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Effects of electrostimulation associated with masticatory training in individuals with down syndrome

Efeitos da eletroestimulação associada ao treino mastigatório em pessoas com síndrome de down

ABSTRACT

Purpose: Investigate and measure the effects of electrostimulation on the orofacial musculature and on the chewing, breathing and swallowing functions of individuals with Down syndrome. **Methods:** Study participants were 16 individuals with Down syndrome (six males and 10 females) from an institutional extension project aged nine to 25 years. Speech-language pathology assessment was performed using the protocol of Orofacial Myofunctional Evaluation with Scores (OMES) pre- and post-intervention. This protocol comprised eight weekly electrostimulation sessions. Functional Electrical Stimulation (FES) current was used at a frequency of 10Hz in warm-up and 30Hz in application, intermittent stimulation (cycling pulses) with ON-time of 5s and OFF-time of 10s common to both stages, and pulse width of 200µs in warm-up and 250µs in application. **Results:** Significant differences were observed between pre- and post-application of FES regarding cheek appearance (flaccidity and arching), tongue mobility (right and left laterality), and musculature behavior during performance of functions of the stomatognathic system: respiration, deglutition (lip behavior), and mastication (bite and trituration). **Conclusion:** Effects of electrostimulation associated with masticatory training of the masseter muscles were statistically identified, with functional gains in chewing, breathing and swallowing performance in individuals with Down syndrome.

RESUMO

Objetivo: Investigar e mensurar os efeitos da eletroestimulação na musculatura orofacial e nas funções de mastigação, respiração e deglutição dos indivíduos com síndrome de Down. **Método:** Participaram da pesquisa 16 indivíduos com Síndrome de Down, sendo seis do gênero masculino e dez do gênero feminino com idade entre 9 e 25 anos, participantes de um projeto de extensão institucional. Foram realizadas avaliações fonoaudiológicas com uso do protocolo AMIOFE antes e após a intervenção, que consistiu em oito sessões de eletroestimulação semanais. A corrente utilizada foi a *Functional Electrical Stimulation* (FES), com uma frequência de 10Hz no aquecimento e 30 Hz na aplicação, em um tempo ON de 5s e OFF de 10s comuns nas duas etapas, e com a largura de pulso de 200(µs) no aquecimento e 250(µs) na aplicação. **Resultados:** Observaram-se diferenças significativas após aplicação da eletroestimulação (FES) em relação ao aspecto das bochechas quando comparadas flacidez/arqueamento pré e pós o estímulo elétrico, diferenças na mobilidade de língua (lateralidade direita e esquerda), no comportamento da musculatura na execução das funções estomatognáticas de respiração, melhoria no comportamento dos lábios durante a deglutição e mudanças expressivas no processo de mastigação (mordida e trituração). **Conclusão:** Foi identificado estatisticamente que houve efeito após a eletroestimulação associada ao treino mastigatório nos músculos masseteres, com ganhos funcionais na execução da mastigação, respiração e deglutição, em pessoas com Síndrome de Down.

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INTRODUCTION

Down syndrome (DS) is a genetic condition caused by the presence of an extra chromosome, most commonly Trisomy 21, originally described by John Langdon Haydon Down in 1866^(1,2).

One of the characteristics most commonly found in individuals with DS is the presence of generalized muscular hypotonia, which directly affects the stomatognathic system (SS). This hypotonia, which emanates from the central nervous system, affects muscles and ligaments, interfering negatively in this system⁽³⁾. In addition to hypotonia, other stomatognathic changes can be observed, such as adapted swallowing and speech impairments due to articulatory imprecision. Individuals with DS present maxillary atresia, which results in reduction of the oral cavity and oval palate and imbalance of forces between the oral and facial muscles, causing alteration of the dental arch, triggering possible open bites in addition to presence of oral breathing and mandibular retraction⁽⁴⁻⁷⁾.

