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Case Report

Psychiatry/Mental Health Section

Temporomandibular Joint Dysfunction as the First Presentation of Tardive Dystonia: A Case Report

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ABSTRACT

Tardive dystonia is a chronic, often disabling hyperkinetic movement disorder that develops after prolonged exposure to dopamine receptor-blocking agents, most commonly antipsychotic medications. It is characterised by sustained or intermittent muscle contractions causing abnormal postures or repetitive movements, frequently affecting the craniofacial, cervical, and axial muscles. Unlike acute dystonic reactions, tardive dystonia has a delayed onset and may persist or worsen even after withdrawal of the offending drug, leading to significant functional impairment and reduced quality of life. Diagnosis is particularly challenging when the orofacial region is involved, as symptoms may mimic Temporomandibular Joint (TMJ) dysfunction, including jaw pain, restricted mouth opening, abnormal movements, and facial discomfort, often resulting in misdiagnosis and delayed intervention. This case report describes a 56-year-old woman with a seven-year history of paranoid schizophrenia who developed insidious, progressive oromandibular dystonia following prolonged antipsychotic exposure. She initially presented with jaw pain, progressive trismus, involuntary tongue movements, dysarthria, and marked functional disability, and was treated for TMJ dysfunction with minimal improvement. Comprehensive dental, neurological, and metabolic evaluations excluded alternative aetiologies, while a detailed medication history revealed sustained neuroleptic exposure, leading to a diagnosis of tardive dystonia supported by a high Abnormal Involuntary Movement Scale (AIMS) score. Gradual discontinuation of the offending antipsychotics and switching to quetiapine resulted in complete resolution of dystonic symptoms and sustained psychiatric stability on follow-up. This case highlights a key diagnostic pitfall and emphasises the need for heightened clinical suspicion of tardive dystonia in patients with atypical orofacial symptoms and long-term dopamine antagonist use.

Keywords: Diagnostic challenge, Hyperkinetic movement disorder, Orofacial dystonia

CASE REPORT

A 56-year-old married woman was brought to the Psychiatry Outpatient Department with a 7-year history of illness of insidious onset and episodic course. The illness was initially characterised by third-person auditory hallucinations, associated with persecutory and referential delusions, which gradually progressed over a span of two years associated with sleep disturbances and poor social interaction. After four years of onset of illness, she had consulted a psychiatrist, and was diagnosed with paranoid schizophrenia, and was initiated on risperidone 2 mg/day, which was gradually titrated to 6 mg/day over three weeks. She had responded well to risperidone with reduction in her psychotic symptoms; however, she was poorly compliant and discontinued medications for two years due to poor insight and lack of supervision. She revisited one-year-ago, where she was tried on risperidone and dose was increased to 8 mg/day gradually over 2-3 weeks and she was observed on this dose for three weeks. In the absence of clinical improvement, amisulpride was initiated at 200 mg/day and escalated to 400 mg/day over four days, with a planned gradual taper of risperidone; however, the patient did not adhere to regular follow-up.

While receiving risperidone 8 mg/day and amisulpride 400 mg/day for seven months, she developed dull, aching jaw pain over the preceding five months, with an insidious onset and progressive course. The pain was initially associated with difficulty chewing solid foods, which gradually progressed to reliance on liquid intake alone, accompanied by a progressive reduction in mouth opening over one to two months. Over the subsequent three months, she also reported involuntary tongue movements associated with dysarthria. The muscle contractions were persistent at rest, diminished during sleep, and exacerbated by stress, resulting in significant distress, impaired oral intake, compromised oral hygiene, and reduced social engagement. Consequently, she increasingly isolated herself, avoided community activities, experienced difficulty engaging in telephone conversations, and was unable to perform domestic responsibilities requiring social interaction. Her speech was slurred, strained, and slow, frequently interrupted by tongue spasms that worsened during conversation. She also experienced an unintentional weight loss of approximately 4 kg over five months, without any other apparent systemic changes. She had a four-year history of hypothyroidism, well controlled on thyroxine 50 µg/day. There was no history of dysphagia, dental pathology, malocclusion, denture use, jaw locking episodes, bruxism, or other dystonic manifestations. Family history was non-contributory for psychiatric or neurological disorders. In view of her symptoms, she had consulted a dentist.

Dental examination demonstrated involuntary, patterned jaw movements with sustained jaw opening. Mandibular range of motion was restricted and there was no TMJ tenderness. On occlusal examination there was some secondary dental wear due to repetitive involuntary movements and there was no primary malocclusion. Panoramic radiography revealed no significant abnormalities, and she was diagnosed with TMJ dysfunction and treated with analgesics and a mouth guard for two to three months, with minimal improvement. A neurology consultation was obtained, and detailed neurological examination excluded alternative neurological conditions that could mimic tardive dystonia; the patient was subsequently referred to psychiatry for further evaluation. She subsequently presented to the Psychiatry Outpatient Department.

