swallowing, nasal reflux, cough during or after swallowing and sensation of food stuck in the throat, if unrecognized and untreated, can lead patients to conditions of malnutrition, dehydration and respiratory complications. Aspiration is a symptom of dysphagia with fatal consequences, since the passage of food and/or liquids into the airway increases the risk of pneumonia[12,13] and, consequently, the morbidity and mortality.

Treatment of dysphagia includes active exercise and other strategies, including compensations designed to improve safety of the swallowing and efficiency of surgical procedures, medications, and dental prosthetic devices. Treatment of dysphagia often takes two parallel courses: compensations to allow patients to eat at least some food *per so* without aspirating and exercises to improve strength and coordination, for the patient to return to full oral intake[10,14,15].

Specifically in the larynx, on the vocal folds, granulomatous lesions can interfere in the voice quality, presenting several degrees of dysphonia or even aphonia. Voice disorders such as hoarseness, increased fatigue and decreased phonatory control[16], commonly found after the scarring of the lesions, what results in functional impairment of the whole structure.

Physicians have several choices for managing hoarseness including observation, medical therapy, surgical therapy, voice therapy, or a combination of these approaches. Voice therapy is effective for treatment of hoarseness and may be applied across the lifespan from children to older adults[17-19].

OBJECTIVE

To evaluate the effectiveness of speech-language therapy in the treatment of dysphonia and/or dysphagia of systemic granulomatous diseases of the larynx and pharynx.

MATERIAL AND METHODS

SEARCH STRATEGY

A systematic review was performed according to Cochrane guideline and the search strategy was run in the main electronic databases: Medline (1966 to May 2012), Embase (1980 to May 2012) Lilacs (1982 to May 2012), Register of the Cochrane Controlled Trials (the Cochrane Library, 2012). There were no language restrictions. The terms and synonyms used in the search strategy are presented in Figure 1.

SELECTION CRITERIA

The intervention of interest was speech-language and voice therapy which could include any vocal and/or dysphagia exercises (e.g. vocal function exercises, resonant voice therapy; and swallow maneuvers, respectively) and recommendations (e.g. to eliminate the vocal abuse or misuse, and dietary modification and postural changes, respectively). The control group could be no intervention, sham procedures, medical/pharmacological treatment and another. The following primary outcomes planned to be considered was: swallowing impairment, frequency of chest infections and patient-reported measures of voice handicap, voice symptoms or voice-related quality of life and swallowing symptoms.

((Granulomatous Disease) **OR** (Granulomatous Diseases) **OR** (Respiratory Tract Granulomas) **OR** (Respiratory Tract Granuloma) **OR** (Leprosies) **OR** (Hansen Disease) **OR** (Hansens Disease) **OR** (Tuberculoses) **OR** (Kochs Disease) **OR** (Kochs Disease) **OR** (Koch Disease) **OR** Syphilis **OR** (Great Pox) **OR** (Histoplasmoses) **OR** (Paracoccidioidomycoses) **OR** (South American Blastomycosis) **OR** (Blastomycoses) **OR** (North American Blastomycosis) **OR** (Gilchrist Disease) **OR** (Gilchrists Disease) **OR** (Gilchrists Disease) **OR** (Leishmaniases) **OR** (Mucocutaneous Leishmaniases) **OR** (Mucocutaneous Leishmaniasis) **OR** (Sarcoidoses) **OR** (Besnier Boeck Schaumann Syndrome) **OR** (Besnier Boeck Schaumann Syndrome) **OR** (Boeck Disease) **OR** (Schaumanns Syndrome) **OR** (Schaumanns Syndromes) **OR** (Boecks Sarcoid) **OR** (Boeck Sarcoid) **OR** (Boecks Sarcoid) **OR** (Schaumann Disease) **OR** (Schaumann Syndrome) **OR** (Besnier Boeck Disease) **OR** (Besnier Boeck Disease) **OR** (Boecks Disease) **OR** (Boecks Disease)) **AND** ((Speech pathology) **OR** (Speech pathologies) OR (Speech Therapy) **OR** (Speech Therapies) **OR** (Voice Training) **OR** (Voice Trainings) **OR** (voice therapy) **OR** (voice therapies) **OR** (vocal treatment) **OR** (vocal treatments) **OR** (voice treatment) **OR** (voice treatments) **OR** (vocal rehabilitation) **OR** (vocal rehabilitations) **OR** (voice rehabilitation) **OR** (voice rehabilitations) **OR** (speech language pathology) **OR** (speech language pathology) **OR** (speech language pathologies) **OR** (speech language pathologies) **OR** (speech language therapy) **OR** (speech language therapy) **OR** (speech language therapies) **OR** (speech language therapies))