Based on physiological concepts that provoke excitability in the nerves and muscle fibers, electrostimulation is a non-invasive method with no systemic implications that does not cause dependence and has no undesirable side effects. It is a possibility of intervention in muscular activity indicated to strengthen the musculature⁽⁸⁾. Aiming to increase the number of application possibilities of technological resources combined with the conventional technique, Speech-language Pathology (SLP) can favor and reveal satisfactory results in the treatments.

Neuromuscular electrical stimulation produces contractions in the skeletal musculature through application of electrical impulses with no cerebral involvement⁽⁹⁾. It has been used in rehabilitation, especially in muscle atrophies or to increase muscle strength, for many years⁽¹⁰⁾. The muscle strength gain resulting from short-duration electrostimulation is similar to that obtained with voluntary training⁽¹¹⁾.

However, the effects of electrostimulation are grounded on different theoretical bases, such as the assumption that direct stimulation of the motor nerves causes the muscles to perform rhythmic contractions^(12,13). In this context, electrostimulation can contribute to the treatment of individuals with DS considering the hypothesis that the presence of hypotonia in these individuals causes muscles to perform slower and/or ineffective contractions, thus leading to shorter response time.

Research evidences that electrostimulation can benefit rehabilitation of neuromotor cases; however, the literature still lacks evidence on these investigations⁽¹⁴⁾.

The present study aims to investigate and measure the effects of electrostimulation on the orofacial musculature and on the chewing, breathing and swallowing functions of individuals with DS.

METHODS

This is a longitudinal, quantitative, applied, field research that is considered an intervention study because the effects of electrostimulation were analyzed pre- and post-therapeutic sessions in individuals with Down syndrome (DS). Study participants were 16 individuals with DS (six males and 10 females) aged

nine to 25 years from an institutional extension project specific for assistance to individuals with DS. Inclusion criteria were as follows: individuals should be undergoing speech-language therapy at the health service and agree to participate in the study. Individuals with cardiopathies and/or non-collaborative were excluded from the survey, considering that electrostimulation application would be hindered. It is important to emphasize that during data collection participants were submitted only to the electrostimulation proposed in the study.

Procedures performed during the speech-language pathology (SLP) assessment were divided into two stages: application of the protocol of Orofacial Myofunctional Evaluation with Scores (OMES), which was performed by trained therapists pre- and post-intervention, observing appearance and postural position of the lips, cheeks and tongue, oral cavity (aspect of the hard palate), and mobility of the lips, cheeks and tongue; evaluation of functions of the stomatognathic system: respiration, deglutition and mastication, with the latter assessed using stuffed cookies (“Bono”) (food indicated in the Brazilian validation process of the OMES protocol)⁽¹⁵⁾. The medians were calculated based on the protocol scores, with higher scores indicating normality and lower scores showing impairment.

Ten sessions were conducted: an initial evaluation and a final reevaluation using the OMES protocol to compare the performance gains of stomatognathic structures and functions pre- and post-Functional Electrical Stimulation (FES), and eight electrostimulation sessions lasting 15 minutes each, with the initial five minutes used for muscle warm-up and the last 10 minutes for application. Of these, five minutes were reserved for masticatory training (with stuffed cookie - “Bono”), always instructing the volunteers to chew bilaterally and alternately and, whenever possible, with the lips sealed.

Electrostimulation procedures were conducted using a Neurodyn II (Ibramed manufactured) intended for current therapies, namely, Transcutaneous Electrical Nerve Stimulation (TENS), which is a technique based on programs that can generate analgesia, Russian Current for high-frequency muscle resistance, and Functional Electrical Stimulation (FES), which was the current used in the present study. FES is applied through electrical impulses intended to produce contractions by pulse trains in muscle groups that will trigger movements and activities of daily living. It stimulates motor nerves by generating nerve impulses that activate nerve fibers⁽⁸⁾.