On general examination, she was thin built, poorly nourished with a body mass index of 17 kg/m^2, without any signs of nutritional deficiencies, and her vital parameters were within normal limits. On inspection, mild deviation of the jaw to the right was noted, with sustained mouth opening of approximately 30-32 mm and

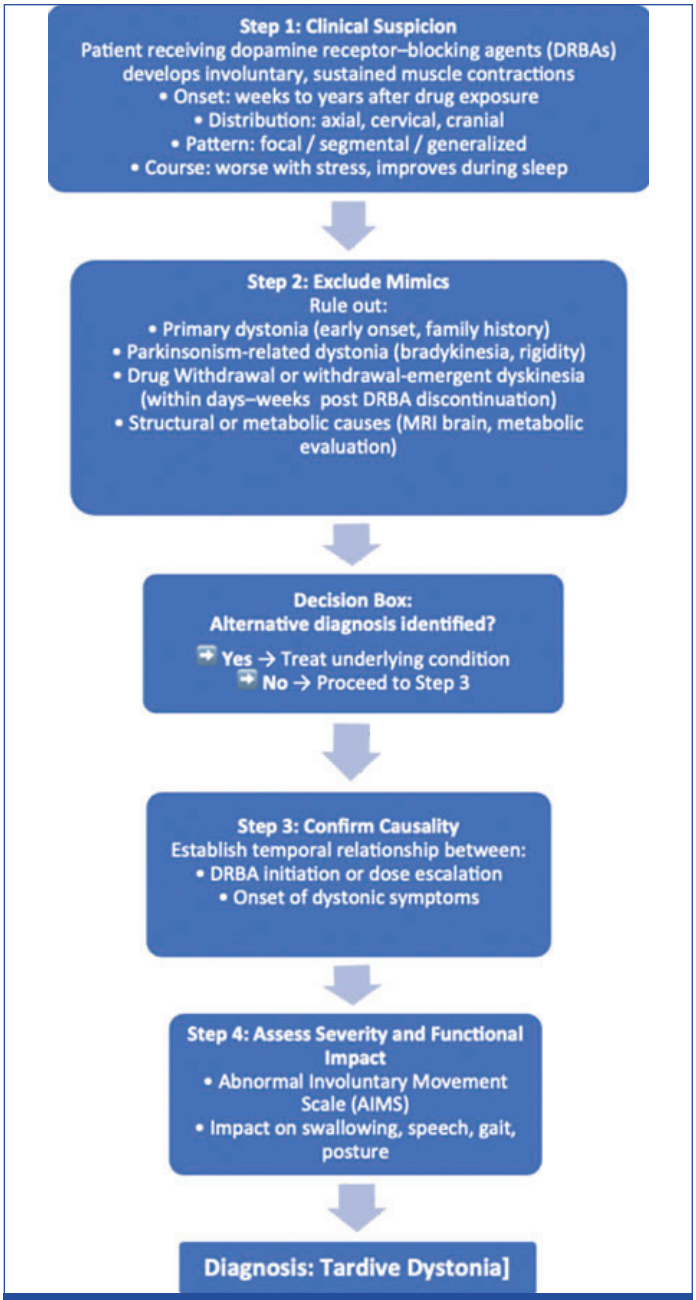
intermittent lower facial muscle contractions during jaw movements, without any facial asymmetry. Tongue protrusion with twisting movements and grimacing were observed, which worsened during speaking and mouth opening, without sensory tricks. On palpation, bilateral masseter hypertrophy was noted with resistance to jaw closure. No crepitus or joint clicking was appreciated in either TMJ.

Comprehensive neurological evaluation revealed intact higher mental functions, including attention, memory, language, and intelligence, with preserved insight into the involuntary movements. Cranial nerve examination demonstrated involvement of the trigeminal nerve (cranial nerve V), with involuntary, sustained jaw opening due to dystonic contractions and mild rightward deviation. Tone of the masseter and temporalis muscles was increased, with preserved muscle strength, intact facial sensation, and a normal jaw jerk. Examination of cranial nerves IX, X, and XII revealed dysarthric speech with lingual dyskinesia. Gag reflex could not be performed and the palate could not be visualised. The remaining cranial nerve examination was unremarkable. On motor system examination, muscle bulk and power were normal. There was increased tone of masticatory muscles without generalised rigidity. There was no evidence of bradykinesia, tremor, or cogwheel rigidity to suggest Parkinson's disease. Sensory examination and deep tendon reflexes were normal and symmetrical. No pathological reflexes were elicited. Gait was normal; there was no truncal or limb dystonia in isolation. There was no buccal dyskinesia, cervical dystonia and limb stereotypies. There were no cerebellar findings. Frontal release signs were absent and no focal neurological deficits was observed. Ophthalmological examination revealed no Kayser-Fleischer rings.