Figure 1- Search Strategy

DATA ANALYSIS

We planned to consider randomized controlled trials (RCTs) and quasi-RCTs (RCTs in which allocation to treatment was obtained by alternation, use of alternate medical records, or other predictable methods) evaluating adults diagnosed with granulomatous lesions of the larynx and pharynx. Two authors (AB and RED) independently screened the trials identified by the literature search. We resolved any disagreements by consulting with the other author and consult with her for quality assurance of the processes.

RESULTS AND DISCUSSION

The search identified 1,140 citations from all electronic databases. After an initial shift we only selected 9 titles to be retrieved in full-text. After full reading, there was no RCT found in this review and therefore, we only described the existing 2 case series studies[20,21] that came out is only in the table contemplated therapeutic outcome in relation to the voice, other symptoms and complaints pertaining to swallowing. The remaining seven studies did not fulfill the inclusion criteria as they were off-topic, reviews and case report (Figure 2).

Yelken et al. [20] analyzed the effects of anti-tuberculosis treatment on the voice quality of laryngeal tuberculosis in 14 patients. The patients were evaluated by patient self-assessment, perceptual analysis and acoustic analysis, before and after treatment. The mean voice handicap index-10 score decreased (11.5; p=0,023), acoustic analytical parameters and perceptual analysis have improvement (p=0.017) after treatment, i.e., median fundamental frequency (p=0.018), jitter and shimmer (p=0.018) and improved in harmonic to noise ratio (p=0.018). The authors concluded that anti-tuberculosis treatment clearly improved the voice outcomes of laryngeal tuberculosis.

Furthermore e Ruas et al. [21] evaluated dysphonia in patients treated for laryngeal tuberculosis, and assessed the effect of speech therapy on patients' vocal quality. Seven of 23 patients with a confirmed diagnosis of laryngeal tuberculosis were treated with speech therapy for six months. After the intervention, dysphonic patients had better vocal quality (15.8%), as demonstrated by statistical analysis of jitter (p =0.017), shimmer (p=0.025), fundamental frequency variability (p=0.032), maximum phonation time (p=0.000) and the ratio between maximum phonation time for voiceless and voiced fricative sounds (p=0.008). The authors concluded that speech therapy seems to improve patients' vocal quality.

Hoarseness is often caused by benign or self-limited conditions, but may also be the presenting symptom of a more serious or progressive condition requiring prompt diagnosis and management. Several approaches to voice therapy for treating hoarseness have been identified in the literature[22-24].

Yelken et al. [20] concerning that after recovery from laryngeal tuberculosis, one would expect the improvement of the voice as the laryngeal inflammatory

process had been treated; however, they could find no reports addressing the evolution of dysphonia after laryngeal tuberculosis treatment. The disappearance of the oedema and tumoural masses returned to a symmetric and periodic vibratory movement of the vocal fold mucosa. As a result perceptual voice quality parameters improved. However, the authors stated that permanent breathy voice might have resulted from irreversible fibrotic changes in the lamina propria after healing of the infection.