The equipment features four output channels with independent intensity control and was configured for two phases. During warm-up, a 10 Hz frequency was used with ON-time of 5 sec and OFF-time of 10 sec, this phase was conducted to ensure that contraction capacity of the muscles continued to respond to electrical stimuli, avoiding fatigue, and then completing a 15 sec cycle. Rise ramps last 3 sec and decay ramps last 2 sec, and pulse width (PW) of 200 μ s (time span from the beginning of the first phase of a pulse to the end of the last phase). Pulse width is generally expressed in microseconds (μ s) and the wider the pulse width, the longer the stimulus time span. During electrostimulation application associated with masticatory training of masseter muscles, the PW parameters were adjusted, increased to 250 μ s, and the frequency used was

increased to 30 Hz, because it is important to minimize muscle fatigue, and the literature suggests modulation frequencies between 30 and 40 Hz^(8,16) (Chart 1).

In this study, the electrodes used were self-adhesive and were placed on the skin according to the size of the muscle region chosen to be investigated. Skin hygiene was performed using 70% alcohol before placing the electrodes, which were then applied to the masseter muscles (superficial bundles) positioned at the origins and inserts on both sides of the face. Intensity was selected according to the level of comfort that the individuals reported at the moment of electric stimulus application. It is worth noting that no myofunctional exercises were assigned for home, and that the participants were not undergoing orofacial motor care at other sectors of the health service.

All participants signed an Informed Consent Form (ICF) prior to study commencement. All other ethical aspects were respected throughout the study procedures, following the precepts of the project approved by the Human Research Ethics Committee based on Resolution no. 466/12 of the National Health Council under protocol no. CAAE 57519316.4.0000.5188.

Data were classified and allocated in a digital spreadsheet. The variables were submitted to descriptive and inferential analysis using the Wilcoxon test for related samples. The R 2.11.0 statistical software was used at significance level of 5%.

RESULTS

Volunteers to this study presented mean age of 15.44 (SD±3.85). Gender distribution was as follows: 62.5% (n=10) female and 37.5% (n=6) male, but no gender differences were analyzed. Statistically significant change was observed regarding postural position of the lips ($p=0.014$). Prior to intervention, the volunteers kept their lips parted (see median: 2.00), whereas after

intervention, they presented sealed lips, assuming a favorable posture (see median: 3.00). The other labial variables did not present statistically significant difference (Table 1).

Statistically significant change was found in appearance of the cheeks ($p=0.002$). In some cases, flaccidity and arching was observed at pre-intervention (see median: 2.00), which improved at post-intervention and was classified as typical (see median: 3.00) (Table 2). With respect to tongue mobility data, it was possible to observe statistically significant effects of electrostimulation, because pre- and post-intervention difference was found regarding tongue lateralization, both to the right ($p=0.049$) and the left ($p=0.049$) sides. Initially, some individuals did not have the ability to lateralize (see median 1.00), but after electrostimulation this ability was retrieved or installed, evolving to normal state (see median 3.00) (Table 3).

Regarding muscular behavior in the performance of functions of the stomatognathic system, the results indicated improved breathing ($p=0.025$). Cases of mild oronasal breathing (see median: 2.00) may have evolved to typical nasal breathing (see median: 3.00). As for deglutition, improvement in behavior of the lips during swallowing ($p=0.033$) was observed. Some volunteers who initially were able to keep their lips sealed only with muscle tension (see median: 2.00), after FES application, were able to seal the lips normally (see median: 3.00); tension of the facial muscles during swallowing also improved, considering that it was initially present and became absent at post-intervention (Table 4).

With regard to the chewing function, bite and trituration were aspects that presented significant difference at the pre- and post-intervention comparison. Bite ($p=0.043$) sought to be performed with the incisors (see change in the median from 2.00 to 3.00) and trituration ($p=0.035$) sought the alternating bilateral normality pattern (median: 4), as shown in Table 4.

