

THE INFLUENCE OF OCCLUSAL STABILIZATION APPLIANCES ON CERVICAL DYSTONIA SYMPTOMS

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Abstract

Objectives. The aim of this study was to assess the therapeutic efficiency of the occlusal stabilization appliance (OSA) in patients with cervical dystonia (CD).

Methods. The study included 11 patients aged between 29 and 80 years, 7 women and 4 men, diagnosed with primary CD. The patients underwent an extra- and intra-oral clinical examination, followed by para-clinical examinations, necessary for the specifications of the OSA. The following data were recorded: demographic parameters, CD duration, management of the disease, dental impression, recording of the centric relationship, recording of the position of the upper jaw with the facial bow. A standardized OSA was manufactured in a private dental laboratory. Patients received instructions for wearing the OSA for 24 hours. Patients filled a questionnaire designed by us, which evaluated the effects of wearing the OSA over a 24-hours period on the symptoms of CD: muscles contraction, pain, discomfort while walking, sleep quality, tremor. The patients kept the dental appliances, and after three months they completed the questionnaire one more time.

Results. The OSA was applied on the lower arch in 3 (27.3%) patients and on the upper arch in 8 (72.7%) patients. The OSA wearing time for the first 24 h was on average 19.2 ± 6 hours. Total relaxation of dystonic muscles was reported by 9 (81.8%) patients, while 2 (18.2%) patients related partial muscle relaxation. Seven (63.6%) patients reported a pain decrease. Increased comfort while walking was observed by 8 (72.7%) patients. Two (18.2%) patients described an increase of sleep quality. In two (18.2%) patients the tremor disappeared. All patients reported difficulties while eating and removed the OSA during meals. Patients who wore the OSA for more hours, experienced a pain decrease ($p=0.08$), an increase in sleep quality ($p=0.1$), the disappearance of the tremor ($p=0.1$). After three months, only seven patients continued to use the OSA. More patients described a pain decrease after three months (5 (71.4%) vs. 4 (57.1%); $p=0.5$), relaxation of dystonic muscles (7 (100%) vs. 6 (85.7%); $p=0.3$).

Conclusions. The use of OSA might be beneficial in CD patients, as it reduced the dystonic symptoms, pain severity and improved the quality of sleep.

Keywords: masticatory system, temporomandibular disorder, occlusal stabilization appliance, cervical dystonia, muscle relaxation

Introduction

Cervical dystonia (CD) is a neurological movement disorder, characterized by sustained involuntary contractions of the neck muscles, which determine abnormal movements and positions of the head and the neck [1]. A study in eight

European countries showed that the prevalence of CD was 5.7 per 100,000 persons [2]. In Romania, CD is an underdiagnosed condition and there are no studies on its prevalence in the general population.

The quality of life in patients with CD is significantly affected. The abnormal position and movements of the head and neck, the pain, the discomfort, the hypertrophy of the cervical muscles, neck and / or shoulder tremor, deficits in

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balance and gait reduce the patient's ability to perform daily activities and it modifies their behavior in their relationship to family and friends. In many cases, the anxiety, depression, and bad mood determine self-isolation and withdrawal from social life [3,4,5,6,7].

The pathogenesis of dystonia is still incompletely elucidated; the current concepts focus on dysfunctions in the complex motor network in which the basal ganglia, cerebellum, cerebral cortex, and other brain regions play a key role. Some forms of dystonia are associated with genetic mutations in more than 100 different genes. Acquired dystonia results from damage to the central nervous system, tumors, infections, exposure to certain drugs or chemical substances [8,9,10]. Therefore, treatment of cervical dystonia is symptomatic and includes oral medication, botulinum toxin injections and surgical treatment. To relieve their symptoms, patients also resort to physical therapy, psychological counseling, acupuncture, relaxation techniques, exercise, swimming, music therapy [11,12]. The first line of treatment are the botulinum toxin injections, with different outcomes in relieving symptoms of dystonia, dependent on the physician's experience, the dose used, and the quality of botulinum toxin. Oral treatment is limited in efficacy, often causing generalized and problematic adverse effects and there is a lack of sound scientific evidence supporting the use of most agents [13,14,15]. Emerging treatment approaches for CD focus on the management of non-motor symptoms and rehabilitation strategies [16].

Through muscles attachments, the joints between them, blood vessels and nerves, the stomatognathic system, also called masticatory system, is in tight connection with the cervical spine and the skull, thus forming the cranio-cervico-mandibular system. The masticatory muscles have tight functional relationships with the muscles of the adjacent regions in maintaining the postural balance of the head [17,18,19,20].

