doi: 10.5505/abantmedj.2019.26878

Abant Tıp Dergisi

Olgu Sunumu / Cilt 8 Sayı 1 Yıl 2019

Abant Medical Journal

Case Report / Volume 8 Issue 1 Year 2019

Diş Çekimi Sonrası Gelişen Orofasiyal Distoni: Organik mi? Psikojenik mi?

Oro-facial Dystonia Following Dental Extraction: Is It Organic or Psychogenic?

Kübra Çoban[1], Barış Çağlar[2], Fuat Büyüklü[1]
[1]Başkent Üniversitesi Tıp Fakültesi Otorinolaringoloji Anabilim Dalı
[2]Başkent Üniversitesi Tıp Fakültesi Plastik ve Rekonstrüktif Cerrahi Anabilim Dalı

Özet

Distoni, sürekli veya aralıklı kas kasılmalarının yol açtığı bükülme benzeri, tekrarlayan hareketler veya postürel anormallikler ile karakterize bir hareket hastalığıdır. Burada 46 yaşında, dental bir prosedürü takiben meydana gelen, tek taraflı, şiddetli, süreklilik gösteren ve üst dudak deviyasyonuna neden olan kas kasılmalarından yakınan kadın hasta sunulmaktadır. Takipte hastanın distonik yakınmalarında değişkenlik saptanmış, en etkin tedavinin uygulanabilmesi amacıyla hasta, Botoks tedavisine ek olarak psikiyatrik tedavi için de yönlendirilmiştir. Vakamız, hem organik hem de psikojenik faktörler içermesi nedeniyle, farklılık göstermektedir. Bu vakalar nadiren Otorinolaringologlar tarafından takip edilmektedir. Bu nedenle karşılaşılabilecek zorluklar ve güncel terapötik uygulamalar konusunda farkındalık gerekmektedir.

Anahtar Kelimeler: Fasiyal distoni, Distoni, Psikojenik distoni, Dental cerrahi, Botoks enjeksiyonu

Abstract

Dystonia is a movement disorder characterized by sustained or intermittent muscle contractions, causing twisting, repetetive movements or postural abnormalities. We present a 46 years old female with a serious persistent complaint of unilateral continous tonic muscle contractions causing upper lip deviation, appearded after a dental procedure. During the follow-up, patient had variable dystonic symptoms and in addition to Botox therapy, psychiatric treatment was included for the best treatment outcomes. This case is unique, since both organic and psychogenic factors are involved.These cases are rarely followed by Otorhinolaryngologists, hence awareness of challenges and current therapeutic modalities are necessary.

Keywords: Facial dystonia, Dystonia, Psychogenic Dystonia, Dental Surgery, Botox Ingection

INTRODUCTION

Dystonia is a movement disorder characterized by sustained or intermittent muscle contractions, causing twisting, repetitive movements or postural abnormalities. It is usually initiated or deteriorated by voluntary actions (1,2).

Most forms of primary adult (late) onset focal or cranio-cervical dystonia have a female preponderance with a male to female ratio 1:2 (3-4).

We present a 46 years old female with persistent tonic muscle contractions causing upper lip deviation, started after a dental extraction procedure. We aim to discuss the etiological factors and the most appropriate treatment modalities regarding this rare, confounding pathology with an otorhinolaryngologist point of view.

CASE REPORT

A 46 years old women admitted to our clinic with a complaint of involuntary facial movements of the left side of her face, for nearly 10 months. She had a dental surgery, nearly 1,5 years ago. It is learned that two premolar teeth in the left upper region had been extracted and continuous pain and numbness had started just after the procedure. Numbness had resolved however, nearly 6 months after the surgery, persistent orofacial muscle movements leading to involuntary left upper lip deviation had started with a sudden onset . She had been occasionally experiencing difficulties while eating and her social life had been deeply disrupted ever since. She neither

İletişim Bilgisi / Correspondence
Uzm. Dr. Kübra Çoban, Başkent University Medical Faculty Otorhinolaryngology Department, Ankara –Türkiye
E-mail: kubracob81@gmail.com
Geliş tarihi / *Received*: 12.02.2018 **Kabul tarihi / Accepted:** 25.02.2019 **Çıkar Çatışması / Conflict of Interest:** Yok / None

had a family history of dystonic pathologies nor a personal history regarding neurological pathologies, traumatic brain injury, facial palsy or exposure to neuroleptic drugs. No specific co-morbidities were noted except gastric reflux. The only medication used was for gastric reflux treatment.

