# Antipsychotic-Induced Tardive Lingual Dystonia Presenting With Bradycardia

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Dystonia is a type of movement disorder wherein a muscle or group of muscles go into sudden sustained contraction resulting in abnormal postures. Dystonia can be classified into focal dystonia, segmental dystonia, multifocal dystonia, hemidystonia, and generalized dystonia depending on the distribution of dystonia. Typical antipsychotics are known to cause drug-induced dystonia, which is focal in distribution and most commonly affects the head and neck area followed by the arms and legs.[1]

Typically, acute dystonia appears within 5 days of starting a dopamine blocker,[2] and tardive dystonia appears much later in the course of treatment but within a year of starting dopamine blockers.[3] Tardive dystonias are usually included under the encompassing term *tardive syndromes*, which also includes the more common orofacial dyskinetic phenotype among others. It is defined as persistent dystonia with onset during or following the use of antipsychotic drugs, in a patient with no family history of dystonia and a negative workup for other secondary dystonias.[4] Tardive dystonias have an incidence of 5% per year.[5] Due to rarity of the occurrence of tardive dystonias in the population, there is not much clarity about its treatment. Here, we present a case of antipsychotic-induced lingual dystonia that led to hemodynamic changes in an individual, which is a rare occurrence.

## Case Report

A 15-year-old boy presented with complaints of persistent tongue protrusion for 1 month. On inquiry, he had been diagnosed with mild intellectual disability disorder and was receiving tablet risperidone 6 mg/d for 1 year for complaints of behavioral disturbances. The tongue movements started around 1 month ago, which the patient described as partially being under his control but that he could not control the movements for a long time. Protrusion of the tongue was not present when he was asleep, and it interfered with his speech to some extent. At the first visit, he was kept in the emergency ward, and he was given injection promethazine 25 mg intramuscular immediately and observed for 4 hours. There was not much improvement. Later, tablet promethazine 25 mg twice/day was added to his existing prescription, and he was sent home. The patient came back after 10 days with no improvement in protrusion of his tongue. He was admitted to the hospital ward and started on injection promethazine 25 mg twice/day. On the second day of admission, it was noted that his pulse and blood pressure were always on the lower side, in the range of 40–55 bpm and 80/50 mm Hg, respectively. On physical examination, orbicularis oris muscle, buccinator muscle, base of the jaw muscles, bilateral sternocleidomastoid muscles, and platysma and intercostal muscles were prominent. His cardiac evaluation was done using echocardiography, which revealed no abnormality. Tablet risperidone was stopped, and injection promethazine 25 mg twice/day was continued. Over the next 2 days, protrusion of the tongue reduced, and his blood pressure and pulse rate returned to normal range, explaining temporality.

## Discussion

Lingual dystonia is a rare oromandibular dystonia. It involves tongue protrusion or curling and can also cause drooling of saliva. It most commonly occurs with speaking and can cause problems with speech, eating, and swallowing.[6] The spasm-caused movements impacted the patient's inspiration due to total closure of the airway on the palate and pharynx after inspiration onset. The persistent lingual dystonia led to repeated Valsalva maneuvers.

Valsalva maneuver consists of a voluntary forced expiratory effort against a closed airway (eg, closed mouth and nose). It can lead to intricate, temporary shifts in cardiovascular and neuro-hormonal functions due to heightened intrathoracic and intra-abdominal pressure, resulting in corresponding fluctuations in blood pressure. There are 4 phases in the Valsalva maneuver, based on the hemodynamic changes.[7,8]

A study by Aebischer et al,[9] which studied the 2D echocardiography dynamics of ventricular interdependence of Valsalva, helped us understand that during phase II of the Valsalva, positive intrathoracic pressure leads to a reduced venous return to the heart. Due to reduced venous return and following reduced preload, stroke volume falls; this leads to a fall in blood pressure. The vagal withdrawal followed by increased sympathetic discharge leads to tachycardia, increased cardiac output, and vasoconstriction, which leads to the recovery of blood pressure to normal values in healthy individuals. Repeated or prolonged Valsalva maneuvers can sustain this reflex response, leading to prolonged bradycardia. Due to the antipsychotic-induced lingual dystonia, the recovery phase was compromised, leading to persistent low blood pressure and low pulse.[9]

It is important to note that while bradycardia is a potential response to the Valsalva maneuver, not everyone will experience it, and individual responses

can vary. Numerous variables, such as blood volume, preceding rest period, cardiac and peripheral sympathetic functions, as well as norepinephrine response, influence the Valsalva maneuver.

There is limited recent literature on effects of repeated Valsalva causing hemodynamic changes in individuals. One study by Khurana and Mittal[10] studied the effect of oral leak size on hemodynamic changes. Results showed that increased neuromuscular effort during the maneuver leads to a rise in overall phase II mean blood pressure, which was positively correlated to oral leak size. These findings were contrary to the findings seen in our case.

This is a rare case in which use of an antipsychotic led to repeated involuntary Valsalva resulting in prolonged hemodynamic compromise. This aspect of lingual dystonia causing hemodynamic changes in individuals has received less attention. Awareness among clinicians regarding this acute side effect of antipsychotics can facilitate early detection and prompt management and lead to improved patient outcomes.

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