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Research Article

Electromyographic study on the effects of an occlusal stabilization appliance on cervical dystonia-affected muscles

[1]Camelia Navrotchi, [2]Bogdan Florea, [3]Mindra E. Badea
[1] "Iuliu Hatieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania; [2] Epilepsy and EEG Monitoring Center Cluj-Napoca, Romania; [3] Department of Preventive Dentistry, Faculty of Dental Medicine, "Iuliu Hatieganu" University of Medicine and Pharmacy, Cluj-Napoca, Romania.

Abstract. Aim: The aim of this study was to analyze the immediate effects of an occlusal stabilization appliance on the electromyographic activity of cervical dystonia-affected muscles. Material and methods: Eleven patients neurologically diagnosed with primary cervical dystonia underwent complex dental examination. An occlusal stabilization appliance was manufactured in the laboratory for each patient. The most contracted neck muscle at the moment of the examination was electromyographically investigated. Results: While wearing the appliance, spontaneous muscle activity decreased significantly in 4 patients, ceased in 4 patients and remained unchanged in 3 patients. Conclusion: Temporary changes in occlusion reduced the electrical activity of the dystonic neck muscles.

Key Words: electromyography, occlusal stabilization appliance, cervical dystonia, spontaneous muscle activity.

Corresponding Author: C. Navrotchi, email: camelianavrotchi@yahoo.com

Introduction

Cervical dystonia (CD) is a neurological disease characterized by prolonged involuntary contractions in the neck muscles causing abnormal movements and postures of the head and neck (Albanese et al 2013). It is a form of focal dystonia defined from the perspective of non-specific changes of the motor system, mainly concerning the planning and execution of voluntary movements (Avanzino et al 2015). Recent studies indicate that besides the alteration of motor functions there are also sensory dysfunctions. The complex sensorimotor integration process through which the central nervous system uses the sensory information received from the peripheral level for assisting motor program execution is modified in patients with dystonia (Abbruzzese et al 2003; Tinazzi et al 2003; Stamelou et al 2012; Berman et al 2015; Patel et al 2014).

CD is mostly primary, idiopathic, although there are also secondary forms involving basal ganglia lesions (Defazio et al 2007). The treatment of CD is symptomatic, aiming to improve quality of life of those affected, and includes oral medication, botulinum toxin injections and surgical treatment. Additionally, patients resort to physiotherapy, various relaxation techniques, psychological counselling and alternative therapies to relieve their symptoms (Skogseid et al 2014; Jinnah et al 2015). In the literature, the use of dental appliances to alleviate symptoms of dystonia is reported in some cases of cervical, oromandibular and lingual dystonia (Sims et al 2012; Gonzalez-Alegre et al 2014; Schneider et al 2011; Maestre-Ferrin et al 2010; Lo et al 2007; Illing et al 1999). Some authors focused on investigating the effects of occlusal appliances on the electrical activity of the masticatory and neck muscles in asymptomatic (Ceneviz et al 2006; Miralles et al 2002) and symptomatic subjects (Santander et al 1994; Miralles et al 1992), showing that temporary changes in the mandible position influences the craniocervical system.

Electromyography (EMG) is performed to investigate the electrical activity of the muscles affected by CD for diagnostic purposes, a proper selection of the muscles to be injected with botulinum toxin or for the evaluation of the efficacy of the botulinum toxin injections (Comella et al 1992; Werdelin et al 2011; Brans et al 1998; Van Gerpen 2000; Nijmeijer et al 2013).

The aim of this study was to analyze the immediate effects of an occlusal stabilization appliance (OSA) on the electromyographic activity of the muscles affected in CD. We also evaluated several factors that could have modified the electromyographic response.

Material and methods

This was an interventional, analytic, longitudinal, prospective, and cohort study.

