Treatment of a Tardive Dyskinesia Patient with Temporomandibular Disorder: A Case Report

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This case report presents a patient with tardive dyskinesia who also suffered from masticatory muscle pain and temporomandibular joint osteoarthrosis. The patient was treated with clozapine in gradually increasing doses and two injections of botulinum toxin type A one year apart. Involuntary movements of mandibular clenching and bruxing disappeared and pain was relieved to a great extent. Reappearances of dyskinetic movements and pain were observed during the follow-up period of 1.5 years. J OROFAC PAIN 2010;24:212–216

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Oral dyskinesias may cause social embarrassment, oral traumatic injury, speech difficulty, chewing and eating disorders, inability to wear removable dentures, or affect professional activities in musicians.[1] Tardive dyskinesia (TD) consists of involuntary, repetitive, purposeless movements that vary in localization and form.[1,2] The lower part of the face is most often involved. Features of the disorder may include grimacing, tongue protrusion, lip smacking, puckering and pursing of the lips, chewing-like movements, and rapid eye blinking.[3,4] Rapid movements of the arms, legs, and trunk may also occur.

TD is associated with the prolonged use of conventional antipsychotic drugs such as phenothiazines, thioxanthenes, and butyrophenones.[2,5] These drugs, such as haloperidol, chlorpromazine, and fluphenazine exert their effects by blocking dopaminergic receptors in the brain, which, in the end, control positive symptoms of schizophrenia, such as hallucinations and delusions.[6,7] Between 15 to 25% of schizophrenia patients using typical antipsychotic drugs develop TD after 1 to 2 years of therapy, although symptoms can start after 3 to 6 months[2,6,7] and, in some cases, even after 3 weeks.[8] Paradoxically, stopping or reducing the drug dosage may unmask symptoms and usually aggravates the disorder. TD is at least three times as likely to occur in patients over 40 years and is twice as common in females than in males.[9]

Not all involuntary movements of the mouth, even in patients who are taking antipsychotic drugs, necessarily reflect TD symptoms.[1,10] For instance, a low percentage of elderly people show spontaneous abnormal movements referred to as oral lingual dyskinesias and senile chorea, although the prevalence increases with concurrent medical illness.[1] The risk of spontaneous orofacial dyskinesia is increased also in edentulous individuals,[11] and about 6% of schizophrenic patients have spontaneous dyskinesia.[12] Patients with diffuse brain pathosis also have an increased risk for TD. Other conditions that can be misdiagnosed as TD include, Huntington's disease, Tourette syndrome, chronic motor tic disorder, Wilson's disease, Meige's syndrome, and habitual oral muscular activity secondary to ill-fitting dentures.[10]

TD is generally pain free but, in a small percentage of cases (3 to 5%), orofacial movements may be so intense and long-lasting that they may cause craniomandibular problems,[1,5] or be accompanied by pain.[13–15] The relentless clenching and bruxing movements typical of this disorder can cause secondary orofacial pain due to the overloading of the masticatory muscles and/or temporomandibular joints (TMJs).[5,16] This tends to lead to the rapid development of internal derangement and subsequent severe degenerative joint disease. Bassett et al[13] reported four patients with complaints of facial pain among 121 TD patients. Ford et al[14] reported 11 patients (out of 204) with painful oral and genital sensory complaints that developed in the setting of TD, tardive dystonia, and tardive akathisia. Hierholzer[15] also described a TD case with tongue pain.

To date, there is no effective treatment for TD.[2] It has been known for many years that clozapine has a diminished risk of inducing TD than the typical antipsychotics.[10,17] On the other hand, botulinum toxin type A (BTA) is used in the treatment of TD as well as in the management of facial pain, severe bruxism and masseter muscle hypertrophy.[1,4,16,18–21] Muscle hyperactivity such as contractures (in the physiological sense), spasms, and focal dystonias are the main indications for BTA therapy.[21,22]

This case report presents the treatment of a TD patient suffering from chronic masticatory muscle pain and TMJ osteoarthrosis.

Case Report

History

A 65-year-old white male was referred to the Department of Dentistry, Dumlupinar University, with a chief complaint of very severe facial pain, particularly in the left masseter region. The medical history revealed that the patient was in treatment for schizophrenia for 8 years. Before dental and craniofacial evaluation, the patient was referred to the psychiatry department. According to the psychiatric assessment, he became angry and eventually violent at the beginning of the complaint. He had sleep disturbances and, soon after, he began having hallucinations. He was diagnosed with schizophrenia and therefore hospitalized for 1 month during which time haloperidol therapy (20 mg/day) was initiated and maintained for 2 months. Thereafter, the dose was reduced to 10 mg/day. With that dose, the symptoms improved in 6 months and he continued taking this dose until the end of the 5th year of treatment. During this period he never had psychotic symptoms, but became more introverted.