On assessment with the AIMS [1], she scored 15, with a severity score of 4 and severe incapacitation due to abnormal movements. She was aware of the movements and experienced significant distress. On the World Health Organisation Disability Assessment Schedule (WHO DAS) [2], she scored in the extreme range for getting along, severe for life activities and participation, and overall demonstrated moderate disability.

Laboratory investigations, including complete blood count, liver and renal function tests, fasting blood glucose, thyroid function tests, serum electrolytes, lipid profile, iron studies, vitamin B12, and folate levels, were within normal limits. Serum copper and ceruloplasmin levels were also normal. Magnetic Resonance Imaging (MRI) of the TMJs was considered; however, in view of the late onset, absence of focal neurological signs, symmetric involvement, and lack of rapid progression, the investigation was deferred.

The differential diagnoses considered included acute dystonic reactions, tardive dyskinesia, Parkinson's disease, bruxism, TMJ disorders, mandibular clonus, orofacial automatisms, Wilson's disease, and idiopathic oromandibular dystonia. Acute dystonia was considered unlikely due to the insidious onset and chronic course of the symptoms. Parkinson's disease was excluded as there was no evidence of parkinsonian features and the minimal jaw tremor was excluded in view of sustained muscle contractions. Tardive dyskinesia was deemed less likely given the absence of typical choreiform movements such as lip-smacking or chewing, with only minimal recent lingual involvement. Bruxism was ruled out as it was usually sleep-related and characterised by repetitive clenching rather than sustained jaw posturing. Orofacial automatisms and mandibular clonus were excluded due to preserved awareness and absence of brief, shock-like movements, respectively. Wilson's disease was unlikely based on age at presentation and a normal metabolic evaluation. Although idiopathic oromandibular dystonia was considered, the history of prolonged exposure to dopamine receptor-blocking agents favoured a diagnosis of tardive dystonia. A stepwise diagnostic approach to tardive dystonia, integrating clinical history, phenomenology, and exclusion of secondary causes, is summarised in [Table/Fig-1].



[Table/Fig-1]: Algorithm outlining the diagnostic approach to tardive dystonia.

In accordance with the Schooler criteria [3], the combination of sustained neuroleptic exposure, exclusion of alternative causes, and a total AIMS score of 15 supported the diagnosis of tardive dystonia.

Management involved the gradual tapering and discontinuation of risperidone and amisulpride over period of 4-6 weeks, with introduction of quetiapine as an alternative antipsychotic agent. She was asked to come for frequent follow ups to monitor for worsening of her dystonic as well as psychotic symptoms. Quetiapine was selected, given the evidence from clinical reviews and case series suggesting improvement in tardive dystonia after switching to antipsychotics with lower dopamine D2 receptor affinity, while preserving antipsychotic efficacy [4,5]. According to Jinnah HA and Factor SA, benzodiazepines are GABAergic agents used as adjunctive therapy in dystonia, where they may reduce dystonic movements by enhancing central inhibitory neurotransmission; accordingly, clonazepam 0.25 mg was added as an adjunct in the present case. [6]. Quetiapine was titrated from 12.5 mg initially for two weeks to 50 mg daily, resulting in a progressive reduction of dystonic muscle spasms, improved jaw mobility, and decreased orofacial pain. By four weeks, the patient's pain had improved, with initiation of semi solid foods, and improvement in her dysarthria, and her mouth opening had improved to 36-38 mm. At eight weeks, she was asymptomatic and able to resume normal eating and engage in

social activities. Her mouth opening had improved to 45-47 mm.

She remained free of both psychotic and dystonic symptoms during one year of follow-up and she was planned for regular follow-ups to assess any worsening and to ensure compliance.

DISCUSSION

Tardive dystonia is a chronic, often disabling hyperkinetic movement disorder that develops after prolonged exposure to dopamine-receptor blocking agents, most commonly antipsychotics. It is characterised by sustained or intermittent involuntary muscle contractions producing abnormal postures or patterned dystonic movements. Unlike tardive dyskinesia, which is typically choreiform, tardive dystonia predominantly manifests as persistent dystonic posturing involving the cervical, axial, or oromandibular musculature [7]. The prevalence of tardive dystonia among patients exposed to antipsychotics is estimated at about 2%, accounting for approximately 10-20% of tardive syndromes, while the overall prevalence of tardive syndromes averages around 20%, with variability attributed to differences in study populations, duration of exposure, and diagnostic criteria [8,9].