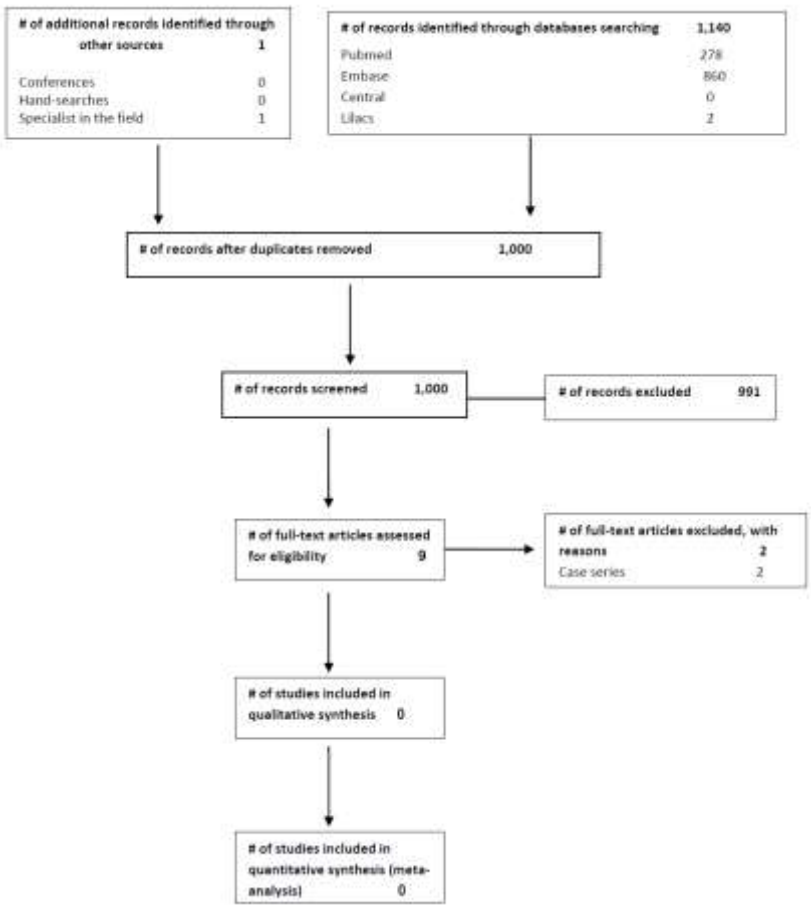

Figure 2- Flowchart of the results about treatment of the dysphonia and dysphagia in granulomatous lesions studies

In both studies selected, the laryngeal pathology was caused by laryngeal tuberculosis. Inflammation of the mucosa and granuloma formation eventually leads to necrosis of the overlying epithelium, which sloughs and ulcerates[20]. The cicatricial tissue observed is made up of dense collagen fibers which are more rigid than the normal vocal fold mucosa. The location of the scar may vary, affecting one or both vocal folds, and may create asymmetry. After speech-language therapy, all patients included in Ruas et al. [21] study, had complete glottal closure, except for one who had an associated vocal fold injury still under investigation.

Although the results positives after speech-language therapy in these two studies, the methodology does not guaranties the effectiveness of intervention in voice quality in patients with granulomatous lesions. It is necessary a more robust design comprising a control group or comparison of speech-language therapy with another therapeutic intervention. Moreover, it is unclear the action of drugs used in patients, with a confounding factor between therapeutic action and drug action on the laryngeal anatomy and physiology.

Still following the objective of this review, the laryngeal mechanisms impaired swallowing in patients with systemic granulomatous diseases already described in several studies[5,11,25,26] where parameters such as odynophagia, dysphagia and cough are present, however, none of these had methodological status to enter the inclusion or exclusion of this review.

Research on treatment of oropharyngeal dysphagia has supported several treatment approaches. Treatment can include postural changes, heightening pre-swallow sensory input, voluntary swallow maneuvers, and exercises. Evidence to support the efficacy of these procedures is variable[10,15].

The efficacy of individual postures, maneuvers, and other therapy procedures reported in the literature has been noted, but although there is considerable evidence that these interventions work with head and neck cancer patients, there are still many questions concerning the relative contributions of the various therapy techniques to improve swallow function, the optimal frequency, timing, and intensity of swallow rehabilitation programs and the impact of patient practice and feedback strategies. Furthermore, randomized clinical trials are considered the gold standard for the evaluation of treatment efficacy studies[27].