The occlusal stabilization appliance (OSA) is a therapeutic device with reversible effects, also called muscles relaxation appliance because it is primarily used to reduce muscle pain in the masticatory muscles in temporomandibular disorders [21,22]. OSA is a removable device applied on the occlusal surfaces and the incisal edges of the teeth of one dental arch. Sims et al. reported having successfully used dental appliances to relieve symptoms of dystonia in three patients diagnosed with CD and temporomandibular dysfunction [23].

The aim of the present study was to assess the therapeutic efficiency of the OSA appliance in CD patients.

Material and methods

The study was interventional, analytic, longitudinal, prospective, and cohort.

The study included 11 patients aged between 29 and 80 years, 7 women and 4 men, diagnosed with primary CD. The patients were recruited from those examined in

three Clinics of Neurology from: Cluj-Napoca, Bucharest, Timișoara, Romania, between July 2016 to March 2017. Patients presenting with other neurological conditions were excluded. This study was approved by the Ethics Committee of the Iuliu Hatieganu University of Medicine and Pharmacy, Cluj-Napoca, with the number 284/29.06.2016. All patients signed the informed consent before participating in the study.

We clinically examined the patients at the level of the masticatory system. Dental x-rays (panoramic and lateral cephalometric x-ray) and temporomandibular joints MRI were performed for each patient in order to complete the clinical data on the dento-maxillary pathology, regarding the odontal, edentulous, occlusion, surgical, orthodontic, periodontal and temporomandibular joints condition.

In order to manufacture the OSA, we took dental impressions and recorded the centric relationship and the position of the upper jaw with the facial bow in each patient. The casts were mounted on the Artex CR articulator (Amann Girrbach) in a private dental laboratory (Ortoclasic Lab). A standardized OSA was manufactured for each patient by the same experienced dental technician, using the established techniques described by Okeson [19]. The OSA was made from hard transparent acrylic (Orthocryl, Dentaaurum), 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. We modified the usual one piece design of the OSA into a three pieces appliance, by cutting it distal to the canines, so that three pieces resulted: one anterior (from canine to canine), and two laterals (Figure 1).

Patients received instructions for wearing the OSA for 24 hours as follows: the lateral pieces during the day, the anterior piece during the night, removal of the appliance during dental hygiene. This type of wearing was designed to facilitate the patient's comfort during daily activities, speaking and mastication. To prevent extrusion of anterior teeth and/or intrusion of posterior teeth, the anterior piece was worn alone at night. At the time of wearing the OSA for 24 hours, all patients were at least 3 months apart from the last botulinum toxin injection.

Patients filled in a questionnaire designed by us, which evaluated the effects of wearing the OSA over a 24-hours period on the symptoms of CD. The questionnaire included questions about: the duration of wearing the appliance, effects on the dystonia symptoms, difficulties in wearing the appliance, side effects, and other observations.

The patients kept using the dental appliances, and after three months they completed the questionnaire one more time. Besides the prior questions, we inquired about the frequency of the usage of OSA, reasons for continuing/discontinuing the use of OSA, symptoms of CD and management of the disease.



Figure 1. The upper/lower occlusal stabilization appliance.

Statistical analysis was carried out using the MedCalc Statistical Software version 17.6 (MedCalc Software bvba, Ostend, Belgium; <http://www.medcalc.org>; 2017). Quantitative variables were tested for normality of distribution using the Kolmogorov-Smirnov test and was described using the mean and standard deviation or median and 25-75 percentiles, when appropriate. Qualitative data were characterized by frequency and percent. Comparison between groups was performed using the Student t test or the chi-square test, when appropriate. The results obtained at three months were compared with the baseline data using the marginal homogeneity test. A p value <0.05 was considered statistically significant.

The following data were analyzed: demographic parameters, CD duration, symptoms of CD (muscles contraction, pain, discomfort while walking, sleep quality, tremor), management of the disease (drug treatment, physiotherapy, relaxation techniques), temporomandibular joint health status, duration of wearing the appliance, effects on the dystonia symptoms, difficulties in wearing the appliance, side effects.

Results

The extra-oral clinical examination shows the dystonic position of the patients (Figure 2).

The demographic, clinical and imaging characteristics of the patients are described in table I.

The results of the questionnaire after 24 hours of OSA use are described in table II. All patients reported difficulties while eating and removed the OSA during

meals. Transient side effects were reported by three patients: discomfort at the lateral surfaces of the tongue, nausea, sensitive teeth.