Chart 1. Description of the FES stages

Warm-up	Application
Frequency – 10 Hz	Frequency – 30 Hz
ON-time – 5 sec	ON-time – 5 sec
OFF-time – 10 sec	OFF-time – 10 sec
Pulse width – 200 (µs)	Pulse width – 250 (µs)
Rise (ramp up time) – 3 sec	Rise (ramp up time) – 3 sec
Decay (ramp down time) – 2 sec	Decay (ramp down time) – 2 sec
Total time – 5 min	Total time – 10 min

FES = Functional Electrical Stimulation

Table 1. Comparison of appearance, postural position, and mobility of the lips pre- and post-FES associated with masticatory training of masseter muscles in individuals with Down syndrome

Variable	Median		p-value
	Pre	Post	
Postural position	2.00	3.00	0.014*
Protrusion	3.00	3.00	1.00
Retraction	3.00	3.00	1.00
Right laterality	1.00	1.00	0.890
Left laterality	1.00	1.00	0.581

*significant $p<0.05$; Wilcoxon test; N=16; FES = Functional Electrical Stimulation

Source: João Pessoa, 2016

Table 2. Comparison of appearance, postural position, and mobility of the cheeks pre- and post-FES associated with masticatory training of masseter muscles in individuals with Down syndrome

Variable	Median		p-value
	Pre	Post	
Appearance	2.00	3.00	0.002*
Inflation	3.00	3.00	1.00
Insufflation	3.00	3.00	0.396
Retraction	3.00	3.00	0.63
Lateralization	1.00	1.00	0.581

*significant $p < 0.05$; Wilcoxon test; N=16; FES = Functional Electrical Stimulation

Source: João Pessoa, 2016

Table 3. Comparison of tongue mobility pre- and post-FES associated with masticatory training of masseter muscles in individuals with Down syndrome

Variable	Median		p-value
	Pre	Post	
Protrusion	3.00	3.00	0.317
Retraction	3.00	3.00	0.414
Right lateralization	1.00	3.00	0.049*
Left lateralization	1.00	3.00	0.049*
Elevation	3.00	3.00	0.102
Lowering	3.00	3.00	0.180

*significant $p < 0.05$; Wilcoxon test; N=16; FES = Functional Electrical Stimulation

Source: João Pessoa, 2016

Table 4. Comparison of differences in muscular behavior during performance of functions of the stomatognathic system pre- and post-FES associated with masticatory training of masseter muscles in individuals with Down syndrome

Variable	Median		p-value
	Pre	Post	
Respiration	2.00	3.00	0.025*
Behavior of the lips - Deglutition	2.00	3.00	0.033*
Behavior of the tongue - Deglutition	3.00	3.00	0.655
Signs of head movements - Deglutition	1.00	1.00	0.083
Signs of tension of the masseter muscles - Deglutition	0.00	1.00	0.049*
Food escape - Deglutition	1.00	1.00	0.257
Solid bolus efficacy - Deglutition Deglutição	3.00	3.00	0.480
Liquid bolus efficacy - Deglutition	3.00	3.00	0.705
Mastication (bite)	2.00	3.00	0.043*
Mastication (trituration)	3.00	4.00	0.035*

*significant $p < 0.05$; Wilcoxon test; N=16; FES = Functional Electrical Stimulation

Source: João Pessoa, 2016

DISCUSSION

Considering the results obtained, substantial gains were verified with the application of electrostimulation associated with masticatory training of masseter muscles in individuals with Down syndrome (DS). Some studies highlight the importance of interventions in SD, considering the altered functions of the stomatognathic system of these individuals, and evidence the aspect of generalized muscular hypotonia as a factor indicative of orofacial myofunctional impairments^(4,5,17,18).

