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LETTER TO THE EDITOR

An unusual dystonia accompanied by tongue tremor

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Dear Editor,

An acute dystonic reaction is a sustained, recurrent muscle spasm characterized by bending, squeezing, pulling, and painful postures, especially affecting the muscles of the face, neck, and back. The primary causes of dystonia include pre-existing neurodegenerative disorders and adverse effects of medication (1, 2). Orolingual tremor is defined as rhythmic, involuntary, oscillatory movements involving the jaw, tongue, pharynx, and lower face, which may result from lesions in the brainstem, extrapyramidal system, or cerebellum, as well as ischemic events, neurological damage associated with metabolic disorders, neurodegenerative diseases, and adverse drug effects (1). There is also a strong association between dystonia and tremor (3). The prevalence of tremor in dystonia has been reported to range between 14% and 86.67% (4). While cases of drug-induced tongue tremor in adults are documented in the literature, there are no reports of drug-induced tongue tremor or dystonia accompanied by tongue tremor in children in the literature. Here, we describe an unusual case of dystonia accompanied by tongue tremor associated with low-dose antipsychotic treatment, which we believe contributes to the literature. Informed consent was obtained from both the patient and her parents.

A 14-year-old girl was under follow-up in our outpatient clinic with a diagnosis of acute stress disorder and depression. The patient weighed 58 kilograms, and risperidone was initiated at 0.5 mg per day. After two weeks, the risperidone dose was increased to 1 mg per day, and sertraline at 25 mg per day was added. Three weeks after the risperidone dose increase, the patient presented to the emergency department with complaints of numbness in her tongue and face, which progressed to convulsions on the right side of her body. On examination, she exhibited dyskinetic movements and facial grimacing, along with convulsions on the left side of her neck. The patient received an intramuscular injection of biperiden 5 mg, but due to an insufficient response, intramuscular diazepam 10 mg was subsequently administered, resulting in the regression of all symptoms. Sertraline and risperidone were discontinued for the patient, who had no known additional medical conditions. She was admitted to the pediatric ward for continued monitoring. That night, unilateral neck contractions and tongue tremor were observed; oral lorazepam 2.5 mg was administered, leading to resolution of the contractions, though the tongue tremor persisted. A neurology consultation was requested. Cranial magnetic resonance imaging (MRI), electroencephalogram (EEG), complete blood count, fasting blood glucose, electrolyte levels, liver, kidney, and thyroid function tests, as well as vitamin B12 and folic acid levels, were all within normal ranges. At her follow-up outpatient clinic visit after discharge, the tongue tremor was noted to persist. When medication use was detailed in the interview, the patient stated that she may have taken risperidone 2 or 3 times a day, as she occasionally forgot her doses on some days, and that she administered the medication herself. The case was evaluated in terms of suicidal ideation or attempt.

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Based on the history obtained from the patient and her family, it was determined that the situation was not an act of self-harm. At a follow-up visit three weeks later, it was noted that the contractions had not recurred, and the tongue tremor had disappeared a few days after the last visit.

The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) defines drug-induced acute dystonia as an unusual and prolonged contraction of muscles that develops following the initiation of drug treatment or a change in drug dosage. Although the physiopathological mechanism is not fully understood, it is suggested that an imbalance between the striatal dopaminergic and cholinergic systems, resulting from a sudden and excessive dopamine receptor blockade, may be responsible. Drug groups reported to cause acute dystonia in the literature include antipsychotics, antiemetics, antidepressants, antiepileptics, and antimigraine drugs (2). The patient was known to have been receiving sertraline and risperidone treatments, both of which can cause movement disorder side effects. Given that the dose of risperidone was increased two weeks prior to the onset of contractions and that the patient may have taken 2–3 mg of risperidone on some days, it was concluded that her condition was likely an acute dystonia side effect resulting from risperidone treatment.

A review of the literature revealed cases of tongue tremor in adult patients due to levosulpiride (5, 6). However, no instances of drug-induced tongue tremor associated with dystonia have been documented in children. There are, however, reports of pediatric cases with isolated tongue tremor in the absence of drug use or neurological issues (7, 8). In the differential diagnosis, conversion disorder, tardive dyskinesia, and other medical conditions were ruled out. Conversion disorder was excluded because our patient had responded well to sertraline and risperidone, the contractions appeared during a period of reduced mental symptoms, and there was no history of conversion attacks.

For the development of dystonia due to antipsychotic drug use, certain risk factors are recognized, including the patient's young age, a first episode of mental disorder, first exposure to an antipsychotic drug, the high potency of the antipsychotic used, high-dose initiation, and recent dose increases (9). In our case, the patient's young age, first episode of mental disorder, first exposure to antipsychotic drugs, and the high potency of risperidone likely contributed to her predisposition to dystonia.

It is unclear whether tremor in dystonia has a distinct pathophysiology independent of dystonia or whether they share some mechanisms. Neurophysiological studies on tremor in dystonia have shown that it shares similar physiology with dystonia (10). Although risperidone is associated with the highest risk of extrapyramidal symptoms (EPS) among atypical agents, risperidone-induced dystonia is linked to alterations in the basal ganglia motor circuit due to dopamine receptor blockade (11). When dopamine D2 receptors in the striatum are blocked, movement disorders may occur, leading to a relative reduction in thalamocortical activity. Similar to dystonia, dysfunction in the basal ganglia network and the cerebello-thalamo-cortical network has been implicated in the pathophysiology of various tremors.

While dystonia is considered a common side effect of antipsychotic drugs, in our case, the clinical presentation was termed "unusual dystonia" to denote a more specific form of the disorder. In this instance, dystonia manifested as torticollis with a 1 mg/day antipsychotic treatment. The neck contraction initially started on the right side, then became bilateral, and was accompanied by a rare tongue tremor, thereby defining the presentation as unusual dystonia. There are a limited number of case reports in the literature on this topic, and it is thought that our case will contribute valuable insights.

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