Temporomandibular joint dislocation due to atypical antipsychotic-induced acute dystonia: a case report

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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, arms or legs [Fahn *et al.* 1987]. It may be focal, segmental (multifocal), or generalized and may also be primary or secondary based on their etiology. It may manifest as oculogyric crisis, deviation of eyes in all directions, protrusion of tongue, trismus, lock jaw, torticollis, laryngeal spasm, difficulty in speaking, facial grimacing, opisthotonus, lordosis or scoliosis and tortipelvic crisis. Drug-induced dystonia are secondary dystonias which occur commonly with drugs with antidopaminergic effects such as antipsychotics and metoclopramide [Ropper and Samuels, 2009; Fadare and Owolabi, 2009]. They reportedly arise from a drug-induced alteration of dopaminergic–cholinergic balance in the nigrostriatum (i.e. basal ganglia). Most drugs produce dystonic reactions by nigrostriatal dopamine D2 receptor blockade, which leads to an excess of striatal cholinergic output. High-potency D2 receptor antagonists are most likely to produce an acute dystonic reaction [Marsden and Jenner, 1980]. Agents that balance dopamine blockade with muscarinic M1 receptor blockade, such as atypical antipsychotics, are less likely to cause dystonic reactions [Marsden and Jenner, 1980; Volkow *et al.* 1998]. The incidence of acute dystonic reactions varies according to individual susceptibility, drug identity and dose of the drug. Increased age may carry less risk for the development of dystonia because of diminished numbers of D2 receptors. It occurs commonly to young males who are naïve to antidopaminergic drugs [Volkow *et al.* 1998].

Oromandibular dystonia is one of the focal dystonias, which can be presented as jaw clenching, jaw opening or jaw deviation and leads to impaired

speech and swallowing [Eken *et al.* 2009]. At times, oromandibular dystonia is so severe that it can cause temporomandibular joint (TMJ) dislocation. So far, few case reports have been published about TMJ dislocation due to antipsychotic medications [Ibrahim and Brooks, 1996]. Among antipsychotic-induced TMJ dislocation, case reports were mainly for high-potency first-generation antipsychotics such as haloperidol [Eken *et al.* 2009; Ibrahim and Brooks, 1996; Zakariaei *et al.* 2012]. It has been proposed that drugs such as risperidone and amisulpride which block specific receptors of serotonin (the 5-HT₂ receptors) and dopamine (the D₂ receptors) are less likely to cause these effects [Owens, 1994]. Therefore, the present paper reports a case of oromandibular dystonia with TMJ dislocation with atypical antipsychotics such as risperidone and amisulpride.

Case report

Mr A, a 25-year-old man, was admitted to the psychiatry ward with complaints of suspiciousness, decreased interaction, irritability, decreased sleep, decreased self-care for 2 years and mutism, staring, negativism, slowness with further deterioration in his self-care for 20 days. He was treated with intravenous (IV) lorazepam 6 mg/day in divided doses and IV fluids to maintain hydration. There was no improvement and oral risperidone 2 mg/day was added. Within 12 hours the patient developed lock jaw with pain at the temporomandibular region. On examination, he was having dystonia of the bilateral mandibular region and no dystonia at other parts of the body. His vitals and systemic examination were normal. Risperidone was stopped immediately, and injections of promethazine 50 mg stat and 25 mg twice daily were given. The next day dystonia decreased and the patient was able to close his mouth and

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started eating. Two days later, because the psychotic symptoms persisted, oral amisulpride 100 mg/day was started and on the same day he again developed lock jaw with pain in the TMJ. On examination there was dystonia of the mandibular region. Amisulpride was stopped and injections of promethazine 50 mg IV twice daily were given. Since symptoms did not improve, baclofen 20 mg twice daily was added and an oral maxillary facial surgeon was consulted to confirm the TMJ dislocation. Subsequently dislocation was reduced under local anesthesia. Subsequently pain at TMJ subsided, baclofen 20 mg and lorazepam continued for 5 days. Finally, the patient was treated with oral olanzapine 10 mg, there was no dystonia and his psychotic symptoms improved in 2 weeks.

Discussion

In the present case we reported an unusual presentation of oromandibular dystonia with TMJ dislocation in a patient treated with oral risperidone 2 mg and with amisulpride 100 mg. In severe form dystonia may cause TMJ dislocation. The mechanical energy of oromandibular dystonia may occasionally be so high that it can cause bilateral TMJ dislocations [Ibrahim and Brooks, 1996; Liu, 1985].

That our patient was a young male and drug naive to antipsychotics were the two risk factors for the development of dystonia. Along with family history and personal history of dystonia, young age, male gender, alcohol and cocaine use are reported as risk factors for the development of acute dystonia [Wirshing, 2001; Freed, 1981]. Patients may experience acute dystonic reactions (ADRs) with the administration of just a single dose [Zakariaei *et al.* 2012]. Although ADRs are often idiosyncratic, they may also be dose dependent and occur in overdose [Levine and Burns, 2007]. Among the second-generation antipsychotics, risperidone is likely to cause more neurological side effects, mainly extrapyramidal symptoms (EPS) in a dose >6 mg/day. Similarly amisulpride could also cause EPS and dystonia. There are few case reports on acute dystonia with risperidone and amisulpride [Zones, 2006; Mendhekar *et al.* 2009; Das *et al.* 2008; Jhanjee and Gupta, 2009; Sankhla *et al.* 1998].

In the present case the Naranjo algorithm adverse drug reactions probability scale score was 9, which shows probable to definite causal relation of the drugs with the dystonia. In general, acute dystonias will be completely reversed with drugs and with supportive management. In this patient we managed him with IV promethazine,

anticholinergics and benzodiazepines. Although he responded well for the first time, his response to similar medications was poor. Therefore, we added baclofen 20 mg/day but the TMJ dislocation was difficult to reduce. It was eventually reduced by oromaxillofacial surgery under local anesthesia. Botulinum toxin injection or surgery may be effective in some cases [Ibrahim and Brooks, 1996; Sankhla *et al.* 1998].

TMJ dislocation is a rare complication of oromandibular dystonia but it can cause considerable fear and discomfort to patient and caregivers. So far, TMJ dislocation has been reported with typical antipsychotics such as haloperidol, but to the best of the authors' knowledge, no cases of TMJ dislocation have been reported with atypical antipsychotics such as risperidone and amisulpride. Therefore, the present case demonstrates the possibility of the development of oromandibular dystonia with TMJ dislocation and olanzapine could be an alternative in such patients.

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Conflict of interest statement

The authors declare that there is no conflict of interest.

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