Sulpiride-Induced Dystonia in Tourette Syndrome; A Stroke Mimicking Presentation

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Case Report

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Abstract

We report a case of sulpiride induced dystonia, which was misleading to identify because of the acute onset during exercise mimicking stroke in a Tourette syndrome patient. After excluding artery dissection through MR-Angiography, Biperiden was infused, which lead to the immediate disappearance of the dystonic symptoms.

Case

A 23-year-old male Caucasian patient was admitted to the emergency department because of an acute onset of difficulty of articulation and weakness of the left arm and throat and neck pain, which occurred while playing tennis. On examination, the symptoms disappeared completely. Except for since childhood known Tourette syndrome (TS), the patient reported no related medical history. Vital signs showed no abnormality.

We initially decided to waive CT-Angiography because of the disappearance of the symptoms and the young age. We so carried out only a native CT Brain and cervical spine, which showed no intracerebral hemorrhage, infarction, tumor, or atlantoaxial subluxation. Subsequently, we admitted the patient to the Stroke Unit for monitoring and Ultrasonography, suspecting a transient ischemic attack (TIA) on top of a possible artery dissection. During the Ultrasonography of the carotid and vertebro-basilar vessels, where the subclavian steal phenomenon was excluded, the patient developed an acute difficulty in articulation and weakness of the left arm again. The patient neurologically showed dysarthria and drift in raising the left arm combined with increased clonic tics under that stressful situation. Although the drift was not convincing, we carried out MRI showed no diffusion/perfusion restriction or altered signal intensity areas. MR-Angiography of the Brain (including Fat suppression imaging) showed no artery dissection or any related abnormality as well. The laboratory values were in the normal range.

After excluding stroke in the emergency, a detailed history taking was carried out. There was no family history of hypercoagulability, cardiac diseases, or stroke in the young. It revealed that the patient started visiting a new neurologist three weeks before and received Sulpiride against Tourette syndrome. Over those three weeks, the patient received a rapid dose-escalating of up to 800 mg daily. A sulpiride induced dystonia, clinically masked by dystonic and clonic tics of the known Tourette syndrome, was suspected. We injected the patient slowly with Biperiden (Anticholinergic Agents) 5 mg intravenous under monitoring. The patient reported an immediate disappearance of the articulation difficulties, movement of the left arm, cervical and neck pains. Clinically there was no dysarthria anymore. Electroencephalography showed a normal Alpha rhythm and no epileptic potentials. The Patient didn't develop such attacks anymore and was discharged the next day.

Discussion

Tourette Syndrome is characterized by chronic motor and vocal tics presenting during childhood, which is more common in males [1]. Our patient had already a Tourette syndrome with mixed motor tics and no vocal tics. Management of Tourette syndrome includes behavioral therapy [1] and medical therapy, which is reserved for severe, refractory cases or social problems due to tics. [2] One of the common medications for the treatment of Tourette syndrome and other tic disorders, according to European clinical guidelines, is sulpiride [2]. Sulpiride belongs to the class of Benzamides, atypical neuroleptic, and primarily a selective dopamine D2 antagonist. In our case, sulpiride was dosed rapidly up to 800mg daily over only three weeks instead of starting with 50-100 mg daily and slowly dosing over many weeks [3]. In our case, sulpiride was dosed rapidly up to 800mg daily over only three weeks.

Acquired dystonia forms can be due to perinatal brain injury, infections, toxicities, vascular diseases, neoplasms, or drug-induced by anticonvulsants, calcium channel blockers, and neuroleptics like dopamine receptor blocking drugs including Sulpiride [4].

Other possible causes of a similar presentation of our case include the subclavian steal phenomenon, resulting in a similar pattern of symptoms like acute weakness and severe dysarthria following exertion of the upper limbs [5]. Subclavian steal phenomenon could be diagnosed during the routine vertebrobasilar sonography. A preceding seizure with regressive weakness or aphasia in the postictal state frame should also be clinically and electroencephalographically excluded [6]. Atlantoaxial subluxation following a possible head trauma during playing could present itself with neck pain, acute weakness, and severe dysarthria that may regress on changing head position [7]. CT- cervical spine can diagnose or exclude this state.

After excluding the other possible causes, we hypothesized that the patient started to suffer from dystonia, including the cervical region, larynx, and left arm because of the rapidly increased dose of sulpiride. The dystonia was masked by the dystonic and clonic tics of the known Tourette syndrome in the background. Only one case in the literature mentioned a similar case of Sulpiride-induced tardive (after five years of sulpiride intake) dyskinesia in a person with Tourette syndrome. [8]

As Biperiden is indicated to treat neuroleptic-induced acute dystonia [9], we injected the patient as mentioned to confirm the diagnosis and manage the patient. Our case is of particular interest for neurologists and psychiatrists because it highlights that treating TS with sulpiride should consider the possibility of developing dystonia in their patients.

Conclusion

Neuroleptic induced dystonia in patients with Tourette syndrome can be challenging to identify since dystonia symptoms can be mimicked or masked by symptoms of TS. A thorough patient and medication history was the key to recognizing an underlying Sulpiride induced dystonia in Tourette syndrome. The

diagnosis could be confirmed after injection of Biperiden as treatment of neuroleptic-induced acute dystonia as mentioned.

Declarations

Conflict of Interest: None.

Consent We certify, that the patient consented to participate and publish their case and clinical data.

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