The pathophysiology of tardive dystonia is multifactorial and incompletely understood. Chronic dopamine D_2 receptor blockade is believed to induce maladaptive neuroplastic changes within basal ganglia circuits, resulting in receptor hypersensitivity and disrupted nigrostriatal signalling [10]. Additional contributory mechanisms include dysfunction of gamma-aminobutyric acidergic and cholinergic neurotransmission, oxidative stress, and abnormalities within cortico-striato-thalamo-cortical networks [8-10]. Comparative studies have shown that while tardive syndromes share several risk factors, tardive dystonia tends to occur at a younger age and shows a male predominance compared with tardive dyskinesia [11].

Second-generation antipsychotics are not devoid of risk for tardive syndromes. Case reports have described risperidone-induced tardive laryngeal dystonia [12] and amisulpride-associated tardive dyskinesia at both standard and low doses in antipsychotic-naïve individuals [13]. These findings reinforce the need for vigilance during long-term antipsychotic treatment.

Oromandibular tardive dystonia may closely resemble temporomandibular disorders, particularly when jaw pain, limited mandibular excursion, or apparent jaw dislocation predominates, frequently leading to misdiagnosis and delayed neurological referral. Temporomandibular disorders are primarily mechanical in nature and are characterised by joint tenderness, clicking, reproducible pain with mastication, or identifiable structural abnormalities, whereas oromandibular dystonia is defined by involuntary, patterned, and sustained jaw movements that may occur at rest or during speech, often accompanied by sensory tricks and overflow phenomena, with typically normal TMJ imaging [14]. This diagnostic challenge has been illustrated in reports by Nikunj A et al., and Prabhakar V et al., [15,16]. These cases highlight the necessity of evaluating alternative aetiologies in instances where orofacial symptoms are persistent, atypical, or accompanied by stereotyped involuntary movements.

Prior to establishing a diagnosis of tardive dystonia, several differential diagnoses must be excluded. TMJ dysfunction presents with mechanical symptoms but lacks the stereotyped involuntary movements characteristic of dystonia [14]. Bruxism and myofascial pain syndromes may mimic jaw clenching but are phenomenologically distinct from dystonia and lack sustained dystonic posturing [13]. Primary (idiopathic) oromandibular dystonia usually has a gradual onset, may be familial, and occurs in the absence of prior neuroleptic exposure [17]. Acute drug-induced dystonia typically develops within hours to days following antipsychotic initiation or dose escalation and is usually reversible with anticholinergic therapy [18]. Structural or metabolic neurological conditions, including stroke, basal ganglia lesions, and Wilson's disease, should also be excluded, particularly in the presence of red flags such as acute onset, asymmetry, or focal neurological deficits; in oromandibular dystonia, a systematic diagnostic approach including clinical assessment and relevant investigations (e.g., MRI, ceruloplasmin) is essential to rule out these secondary causes [19].

Early recognition and timely intervention may lead to gradual clinical improvement. Focal oromandibular dystonia often shows a favourable response to antipsychotic modification, botulinum toxin injections, and adjunctive pharmacotherapy, with symptom stabilisation occurring over time [19].

The occurrence of tardive dystonia despite second-generation antipsychotic use highlights the need for judicious prescribing, sustained monitoring, and consideration of adjunctive therapies beyond antipsychotic modification, including Vesicular Monoamine Transporter-2 (VMAT-2) inhibitors such as valbenazine and deutetrabenazine, which reduce presynaptic dopamine availability and are increasingly used off-label [20]. Botulinum toxin injections are considered first-line therapy for focal and segmental dystonia, exerting their therapeutic effects through targeted chemodenervation at the neuromuscular junction. Pharmacological agents, including anticholinergic medications and benzodiazepines, may be employed as adjunctive treatments, although their clinical utility is often constrained by limited efficacy and tolerability [6]. In severe, refractory cases of dystonia including tardive dystonia, deep brain stimulation of the globus pallidus internus has demonstrated sustained clinical benefits in motor symptom reduction and disability [20].

In this case, quetiapine - a second-generation antipsychotic with relatively low dopamine D_2 receptor affinity-was selected to minimise further dopamine blockade and reduce the risk of worsening dystonia. An integrated management strategy allows for effective psychiatric treatment while optimising movement disorder control, underscoring the importance of early recognition and timely intervention in achieving favourable outcomes while preserving psychiatric stability.

CONCLUSION(S)

Tardive dystonia should be considered in patients on long-term antipsychotic therapy who present with TMJ-like symptoms. Careful review of medication history and neurological signs is essential for accurate diagnosis. Early recognition and timely treatment modification can significantly improve outcomes and prevent persistent orofacial dysfunction.

Declaration of the author: The authors confirm that informed consent was obtained from the patient for publication of clinical information and images. The patient has provided consent for the use of her clinical data in this report and understands that identifying details will not be published; however, while all reasonable efforts will be made to ensure anonymity, complete confidentiality cannot be guaranteed.

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