Her otorhinolaryngological examination was unremarkable, except for tonic sustained muscle contractions in the left peri-nasal and upper peri-oral area, with ipsilateral upward deviation of the left side of the lip (Figure 1). Contractions were especially triggered with the closure of the mouth. During speech, no remarkable dystonic movements were observed. Associated blefarospasm, lingual, pharyngeal and/or laryngeal spasm, involuntary jaw movements, platismal contractions were not detected.

Figure 1: Involuntary muscle tonic contractions in the left face. Upper lift deviation and the apparent perinasal groove and hypertrophic muscles are observed.

Her neurological evaluation, magnetic brain and internal acoustic canal imaging, ultrasonographic evaluation of the head and neck region were within normal limits. Electromyographic recordings were insignificant for facial nerve denervation, however increased discharge caused by spontaneous muscle activities of orbicularis oris were detected.

Possible treatment measures were thoroughly discussed with the patient and she approved Botulinum toxin therapy. Dysport®(Ipsen Limited, Slough Berkshire, UK) was injected subcutaneously into the upper lift bilaterally (3.125 Dysport unit in each 4 locus) and into the left lateral region of the alar curvature (6.25 Dysport unit) (Figure2).

Figure 2: The initial Dysport injection sites are marked.

One week after the injections, mild improvement in the lip movements were noted. She and her mother was contented with the results because the time interval between the dystonic attacks extended. Her mother mentioned that the frequency of the movements was decreased especially when she was distracted (i.e: watching television). Additionally, pain relief was satisfactory. However, a novel remarking sign of bilateral lower lip dystonia, leading to 'rabbit –like' facial movements was clearly observed when the mouth was closed. Additional low dose Dysport® was injected into the lower lip bilaterally (3.125 Dysport unit into both sides) and into the left lateral alar crest (6.25 Dysport unit) (Figure 3).

Figure 3: Injection sites in the second Dysport application are marked.

Two weeks later, significant recovery was observed. However, after 45 days of follow-up, she had apparent perioral muscular contractions

which significantly differed from her previous examinations. No additional injections were considered. The patient was referred to the psychiatry department for further evaluation.

Patients' informed consent was obtained regarding this study.

DISCUSSION

Chong-Han Pek and colleagues reported a case report in 2010, with similar upward lip deviation triggered by mouth closure. They also observed changes in the movement patterns between the visits. However, our case differs by having a history of pre-onset dental procedure and that our patient exhibits distimic behaviours giving rise to a thought of both traumatic (organic) and functional causality.

Sankhla et. al. analysed 27 peripherally induced oromandibular dystonia patients and stated that oromandibular dystonia may occur after injury, oro-mandibular surgery or a dental procedure. They found that severity of the symptoms and the disease course are more distinct in the post traumatic patients (5).

Since there is a close temporal and anatomical relevance between the dental injury and the manifestations of the symptoms, we believe, in this case, there is a strong cause-effect relation.

Additionally, this current case was a female in her late forties, showing depressive symptoms which at first was erroneously considered to be related to her embarrassing dystonic movements. However, thorough evaluation showed a single woman living with her parents, unmarried, un-employed and showing recessive behaviours. She had been using anti-reflux therapy for non-specific reflux symptoms which may be also related to her psychological status by that time. These findings were confounding and brought up the idea that our case may also be an example of psychogenic dystonia. With a more accurate explanation, a rare case of focal oro-facial dystonia based on psychogenic factors, which was induced by a dental intervention.

Fasano and colleagues investigated psychogenic facial movement disorders. They reported that the most common sign was lip pulling, predominantly downward. Unilaterality, sensorial complaints and platysma contraction was more common in the psychogenic group (6). The common clinical appearance was unilateral, dystonic lower facial movements akin oromandibular dystonia, mostly seen in young women in whom additional variable psychosocial abnormalities may be observed.

According to Fassano and colleagues, functional facial movement disorders should be considered if one has combinations of: 1) fixed unilateral facial contractions especially with lower lips, 2) changes in pattern and side during or between examinations, 3) associated somatoform, or nonphysiologic sensory or motor signs, 4) reduction of the symptoms with distraction, 5) response to pyschotherapy, 6) rapid onset, spontan remissions, 7) normal neurologic examinations (6).