Comparison of data on the differences in appearance, postural position, and mobility of the lips pre- and post-intervention showed a change in the posture of the lips, which became

sealed, after application of Functional Electrical Stimulation (FES). Masticatory training during electrostimulation with instruction to chew with the lips sealed may have contributed to proprioception and correction of lip posture, considering that masticatory function has been used as an important speech-language therapy resource when there is intention to exercise the mandibular elevator muscles, buccinator muscles, orbicularis oris muscle, and tongue musculature⁽¹⁹⁾.

Comparison of differences in the appearance, postural position, and mobility of the cheeks pre- and post-electrostimulation showed significant change regarding their structural aspect - previously flaccid and arched, evidencing satisfactory effects for a tendency to normality patterns. Masticatory training can assist with stability

of the gains achieved through Speech-language Pathology (SLP) intervention, because not only the masseter muscles, but also the buccinator muscles, orbicularis oris muscle, and tongue musculature are recruited, providing constant muscular exercise. Muscle training may bring changes in the dynamics and strength of the muscular structures that compose the masticatory system⁽²⁰⁾.

In the present study, significant changes in the mobility pattern (right and left lateralization) were observed when differences in tongue mobility were compared pre- and post-FES in patients with DS. In addition, significant differences were found in muscular behavior during the functions of the stomatognathic system: respiration, deglutition and mastication, pre- and post-electrostimulation.

Muscle fibers can modify their physiological and biochemical characteristics according to the stimuli to which they are subjected, with results reflecting on the amount or type of muscle proteins. Such adaptive ability involving several fiber components corresponds to muscle plasticity. The contraction mechanism of the skeletal musculature can occur by means of a controlled voluntary command coordinated by the brain or through an involuntary command induced by an external electrical stimulus^(21,22).

Regarding the breathing function, this study demonstrated that this function was characterized as oronasal prior to intervention, clinically evolving after the application of FES, when the volunteers began to present nasal breathing with considerable improvement of the mandibular posture and predominance of lip sealing. Phenotypic facial features and oral motor development in DS demonstrate the possibility of adapted deglutition, presenting tongue protrusion, inadequate tongue posture at rest, accumulation of saliva in the oral cavity, as well as impairments in speech production⁽³⁾. This adaptation promotes, in development, behaviors that hinder nutrition, such as oral refusal or aversion, limiting oral motor experiences and compromising the performance of these skills⁽²³⁾.

In this study, improvement in lip behavior during swallowing was also observed; initially, some patients were able to perform lip sealing with muscular tension and after application of FES with masticatory training they were able to seal their lips without perioral muscular tension and tension of the facial musculature. With respect to the aspects of bite and trituration of the masticatory function, significant gains were observed pre- and post-electrostimulation, with bilateral alternate trituration and bite performed using the incisors, both with a tendency to normality standards.

A literature review on the use of electrostimulation in SLP practice showed very few published articles on the theme, because application of this technique is recent in this area. In this review, the authors emphasized the effect of use of electrostimulation in the treatment of dysphagia and concluded that electrostimulation associated with therapy is more beneficial than traditional therapy in dysphagia, increasing oral intake, reducing degree of dysphagia and laryngotracheal aspiration, and enabling return to the oral route. Thus, according to the authors, traditional therapy associated with electrostimulation brings more benefits to patients than traditional therapy alone⁽⁸⁾.

Aware that there are still few studies on electrostimulation in SLP, it is hoped that the present research, given the significant results presented, contribute to the clinical practice of Orofacial Motricity when intervening in individuals with DS.

CONCLUSION

Electrostimulation associated with masticatory training provides statistically significant gains to the orofacial musculature and the chewing, breathing and swallowing functions in individuals with Down syndrome. Results show new possibilities for intervention, associating Functional Electrical Stimulation with conventional orofacial myofunctional therapies.

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Author contributions

DLSAP participated in collection, analysis and interpretation of the data and writing of the manuscript; GASA was responsible for the study design, collection, analysis and interpretation of the data, and writing of the manuscript; FMMF, LAS, and SMFP participated in data collection; LSFP and LNAA were in charge of analysis and interpretation of the data and writing of the manuscript.