A number of randomized clinical trials have been instituted in the past few years that address some of these issues and may demonstrate the superiority of various therapy procedures in, e.g., treated head and neck cancer patients[14], but since this revision is possible

to verify the absence of such methodological models in studies that include speech and hearing therapy in patients with dysphonia and dysphagia resulting from granulomatous diseases.

The authors of this review were very critical about the search strategy covering all databases and ensuring the independence of the selection of studies. These aspects are the strengths of this study. Although with limitations of the evidence found, this does not detract from this review, but draws attention to the urgent need to have systematic clinical studies in the literature.

CONCLUSION

There is no evidence from high quality studies about the effectiveness of speech-language therapy to treat dysphagia and dysphonia symptoms in systemic granulomatous diseases of the larynx and pharynx. Clinical trials need to be conducted. The investigators could rely in the outcomes suggested in this review to design their own clinical trials.

RESUMO

As lesões granulomatosas são freqüentemente encontradas em doenças infecciosas, podendo envolver a laringe e a faringe causando vários graus de disfonia e disfagia. Ainda não há revisão sistemática que analisa a eficácia da terapia fonoaudiológica nas doenças granulomatosas sistêmicas. Estratégia de pesquisa: Uma revisão sistemática foi realizada de acordo com a base Cochrane considerando a inclusão de ECRs e quase-ECRs sobre a eficácia do tratamento fonoaudiológico para tratar os sintomas da disfagia e disfonia em doenças granulomatosas sistêmicas da laringe e faringe. Critérios de seleção: Os desfechos considerados para esta revisão foram: dificuldades na deglutição, freqüência de infecções pulmonares e sintomas na voz e na deglutição. Análise dos dados: Foram identificados 1.140 citações de todos os bancos de dados eletrônicos. Após análise inicial, foram selecionados apenas 9 títulos a serem recuperados em texto completo; destes, não foram encontrados ECR. Nesta análise, portanto, só descrevemos dois estudos de série de casos. Resultados: Não foram encontrados ensaios clínicos randomizados na literatura. Portanto, dois estudos foram selecionados e incluídos apenas para análise narrativa. Conclusão: Não há evidências de estudos de alta qualidade sobre a eficácia da terapia fonoaudiológica em pacientes com doenças granulomatosas da laringe e faringe. Os investigadores podem confiar nos resultados sugeridos nesta revisão para desenhar seus próprios ensaios clínicos.

Palavras chave: Fonoaudiologia; Faringe; Laringe; Deglutição.

RESUMEN

Las lesiones granulomatosas se encuentran a menudo en enfermedades infecciosas, y puede implicar la laringe y la faringe causando diversos grados de disfonía y disfagia. Ninguna revisión sistemática examina la eficacia de la terapia del habla en las enfermedades granulomatosas sistémicas. Estrategia de investigación: una revisión sistemática se realizó de acuerdo a la base Cochrane considerando la inclusión de los ECA y ensayos controlados cuasialeatorios sobre la eficacia de la terapia del habla para tratar los síntomas de disfagia y disfonía en las enfermedades granulomatosas de la laringe y la faringe. Criterios de selección: Los resultados considerados para esta revisión fueron: dificultad para tragar, la frecuencia de infecciones en los pulmones y los síntomas en la voz y la deglución. El análisis de datos: Se identificaron 1140 citas de todas las bases electrónicas. Tras el análisis inicial, se seleccionaron sólo 9 títulos a ser recuperados a texto completo y de éstos, no hay ECA. En este análisis, por lo tanto, sólo dos estudios describieron series de casos. Resultados: No hubo ensayos controlados aleatorios en la literatura. Por lo tanto, dos estudios fueron seleccionados para análisis e incluía sólo narrativa. Conclusión: No existe evidencia de estudios de alta calidad sobre la eficacia de la terapia del lenguaje en pacientes con enfermedades granulomatosas de la laringe y la faringe. Los investigadores pueden confiar en los resultados propuestos en esta revisión para sacar sus propios ensayos clínicos.

Palabras clave: Fonoaudiología; Faringe; Laringe; Deglución.

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