Young female patients with additional findings such as headache, various other functional problems (gastrointestinal, etc.), fibromyalgia, facial pain, depression may be also considered having functional facial movement disorders (6-8).

Our case initially expressed unilateral upper lip deviation, which was altered during further examinations. Additional lower lip contractions were included afterwards. Blepharospasm was never observed during any visit, however she stated having such contractions previously. Having severe facial pain which is commonly unexpected in organic dystonic pathologies and suffering from gastric reflux without any diagnosed gastric pathologies may support the psychogenic nature of the disease in this case.

Despite all, the history of a dental extraction procedure is a confounding information, which we know that these procedures are related to facial dystonia. Nevertheless such procedures are also known to trigger movement disorders especially in vulnerable patients. Psychogenic dystonic pathologies commonly do not involve an initiating voluntary action, but in our case the symptoms were started with mouth closure.

Botulinum toxin (Dysport®) was the choice of treatment in this patient. Post injection outcomes were promising however complete improvement was not observed. The patient was also referred to psychiatric evaluation for additional psychosocial support. Botulinum toxin injection in facial disorders is a common application which is mostly preferred in patients who were unsatisfied with medical treatments or in whom adverse effects of the medications are refrained. It is considered a valuable treatment alternative to medical treatments (9).

Neurotoxin type A is the widely used preparations for treatment. One of the three most commonly used types is abobotulinum toxin A (Dysport, Ipsen Limited, Slough Berkshire, UK). All the neurotoxin A types are reported to have similar mechanisms of action, however their potency and therapeutic profiles may vary and appropriate adjustment of equivalant ratio of doses should be prepared (9). For Dysport, a 1:3 conversion ratio is suggested. Abobotulinum toxin (Dysport) is reported to be more cost-effective (9).

We applied Dysport in our case. It was prepared in similar manners as recommended in the literature. Frozen Dysport was applied, which Thomas et. all. found that it may be safely used for treatments and may be cost-effective due to its cost (10).

CONCLUSION

Facial movement disorders, as well as focal dystonias are commonly managed in neurology clinics. However, head and neck dystonias are also a concern in otorhinolaryngology practice. We aimed to increase the acknowledgement in facial focal dystonias and highlight the efficacy of botulinum toxin therapy in these patients. This case is unique among other focal dystonia cases presented previously in the literature, since it has both organic and psychogenic factors involved. That is why a detailed history and meticulous physical and psychological examinations are essential for better outcomes.

Conflict of Interest: There is no conflict of interest declared by the authors.

Financial Disclosure: This research received no specific grant from any funding agency, commercial or not-for-profit sectors.

KAYNAKLAR

1. Camargo CH, Teive HA. Evolution of the concept of dystonia. Arq Neuropsiquiatr. 2014 Jul;72(7):559-61.
2. LeDoux MS. Meige syndrome: what's in a name? Parkinsonism Relat Disord. 2009 Aug;15(7):483-9. doi: 10.1016/j.parkreldis.2009.04.006. Epub 2009 May 19.
3. LeDoux MS. Dystonia: phenomenology. Parkinsonism Relat Disord. 2012 Jan;18 Suppl 1:S162-4.
4. Pek CH, Seet RC, Yik JH, Lim EC. Orofacial dystonia triggered by mouth closure. Clin Neurol Neurosurg. 2010 Jan; 112(1): 79-81.
5. Sankhla C, Lai EC, Jankovic J. Peripherally induced oromandibular dystonia. J Neurol Neurosurg Psychiatry. 1998 Nov;65(5): 722-8.
6. Fasano A, Valadas A, Bhatia KP, Prashanth LK, Lang AE, Munhoz RP et al. Psychogenic facial movement disorders: clinical features and associated conditions. Mov Disord. 2012 Oct; 27(12): 1544-51.
7. Fasano A, Tinazzi M. Functional facial and tongue movement disorders. Handb Clin Neurol. 2017; 139: 353-365.
8. Colosimo C. Tonic lip deviation as a distinctive form of psychogenic facial dystonia. J Neurol Sci. 2015 Nov 15; 358(1-2): 486-7.
9. Scaglione F. Conversion Ratio between Botox®, Dysport®, and Xeomin in Clinical Practice. Toxins (Basel). 2016 Mar 4;8(3). pii: E65.
10. Thomas JP, Siupsinskiene N. Frozen versus fresh reconstituted botox for laryngeal dystonia. Otolaryngol Head Neck Surg. 2006 Aug;135(2):204-8.