The study included 11 patients with a mean age of 46.5±14.8 years, 7 (63.6%) women and 4 (36.4%) men, diagnosed with primary CD. The patients included were treated in 3 Departments of Neurology from Cluj-Napoca, Bucharest and Timisoara, between July 2016 and March 2017. Patients diagnosed with any other neurological diseases were not included. This study was approved by the Ethics Committee of "Iuliu Hatieganu" University of Medicine and Pharmacy, Cluj-Napoca. All patients signed the informed consent before participating in the study.

The patients were examined extra and intra-orally. The following parameters were recorded: demographics, CD duration, and specific drug or non-drug therapies. The MRIs of temporomandibular joints (TMJ) were performed in order to study the health status of TMJ.

Dental impressions, centric relation recordings, upper jaw position recordings using the face bow, casts mounted on the Artex CR articulator (Amann Girrbach) were necessary to manufacture standardized OSAs for each patient. The OSAs were manufactured in a private dental laboratory (Ortoclasic Lab) by the same experienced dental technician for each patient, using the established techniques described by Okeson (2013). OSA is a removable dental device applied on the occlusal surfaces and the incisal edges of the teeth of one dental arch. The OSA made from hard transparent acrylic (Orthocryl, Dentaurum) was used in the upper or lower jaw, depending on the clinical situation of each patient's dental arches, with double retention: through friction and two Stahl wire hooks. The OSA had a flat occlusal surface, stable and even contacts with the teeth of the opposing arch and functionalized guidance in the protrusive and lateral movements of the mandible.

The electrical activity of the muscles affected by CD was recorded in a private neurology practice by needle EMG using the two-channel NeuroSoft electromyograph. We used 25 mm SEI type bipolar disposable needles. Prior to EMG, a check-up session was conducted in the dental office and the patients were trained to apply and remove the appliance on and from the dental arches.

The patients were comfortably seated in an ergonomic chair in a relaxed position, with the back to the electromyograph. The selection of the muscle to be electromyographically investigated was based on: patient symptoms, i.e. the most contracted muscle in the cervical area at that moment; clinical palpation of the neck muscles by the examiner; the patient's medical history of botulinum toxin injections, regarding doses and injected muscles. When electromyography was performed, none of the patients studied were under the influence of botulinum toxin, the last botulinum toxin injection being administered at least three months ahead, time interval that makes the effects of botulinum toxin disappear. A recording of 20 seconds of the electrical activity of the selected muscle was performed, with the mandible in resting position, after which the patient, without changing the position in the examination seat, applied the OSA on the dental arch, bit on it and remained in this position for ten minutes. The needle was kept motionless in the examined muscle throughout this time by the examiner, in order not to change the electromyographic recording site. After 10 minutes, a second recording of the electrical activity of the examined muscle was performed, with the OSA in the oral cavity, in resting position of the mandible.

We analyzed the spontaneous activity of the examined muscles, i.e. the presence, amplitude and frequency of positive sharp waves, fibrillations.

Statistical analysis was performed using MedCalc Statistical Software version 17.6 (MedCalc Software bvba, Ostend, Belgium; http://www.medcalc.org; 2017). Continuous variables were characterized using the mean and standard deviation or median and 25th-75th percentiles, depending on the situation. Categorical variables were described by frequency and percentage. Differences between groups were assessed by the Kruskal-Wallis test or the chi-square test. The differences between the two EMG recordings were tested using the marginal homogeneity test. A p value <0.05 was considered statistically significant.

Table 1: EMG results before and during OSA use

Variable		Without OSA	With OSA	P
Positive waves	0	-	4 (36.4%)	0.008
	1	5 (45.5%)	6 (54.5%)	
	2	4 (36.4%)	1 (9.1%)	
	3	2 (18.2%)	-	
Fibrillations	0	4 (36.4%)	6 (54.5%)	0.04
	1	5 (45.5%)	5 (45.5%)	
	2	2 (18.2%)	-	

Results

Ten patients had rotational torticollis along the vertical axis, 4 turning to the left and 6 turning to the right side. One patient had laterocollis, shifting the head to the left side. The examined muscle was the left trapezius muscle in 6 patients, the right trapezius muscle in 4 patients and the left splenius capitis in one patient. The EMG analysis before and during OSA use showed a significant reduction in positive waves and fibrillation amplitude and frequency (Table 1).