A pain decrease was described by patients that used the OSA for more hours (23 (22; 23) hours vs. 15 (7; 22) hours; $p=0.08$), patients with a shorter disease history (8 (6; 11) years vs. 10.5 (6.2; 15.5) years; $p=0.4$) and were older (48.1 ± 17.5 years vs. 43.7 ± 10 years; $p=0.9$). Patients that used relaxation techniques were more likely to experience a pain reduction (4 (57.1%) vs 0; $p=0.2$). Gender and physiotherapy did not influence the pain perception ($p=1$).

The comfort while walking was not influenced by the longer use of OSA, disease history, age or gender. Patients that used relaxation techniques or physiotherapy described an increase in comfort while walking ($p=0.2$; $p=0.1$ respectively).

After three months, only seven patients continued to use the OSA. Two patients discontinued the use of OSA because of dental procedures, and the other two felt no effect on CD symptoms. Four (57.1%) of these seven patients used the OSA during the day and the other 3 (42.9%) patients used the OSA during the night, when the contractions of the muscles affected by dystonia were too strong. Four (57.1%) patients continued to take anticholinergic agents, benzodiazepine and muscle relaxants, while the others had to stop the drugs due to their side effects. Four (57.1%) patients used relaxation techniques and one (14.3%) patient used physiotherapy. The evolution of symptoms is described in table III.



Figure 2. Right spasmodic torticollis and retrocollis, left spasmodic torticollis, right spasmodic torticollis and laterocollis (from the left to the right).

Table I. Group demographic, clinical and imaging characteristics.

Variable	Description	
Age (years) (mean±standard deviation)	46.5±14.8	
Gender	Men (number (%))	4 (36.4%)
	Women (number (%))	7 (63.6%)
CD duration (years) (mean±standard deviation)	9.3±3.7	
CD type	Left side torticollis (number (%))	5 (45.5%)
	Right side torticollis (number (%))	6 (54.5%)
Temporomandibular disorder (number (%))	2 (18.2%)	
MRI detected incipient condyle surface remodelling arthrosis (number (%))	2 (18.2%)	
Temporomandibular joints within the normal limits (number (%))	6 (54.5%)	
Use of relaxation techniques (number (%))	4 (36.4%)	
Use of physiotherapy (number (%))	8 (72.7%)	

Table II. Results after 24 hours of OSA use.

Variable	Description	
The OSA wearing time for the first 24 hours (mean±standard deviation)	19.2±6	
OSA application	Lower arch (number (%))	3 (27.3%)
	Upper arch (number (%))	8 (72.7%)
Total relaxation of dystonic muscles (number (%))	9 (81.8%)	
Partial muscle relaxation (number (%))	2 (18.8%)	
Pain decrease (number (%))	7 (63.6%)	
Increased comfort while walking (number (%))	8 (72.7%)	
Increase of sleep quality (number (%))	2 (18.8%)	
Disappearance of tremor (number (%))	2 (18.8%)	

Table III. Evolution of symptoms at three months.

Variable	After 24 h	After 3 months	p
Pain decrease (number (%))	4 (57.1%)	5 (71.4%)	0.5
Relaxation of dystonic muscles (number (%))	6 (85.7%)	7 (100%)	0.3
Increase in sleep quality (number (%))	2 (28.5%)	2 (28.5%)	1
Disappearance of the tremor (number (%))	1 (14.2%)	1 (14.2%)	1
Increase comfort while walking (number (%))	6 (85.7%)	2 (28.5%)	0.04

Discussion

To our knowledge, this study is the first to analyze the influence of OSA on cervical dystonia symptoms. Our results show that OSA induced muscles relaxation, reduction of pain, comfort while walking, a better quality of sleep and tremor disappearance.

OSA changes the occlusion and the position of the mandible and reduces the temporomandibular intra-articular pressure, determining an alteration of the information transmitted from the peripheral level to the central nervous system and the reorganization of the reflex neuromuscular activity, with consecutive reduction of abnormal muscle activity and muscle pain [20,24,25]. Wearing the OSA for 24 hours reduced the pain in seven (63.6%) patients. These results are in accordance with the results of Kashima et al.'s which showed that the short-term wear of an OSA may inhibit certain harmful and sensory stimuli from the innervated cervical structures. That makes the OSA suitable for the treatment of painful cervical disorders [26].

