Temporomandibular Joint Dislocation Due to Haloperidol Induced Acute Dystonia: A Case Report and Review of the Literature

Haloperidolün İndüklediği Akut Distoniye Bağlı Gelişen Temporomandibular Eklem Dislokasyonu: Vaka Sunumu ve Literatürün Gözden Geçirilmesi

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Abstract
Dystonia is a movement disorder that causes sustained muscle contractions, repetitive twisting movements, and abnormal postures of the trunk, neck, face, or arms and legs. Antipsychotic agents and neuroleptic agents which are used primarily in the treatment of schizophrenia and also agitated states have some extrapyramidial side effects such as acute dystonia via antagonizing the D_2-dopamin receptor activity. In this case report, we are presenting a case with temporomandibular joint dislocation due to the dystonic reaction induced by haloperidol.

Key words: **Dystonia; Haloperidol; Temporomandibular Joint Disorders.**

Özet
Distoni uzamış kas spazmları, tekrarlayan bükülme haraketleri ile gövde, boyun, yüz, kol ve bacakların anormal postürü ile karakterize bir hareket bozukluğudur. Antipsikotik ve nöroleptik ajanlar D_2-dopamin reseptörler yoluyla distoni gibi ekstrapiramidial yan etkilere neden olurlar. Bu vaka sunumunda, haloperidolün indüklediği temporomandibüler eklem dislokasyonu ile presente olan olguyu bildirmeyi amaçladık.

Anahtar Kelimeler: **Distoni; Haloperidol; Temporomandibular Eklem Bozukluğu.**

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Introduction

Dystonia is a movement disorder that causes sustained muscle contractions, repetitive twisting movements, and abnormal postures of the trunk, neck, face, or arms and legs (1).

The Epidemiological Study of Dystonia in Europe Collaborative Group reported a prevalence rate of 152 per million for primary dystonia, 117 per million for focal dystonia and 57 per million for cervical dystonia (2). However these rates are likely to be underestimated because of the failure to diagnose dystonia particularly in the primary care. Dystonia results from involuntary concomitant contraction of agonist and antagonist muscles, with overflow of unwanted muscle contractions into adjacent muscles. Dystonic movements can be either slow or rapid, can change during different activities or postures, and may become fixed in advanced cases. Dystonia can be classified into four categories as primary dystonia, secondary dystonia, dystonia-plus syndromes and paroxysmal dystonia (3). Primary dystonia have genetic based disorders and cervical dystonia is the most common type of primary dystonia that begins in the middle ages and usually misdiagnosed as musculoskeletal disorders. Primary focal dystonia may also emerge as cranial dystonia which can show itself as blepharospasm, oromandibular dystonia and spasmodic dysphonia, dystonia of the vocal cords. Secondary dystonia may be related to drug induced dystonia, structural abnormalities and heredodegenerative disease and metabolic diseases such as Wilson's disease, Parkinson disease, Huntington's disease, lysosomal storage disease, etc. Dystonia plus syndromes includes the rare genetic disorders like dopa-responsive dystonia and myoclonus dystonia (3).

Although the pathological abnormities are not clear in primary dystonia, neurochemical changes such as imbalance of dopamine transmission may cause primary dystonia as well as dystonia plus syndromes. The lesions in basal ganglions, putamen and thalamus, may also cause secondary dystonia (4).

In the present case, we state an unusual presentation of haloperidol induced acute dystonia presented with temoporomandibular joint (TMJ) dislocation.

Case Report

A 34 years old woman admitted to the emergency department with contraction, pain in her jaw and inability to speak. In her history, she admitted to the emergency department one day ago because of a demoralized mood due to a discussion with her husband. 5 mg diazepam and 5 mg haloperidol via intravenous access were administered in the first presentation of the patient because of an intense anxiety and agitation in the physical examination. One day after haloperidol administration, contractions started in her jaw and an asymmetry occurred in her jaw due to the contraction. She also stated muscle contractions in her back. She had a history of depression and she was taking escitalopram, selective serotonin reuptake inhibitor. There was no other pathological condition in the past medical history of the patient. The physical examination revealed left TMJ dislocation. She denied the other causes which can cause TMJ dislocation such as trauma or yawning before the event. The vital signs of the patient were normal and there was no electrolyte abnormality in the biochemical analyses. TMJ dislocation was relocated by performing downward and backward maneuver after the administration of 30 mg tramadol and 7 mg diazepam. After the reduction of TMJ dislocation, patient's pain ceased and she immediately started to speak.

Discussion

Antipsychotic agents and neuroleptic agents are mainly used in the treatment of schizophrenia but are also effective in the other psychotic and agitated states. Agents with neuroleptic effects have the potential to antagonize the D_2-dopamin receptor activity with the risk of extrapramydial side effects. The extrapramydial side effects of anitpsychotic agents are associated with the potency of D_2 antagonism. Haloperidol is a butyrophenone derivate which has the highest affinity to D_2 receptors when compared to the other dopaminergic and serotonergic binding sites that causes a high incidence of extrapyramydal adverse effects than the other antipsychotic agents (5, 6). In the present case, 5 mg haloperidol was administered via IV route one day ago before the dystonic reaction.