We analyzed the results of the EMG in correlation with the TMJ health status (Table 2). Although our results showed that the OSA produced immediate relaxation effects on the dystonic muscles, even in four out of the six patients with healthy TMJ, the differences were not statistically significant (p=0.5).

We studied several factors that could have influenced muscle activity after the patients used the OSA (Table 3). Although we could not find a statistically significant difference, we observed that older patients, patients with a longer history of the disease, and patients that underwent physiotherapy, were more likely to have a reduced or even complete cessation of muscle activity.

Discussions

To the best of our knowledge, this study is the first to investigate the electromyographic activity of the muscles affected in CD when using an occlusal stabilization appliance. We chose this type of dental appliance for our study as it is a therapeutic dental appliance with reversible effects, primarily employed to reduce the pain in the masticatory muscles in temporomandibular disorders. It is also known as muscle relaxation appliance (Okeson 2013).

In our research, the examined muscles were affected by involuntary contractions caused by the present neurological movement disorder. The initial spontaneous muscle activity of the studied muscles presented positive sharp waves and fibrillations, which in 4 out of the 11 studied patients reduced their frequency and amplitude and in 4 of them completely disappeared, when the appliance was in the oral cavity. In the other 3 patients, the spontaneous muscle activity remained the same as before using the appliance. These findings were statistically significant.

Table 2: EMG results in correlation with the health status of the TMJ

Variable	Healthy TMJ	TMJ disorder	MRI detected pathological changes, but no signs or symptoms of TMJ disorder
Complete cessation	2 (33.3%)	1 (33.3%)	1 (50.0%)
Reduced activity	2 (33.3%)	2 (66.7%)	-
Unchanged activity	2 (33.3%)	-	1 (50.0%)

Table 3. EMG results in correlation with other variables

Variable		Complete cessation	Reduced activity	Unchanged activity	P
Age		45.5 (37.5; 52)	56 (35; 74.7)	36 (30; -)	0.3
Gender	**M**	-	2 (50%)	2 (66.7%)	0.1
	F	4 (100%)	2 (50%)	1 (33.3%)	
Disease duration		10.5 (8.5; 15.5)	8 (5.5; 12.7)	6 (5; -)	0.3
Muscle relaxants		4 (100%)	3 (75%)	3 (100%)	0.3
Physiotherapy		4 (100%)	3 (75%)	1 (33.3%)	0.1
Relaxation		2 (50%)	2 (50%)	-	0.3

The following aspects explain the way OSA influenced the electrical activity of the examined muscles: a more stable and optimal occlusion; normal periodontal ligament proprioception, because OSA dissipates the forces placed on individual teeth, thus balancing the load and allowing for muscle symmetry; increase in the vertical dimension of the occlusion, thus a decrease in the temporomandibular intra-articular pressure and a consecutive decompression of the TMJ tissues; mandibular position changes; a more stable condyle-disc-eminence relationship; changes in the peripheral input to the central nervous system, followed by the reorganization of the reflex neuromuscular activity, with consecutive reduction of abnormal muscle activity (Okeson 2013; Nitzan 1994; Casares et al 2014, Sims 2012).

Patients undergoing physiotherapy were more likely to have a reduced or even complete cessation of the dystonic muscle activity. This result is in accordance with De Pauw et al research findings, suggesting that physiotherapy adjunct to BoNT/A injections may improve head position, decrease pain levels in patients with CD and improve functioning in everyday activities in the short term (De Pauw et al 2014).