Involuntary chewing-like movements appeared at the end of the 4th year of haloperidol therapy and gradually increased in intensity. A diagnosis of TD was made during the 5th year of therapy and the haloperidol administration was replaced with clozapine. The initial dose of 25 mg/day was gradually increased up to 150 mg/day (the optimum dose for schizophrenia treatment is 600 to 800 mg/day). The dose had to be reduced to 50 mg/day for the following 3 years as the patient developed somnolence at the previous dosages. During this period of time, his chewing-like movements persisted. In the 7th treatment year he developed facial pain. He had no extraoral dyskinetic movement. At the psychiatric assessment, during the 8th treatment year, he did not mention any delusion or hallucination. Thus, there were no positive schizophrenia symptoms, such as hallucinations or delusions but negative symptoms such as flattening of effect, poverty of speech, and social withdrawal were apparent.

After the psychiatric assessment, the patient was referred to the Department of Dentistry because of his facial pain. During the visit, he reported that he first noticed pain after he had given up smoking, ie, during the 7th treatment year. Pain started on the left side of the face, spread thereafter to the temporal area and increased progressively in intensity. At the examination time, the pain was fluctuating in intensity and aggravated by chewing and clenching. The patient denied a history of orofacial injury that could have contributed to TMJ dysfunction.

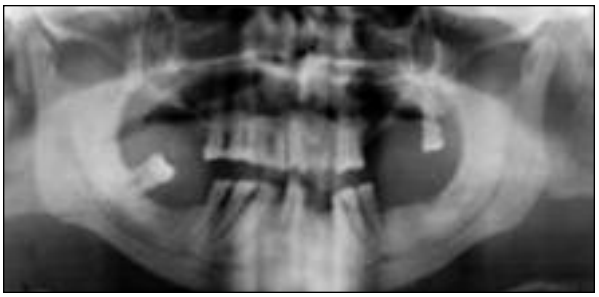

Fig 1 Panoramic radiography demonstrating severe degenerative changes of the right TMJ.

Clinical Examination

Inspection showed severe clenching and bruxing but no extraoral dyskinetic movements. He had a marked bilateral temporal and masseter muscle hypertrophy. The clinical examination revealed a painful maximum opening of 43 mm with a 4 mm deviation to the right side. There was a crepitation sound in the right TMJ and a reciprocal clicking in the left TMJ (opening click at 17 mm and closing click at 14 mm). The left TMJ area was painful to palpation upon opening. The temporal and masseter muscles were tender to palpation on both sides and there was severe pain in the left masseter upon clenching. The intraoral examination revealed a severely reduced dentition. The remaining teeth were severely worn with marked wear facets on the canines and premolars in particular.

Transcranial and panoramic radiographies showed a sclerosis of the posterior slope of the articular eminence on both sides and a flattening of the anterior superior aspect of the right condylar head (Fig 1). The diagnosis was degenerative osteoarthrosis.

Therapy

In the Department of Neurology, 35 units of BTA (BOTOX, Allergan Pharmaceuticals) were injected into each masseter of the patient in order to weaken the muscle and to treat his severe pain. In addition, clozapine was increased to 100 mg/day. These treatments led to a significant decrease in orofacial dyskinetic movements and muscle pain. Three months later, some recurrences of these involuntary movements were seen but without severe pain. Thus, the clozapine dose was gradually increased to 200 mg/day and resulted in a marked reduction in dyskinetic movements and the patient remained stable for the next 6 months. However, the patient started again to suffer from the pain and orofacial dyskinetic movements during the following 3 months. Therefore, after 1 year from the first BTA injection, both masseter muscles were injected with a second dose of 35 units of BTA. This injection resulted in a reduction of the pain and chewing and clenching dyskinetic movements. Since then, the patient has been followed up regularly at 3-month intervals for 1.5 years. During this period, reappearances of dyskinetic movements and muscle pain were observed.

