Acute Haloperidol Induced Oro-Mandibular Dystonia


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**Objective**
To report a case of acute oro-mandibular dystonia secondary to haloperidol occasional intake.

**Case summary**
A 49-year-old white woman was referred to the emergency room with severe involuntary tongue protrusion. She was in severe distress, oral examination revealed sustained and involuntary protrusion of a turgid and purplish tongue, speech and swallowing difficulty. After two or three minutes of continuous tongue protrusion, masseter contraction appeared for one minute with gnashing of teeth and jaw involuntary forced closure. One of these spasms caused tongue biting with a wound on the center of her tongue requiring suture. The symptoms disappeared within five minutes but recurred five minutes later. Between two contractions the patient was able to move (pull out-put in) her tongue with moderate difficulty and she was able to speak. These clinical features recurred several times during the emergency monitoring. The clinical presentation was interpreted as dystonia and was successfully treated with diazepam 10 mg i.v.. Eighteen months before she had presented a similar spontaneously resolved episode interpreted as tongue angioedema. She did not have any history of head trauma, seizure, infections, substance abuse or family history of major illness or motor disorder, except for her mother’s psychosis. She was not on any home-based therapy. Neurological exam was normal except for tongue dystonia. Rigidity was present only in the masseter muscle, while limbs and trunk were free from rigidity, bradykinesia and tremors. Routine haematology exams, oxygen saturation, CT and MRI brain scan were normal. The day after admission, she revealed that a few hours before the onset of dystonia she had felt anxious and had taken unspecified dosage of haloperidol prescribed to her mother. Moreover, she reported to have taken haloperidol also a few hours before the onset of the previous episode of tongue protrusion.

**Conclusion**
This is an example of acute drug induced oro-mandibular dystonia, appeared in the early hours after occasional haloperidol intake. Approximately only 10% of acute drug-induced dystonias may appear in the early hours of treatment, 90% within the first three days. The pathophysiologic mechanism seems to be a hyperactivity of dopaminergic transmission in the basal ganglia, which occurs during reduction in drug concentration. Management and treatment are often difficult. Tetrabenazine and trihexyphenidyl can be an useful treatment but they are ineffective in many cases. Benzodiazepines, clonazepam and diazepam, are often used with mild symptomatic relief. Drug-induced acute dystonic reaction is a common presentation at emergency department (1) and poses a very serious challenge because of the high probability of misdiagnosis like allergy with swollen tongue, which may delay intervention (2). In the management of acute dystonias, a high level of clinical suspicion is crucial and an accurate drug history is necessary.

**References**