In our study, patients who used relaxation techniques were more likely to experience a pain reduction, an increase in sleep quality and an increase in comfort while walking. These results were close to the statistical significance threshold. Our findings are in accordance with Boyce et al. research results which show that, given the fact that mental and physical load may aggravate the symptoms of CD, it is possible that whole body relaxation in itself has a beneficial effect on the symptoms of CD [27].

Patients who used physiotherapy described an increase in comfort while walking after 24 hours of wearing the OSA. After three months, only one patient still used physiotherapy, and the number of patients who reported an increased comfort while walking was reduced to two patients. This finding is in agreement with other studies on rehabilitation which suggest that a multimodal physiotherapy program consisting of active exercises, stretching and relaxation may improve disease severity and the quality of life in patients with CD [28,29,30].

Patients that used the OSA for more hours were more likely to experience a pain decrease CD, an increase in sleep quality and the disappearance of the tremor. This results suggests the fact that OSA's influence on CD

symptoms is time dependent.

At least half of the patients with CD suffer from sleep disturbances. Sleep disturbance is associated with depressive symptoms. The frequency and duration of dystonic movements is markedly reduced during sleep [31]. Eichenseer et al. found that sleep quality was not improved after botulinum toxin treatment, despite an important improvement in motor symptoms [32]. In our study, two patients described an increase of sleep quality after 24 hours of wearing the OSA and at three months follow-up they reported to use the OSA during the night, when contractions were too strong, in order to get muscle relaxation and a good sleep. This result is in accordance with Rosar et al.'s findings that short-term interocclusal appliance therapy had a positive effect on sleep quality [33].

Esposito et al. performed a computerized gait analysis in CD patients, before and after botulinum toxin injections, and in healthy controls and found that CD patients demonstrated a significant reduction of velocity, stride length and dynamic stability index while stride and swing time were increased. No significant effect of botulinum toxin injections was detected [34]. In our study, 72.7% of patients reported an increase comfort while walking during the 24 hours wearing time of the OSA.

Sims et al. state that there is growing clinical evidence that temporomandibular disorders play an important role in the pathogenesis of CD when they coexist. They reported having successfully used dental appliances in three patients diagnosed with both CD and temporomandibular disorder to relieve symptoms of dystonia. Their hypothesis was that their dental appliances increased the vertical dimension of the occlusion and produced a decompression of the joint tissues, followed by the cessation of continuous irritation of the peripheral branches of the auriculotemporal nerve, which provides the temporomandibular joint innervation. There may be a neuritis of the auriculotemporal branch of the trigeminal nerve, which has direct input into the reticulate formation, and it may activate the cells of the pontine region of the reticulate formation, known for the control and deviation of the head posture [22]. In our study, all the participants have reported relaxation of the muscles affected by dystonia by wearing the dental appliance for 24

hours, even the 6 patients with healthy temporomandibular joints. Also, in the long run, 4 of these 6 patients with healthy temporomandibular joints, continued to use the OSA, alongside medication, botulinum toxin, physiotherapy and relaxation techniques to relieve symptoms of dystonia.

All patients investigated in this study were following treatment with botulinum toxin, at varying periods of 3-6 months, depending on the recurrence of symptoms, with different outcomes in relieving symptoms of dystonia, depending on the dose used, the injection technique and the quality of botulinum toxin. Studies show that efficacy of botulinum toxin injections in CD patients on motor symptoms varies from 20 to 70% [16]. In the long run, seven out of the eleven patients who participated in this study continued to use the OSA alongside other therapies. These aspects indicate the fact that no treatment that patients follow brings about a complete disappearance of the symptoms of dystonia and that they combine various therapies according to the severity of the symptoms at any given time. Our research shows that the OSA can be used in combination with other therapies to provide the patient with a better quality of life, the main objective of CD treatment.

A limitation of our research is related to the small size of the studied group of 11 patients diagnosed with primary CD, due to the fact that this is a rare disease, little known in Romania, with an etiology and pathogenesis left unknown at this moment. A study of the influence of OSA on CD symptoms on larger patient populations could provide us with more information about the connection between CD and the masticatory system, respectively the effects of the OSA on CD symptoms.

Conclusions

Due to the close anatomical and functional relationship and to the common role in maintaining the head posture, changes at the level of one element of the cranio-cervico-mandibular system may cause alterations throughout the whole system. Our results show that the changes at the level of the masticatory system of the patients produced by the OSA reduced the dystonic symptoms, pain severity and improved the quality of sleep and the comfort while walking on short and long term. The use of OSA might be beneficial in CD patients in a multidisciplinary treatment approach of CD.

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