Discussion

In oral dyskinesia patients, orodental complications are likely to be first diagnosed by the dentist. In TD patients, complications such as masticatory muscle hyperactivity, TMJ degenerative diseases, and pain must be evaluated and treated diligently.[1] Other complications include tooth wear, tooth and denture damage, accelerated bone loss in edentulous patients, mandibular luxation, speech impairment, dysphagia, chewing difficulties, and inadequate food intake, and weight loss. Some TD patients experience pain as a result of dyskinetic movements rather than mechanical trauma or oral lesions.[1,9,10] In some instances, the pain is called tardive pain, a complication of chronic antipsychotic exposure.[13–15] Nevertheless, in the present case it is thought that the patient developed severe masticatory muscle pain and TMJ osteoarthrosis as a result of the overload caused by the chronic dyskinetic movements.

Occlusal appliances are used in orofacial dyskinesia patients to prevent tooth wear and TMJ dysfunction.[5,23] Nonsteroidal anti-inflammatory drugs (NSAIDs) together with the application of moist warm heat to the affected side may be beneficial for relieving the pain.[5] However, without eliminating the main cause of the pain or reducing the dyskinetic movements, it is impossible to achieve successful results in these type of patients. Hence, occlusal appliances and NSAIDs are only palliative or supportive treatments for orofacial dyskinesia cases. BTA is a promising alternative for these patients because of its effects on both muscle hyperactivity and pain. It is a powerful neurotoxin selectively taken up by cholinergic nerve terminals, where it prevents the release of acetylcholine by damaging a protein needed to fuse vesicles with the cell membrane.[22] The response to BTA is influenced by the dosage (ie, a dose-dependent effect), the injection technique, and the size (mass) of the injected muscle.[16,18] The muscle weakens within a few days or weeks. The effect of BTA therapy generally lasts for 3 to 6 months and additional injections are needed to have long-term results.[4,22] In the present case, the initial treatment objective was to weaken the masseter muscles by means of BTA. Each masseter was injected with only one dose of BTA. The relapse of orofacial movements and slight pain after 3 months might be attributed to the diminishing effect of the medication. Because clozapine was accompanying BTA treatment and its dose was increased after 3 months, it was impossible to identify the respective effect of the two treatment methods.

There is some evidence that BTA also has a direct analgesic effect, although the analgesic mechanisms are not clear. One possibility is that the BTA not only inhibits the release of acetylcholine but also of neurotransmitters from nociceptive nerve endings.[21,24] Another site of an analgesic action could be the postganglionic sympathetic nerve ending that uses norepinephrine and adenosine triphosphate as transmitters.[21] Inhibition of the release of these transmitters could have an analgesic effect in the presence of sympathetically maintained pain conditions, such as the complex regional pain syndrome.

In the treatment of TD, clozapine is a well-known drug of choice.[2,17] When compared to the atypical antipsychotics, clozapine is thought to have a lower risk of TD. It significantly reduces dyskinetic movements and improves motor symptoms in patients with TD.[1,25] Nevertheless, clozapine is often a later choice because of its potential risk of agranulocytosis. Switching to clozapine earlier can be beneficial in patients with significant TD who agree to the blood-monitoring regime.[17] In the present case, a clozapine dose of 50 to 100 mg/day might not have been sufficient in order to compensate for the dopaminergic hyperactivity in the striatum. After 3 months of treatment, the dose was gradually increased to 200 mg/day and dyskinetic movements decreased considerably. The patient remained stable for the next 6 months. The relief in pain during this period could be attributed to the decreased dyskinetic movements. However, the patient suffered again from considerable pain and dyskinetic movements for the following 3 months. Therefore, 1 year after the first BTA injection, both masseter muscles were injected with a second dose of 35 units of BTA. This injection resulted in a reduction of the pain and chewing and clenching dyskinetic movements.

Instead of clozapine, atypical drugs such as olanzapine and risperidone are also used for the treatment of TD. However, the TD risk associated with these drugs is felt by some investigators to remain significant.[25] Other approaches used in the treatment of pain in TD patients include 2% lidocaine and glyoxide, electroconvulsive therapy, rezerpine, imipramine, propoxyphene, tetrabenazine, and amitriptyline.[14]

When a patient has clinical observable involuntary movements and a positive antipsychotic drug history, the dentist should suspect a TD. The longer the facial dyskinetic movements are allowed to occur without intervention, the greater the risk to develop temporomandibular disorders.[1,5] A patient with these signs might have a neuropsychiatric assessment by a specialist, psychiatrist, or neurologist who is familiar with movement disorders. A psychiatrist will be able to decide whether the drug no longer seems necessary or the psychiatric condition allows a gradual tapering and/or discontinuation of antipsychotic drugs. Thus, a multidisciplinary approach is necessary in treating patients with TD and temporomandibular disorder, and dentists and physicians should cooperate and assess the very early signs and symptoms of temporomandibular disorders.

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