Acute dystonic reaction in the present case can be defined as a drug induced oromandibular dystonia. Oromandibular dystonia causes involuntary clenching, opening or deviation of the jaw. Severe cases may present with jaw pain, dysarthria, dental and temporamandibular traumas such as temporamandibular dislocations. There are three case reports in the literature reporting neuroleptic induced

temporamandibular joint dislocation (7-9). The mechanical energy of oramandibular dystonia may occasionally so high that can cause bilateral TMJ dislocations as the cases reported by Ibrahim and Brooks (7) and Liu (9).

The first step in the treatment of dystonia should be to establish the underlying cause. There has been no known exact therapy existed for the treatment of primary dystonia. Medical therapy for the treatment of drug induced dystonia, secondary dystonia, should be considered. Drugs with anticholinergic effects such as diphenhydramine, a sedative antihistamine, and trihexyphenidyl should be given either parenterally or orally. Anticholinergic agents block the action of the neurotransmitter acetylcholine results deactivating of the muscle contractions. Benzodiazepines are sedative-hypnotic agents binding GABAA receptors which activates the GABA, the major inhibitor neurotransmitter in central nervous system. They can reduce communication between neurons and may also relax muscles. Consequently, benzodiazepines should be an alternative in the treatment of dystonia. Diazepam and clonozepam are the most commonly used benzodiazepines in the treatment of dystonia. Baclofen is another choice used to treat the spasticity. It shows its effects by reducing the release of the neurotransmitters that stimulate muscle activity at the spinal cord level. Baclofen has been used both for primary and secondary dystonia (10, 11). The exact pathophysological mechanisms of dystonia are unclear as aforementioned. Both dopamine agonists and dopamine blocking or depleting agents are used in the treatment of dystonia. Carbidopa or levodopa should be used in generalized or focal dystonia of unknown cause to establish or rule out dopa responsive dystonia. On the other hand tetrabenazine, a presynaptic depleter of dopamine and a weak dopamine receptor blocking agent produced marked improvement in two thirds of patients with focal and generalized dystonia (12). In the present case after the reduction of temporamandibular joint, the dystonic reaction improved and there was no need of additional drug to treat the dystonic reaction. We thought that this improvement may be a result of diazepam administration which is one the alternative drugs in the treatment of dystonia as aforementioned.

Botulinum toxin which inhibits the release of acetylcholine into the neuromuscular junction and surgery in selected patients are also the alternatives that should be performed in the treatment of dystonia.

As a conclusion, acute dystonia induced by either neuroleptic or other drugs may cause serious traumas like TMJ dislocations as presented in this case report. Physicians should be aware of the neuroleptic induced dystonia and admonish the patients about the possible complications.

References

1.Fahn S, Marsden CD, Calne DB. Classification and investigation of dystonia. In: Marsden CD, Fahn S, editors. Movement disorders 2. London: Butterworths; 1987. p.332-358.

2.Epidemiological Study of Dystonia in Europe (ESDE) Collaborative Group. A prevalence study of primary dystonia in eight European countries. 2000; 247:787-792.

3.Tarsy D, Simon DK. Dystonia. N Engl J Med 2006;355:818-829.

4.Marsden CD, Obeso JA, Zarranz JJ, Lang AE. The anatomical basis of symptomatic hemidystonia. Brain 1985;108: 463-483.

5.Potter WZ, Hollister LE. Antipsychotic Agents and Lithium. In: Katzung BG, editor. Basic and Clinical Pharmacology. 10th Edition. New York: McGraw-Hill Company; 2007.

6.Baldessarini RJ, Tarazi FI. Pharmacotherapy of psychosis and mania. In: Brunton LL, Lazo JS, Parker KL, Editors. Goodman&Gilman's The Pharmacological Basis of Therapeutics. 11th Edition. New York: Mc-Graw Hill Company; 2005. p. 461-500.

7.Ibrahim ZY, Brooks EF. Neuroleptic-induced bilateral temporomandibular joint dislocation. Am J Psychiatry. 1996; 153:293-294.

8.O'Connor M, Rooney MD, Nienaber CP. Neuroleptic-induced dislocation of the jaw. Br J Psychiatry. 1992; 161:281-282.

9.Liu ZZ. Bilateral dislocation of mandibular joints induced by haloperidol therapy—a case report. (Article in Chinese) Sichuan Yi Xue Yuan Xue Bao 1985; 16:82-83.

10.Ford B. Intrathecal baclofen for dystonia and related motor disorders. In: Tarsy D, Vitek JL, Lozano AM, editors. Surgical treatment of Parkinson's disease and other movement disorders. Totowa, New Jarsey, USA:Humana Press; 2003. p.287-298.

11.Albright AL. Intrathecal therapy for dystonia in children. In: Brin MF, Comella CL, Jankovic J, editors. Dystonia: etiology, clinical features, and treatment. Philadelphia: Lippincott Williams & Wilkins; 2004. p.87-92.

12.Jankovic J, Beach J. Long-term effects of tetrabenazine in hyperkinetic movement disorders. Neurology 1997; 48:358-362.