Sims et al (2012) stated that the success rate in relieving painful symptoms and dystonic head posture in CD patients by means of an occlusal appliance is good for those with well-defined TMJ disorders. Their hypothesis is that an internal derangement of the TMJ (disc displacement, arthrosis) may constantly stimulate the auriculotemporal branch of the trigeminal nerve, which has direct input into the reticulate formation, and may activate the cells of the pontine region of the reticulate formation, known for the control and deviation of the head posture. By increasing the vertical dimension of the occlusion, the dental appliances determine a decompression of the TMJ tissues, followed by cessation of stimulation of the auriculotemporal nerve (Sims et al 2012). In our study, all patients underwent complete examination at the level of the masticatory system prior to electromyography. We analyzed EMG results in correlation with the TMJ health status and we found different results from those reported by Sims et al, namely that OSA produced immediate relaxation effects on the dystonic muscles, even in four out of the six patients with healthy TMJ.

Another aspect we need to discuss is whether the occlusal appliance can be considered as a sensory trick. The pathophysiological mechanism of sensory tricks is unknown. Sensory tricks or antagonistic gestures are those actions intentionally performed by the patient in order to diminish the symptoms of dystonia and they are also characteristic for other forms of focal dystonia (blepharospasm, musician's dystonia). Thus, patients with torticollis frequently touch their chin, face or neck, as touching the affected area or the neighbouring area reduces muscle contractions (Jahanshahi et al 2000; Patel et al 2014; Schramm 2004). The duration of sensory trick effect upon introduction of the stimulus varies between a few second to hours (Ramos et al 2014; Muller et al 2001; Ochudlo et al 2007).

Schneider et al applied a pivotal dental appliance to a patient diagnosed with oromandibular dystonia and dysarthria that allowed contact with the teeth on the lower arch only in the area of the molars on the right side and noted the complete disappearance of dystonia symptoms and normal speech for one month. At the three-month check-up, dystonia symptoms returned, but not with the same severity. By modifying the pivotal area of the appliance, they managed to relieve symptoms of dystonia again, but did not achieve a complete disappearance, as it did the first time, and the effect was temporary. This evolution determined by the use of the appliance was explained by the fact that changes in the neuromuscular proprioception caused by the insertion of the device can restore the mechanisms that initiated dystonia to normal until the nervous system becomes accustomed to this position and then dystonia returns (Schneider et al 2011). In our study, temporary changes in occlusion influenced the electrical activity of dystonic muscles. These immediate effects on cervical dystonia-affected muscles obtained using OSA suggest that 8 of the studied patients can use them to relax their contracted muscles over very short periods of time.

Our research has a few limitations related to the small sample size and to the fact that only one muscle affected by dystonia was investigated in each patient. The research should continue on larger groups of patients with CD, extending the electromyographic investigation of OSA effects on more neck muscles affected by dystonia in each patient. There is need for a study

that would also investigate the sensory trick of the appliance by extending the investigation time. By continuing this type of research, we could assess the possibility of using these removable appliances in combination with other treatments to improve quality of life in these patients.

Conclusions

Temporary changes in occlusion induced by OSA modified the electrical activity of dystonic neck muscles by reducing the frequency and amplitude of initial positive sharp waves and fibrillations. Older patients, patients with longer history of the disease, and patients that underwent physiotherapy were more likely to have a reduced or even complete cessation of muscle activity.

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Authors

•Camelia Navrotchi, "Iuliu Hatieganu" University of Medicine and Pharmacy, 31 Avram Iancu Street, 400083, Cluj-Napoca, Cluj, Romania, EU, e-mail: camelianavrotchi@yahoo.com

•Florea Bogdan, Epilepsy and EEG Monitoring Center, 10 Iuliu Moldovan Street, 400348, Cluj-Napoca, Cluj, Romania, EU, e-mail: bogdan_florea@yahoo.com

•Mindra Eugenia Badea, Department of Preventive Dentistry, Faculty of Dental Medicine, "Iuliu Hatieganu" University of Medicine and Pharmacy, 31 Avram Iancu Street, 400083, Cluj-Napoca, Cluj, Romania, EU, e-mail: mindrabadea@yahoo.com

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