Piriformis Syndrome Misdiagnosed as Antipsychotic-Related Dystonia

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ABSTRACT:
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Dystonia associated with antipsychotics is common and well known. The current case presents a 20-year-old male patient during the manic phase of bipolar disorder, developing piriformis syndrome (PS) under antipsychotic medication treatment. Initially, the anteflexion position of the patient was considered to be dystonia secondary to antipsychotic treatment and treated accordingly with biperiden, benzodiazepine, baclofen, tetrabenazine, electroconvulsive therapy (ECT), and botox; however, the patient did not respond to these treatments. Subsequently, after consulting the algology department, PS was diagnosed as a distinct entity. The patient recovered from the symptoms of PS upon the injection of bupivacaine and triamcinolone into his piriformis muscle. This case reminds us that the accuracy of the diagnosis in the treatment of possible drug-related side effects should be carefully reviewed, especially in resistant cases.

Keywords: antipsychotic, dystonia, piriformis syndrome

INTRODUCTION

Movement disorders are common side effects of antipsychotics due to blockage of dopamine D2 receptors, and these are well known to clinicians. One of them is acute dystonia, which is characterized as “sustained abnormal postures or muscle spasms that occur within a few days of starting or rapidly raising the dose of the antipsychotic, or of decreasing the medication used to treat or prevent acute extrapyramidal symptoms (e.g., anticholinergics). The prevalence is quite high, varying widely, from 2% to 90%¹. The prevalence of tardive dystonia is 0.4-21.6% (mean 5.3%) in patients treated with antipsychotics². Acute dystonia is generally improved by anticholinergic treatment, but there may be some difficulty in treating chronic or late dystonia³. Except for being a side effect of antipsychotics, there are numerous causes of dystonia; including idiopathic dystonia, Parkinson disease, Multiple Sclerosis, Huntington disease, Wilson disease, Rett syndrome, ataxia telangiectasia, homocystinuria, Hartnup disease, tyrosinosis, metachromatic leukodystrophy, gangliosidosis, Lesch-Nyhan syndrome, vitamine E deficiency, perinatal cerebral events, viral encephalitis, Reye syndrome, cerebral tumors, arterio-venous...
malformations, cerebral trauma, cerebral operations, certain toxins, levodopa, bromocriptine, metoclopramide, fenfluramine, flecainide, ergot alkaloids, anticonvulsants, and calcium channel blockers.

Piriformis syndrome (PS) is a sciatic nerve neuropathy developed through compression of the sciatic nerve by the piriformis muscle (PM). PS may be caused by inflammation, a tumor near the PM, or anatomic variation of the PM and sciatic nerve, micro/macro trauma to the PM, edema, and local ischemia. PS is characterized by hip and buttock pain. This syndrome is not often considered in clinical situations because its presentation may be similar to that of lumbar radiculopathy, primary sacral dysfunction, or innominate dysfunction. The most common symptoms of patients with PS is increasing pain after sitting for longer than 15 to 20 minutes. Many patients complain of pain above the PM. Difficulty in walking and pain during internal rotation of the ipsilateral leg may exist. Reported incidence rates for PS among patients with lower back pain vary widely, from 5% to 36%.

Here, we report the management of a patient who had initially been misdiagnosed with antipsychotic-related dystonia and recovered after being treated for PS.

**CASE REPORT**

A 20-year-old male patient hospitalized with typical manic symptoms that lasted for one week. He had had three episodes of hypomania previously and recovered without treatment. Daily doses of 6 mg risperidone, 900 mg lithium, 2 mg biperiden, and 1 mg clonazepam treatment were initiated. Two weeks later, risperidone was increased to 8 mg and lithium was increased to 1200 mg. The patient recovered and was discharged. Risperidone and clonazepam doses were reduced at follow-up.

The patient was doing well at the three-month follow-up and was using 4 mg risperidone, 2 mg biperiden, and 900 mg lithium; however, he had complaints of spasms and left leg pain, considered to be related to the antipsychotics. Risperidone was reduced to 2 mg, while the other medications were continued at the same doses.

Two months later, the patient had manic symptoms that lasted for two weeks, and was in the anteflexion position. The patient was hospitalized for a manic episode and acute dystonia. Risperidone was stopped and a treatment regimen was initiated including 6 mg biperiden, 900 mg quetiapine, 900 mg lithium, and 2 mg clonazepam. The manic symptoms regressed; however, dystonia continued. Biperiden was discontinued on the 16th day of hospitalization; 15 mg baclofen was initiated and increased to 30 mg; however, the dystonia did not improve. A daily dose of 100 mg tetrabenazine was initiated. Then the patient’s mood was elevated again. ECT and 25 mg clozapine were added to the ongoing treatment. Quetiapine was terminated. Tetrabenazine was increased to 200, then 300 mg, with no improvement in dystonia. The patient did not respond to walking, balance, posture, and isometric strengthening exercises prescribed by the Physical Therapy Department. His mania improved. The patient received 30 sessions of ECT in total. He was discharged and prescribed 200 mg clozapine, 300 mg tetrabenazine, and 1500 mg lithium. The neurology department administered botox in the outpatient clinic, but he did not recover from dystonia. Later, he was referred to the algology department.

The algology assessment revealed posture disruption while walking, sitting, and going upstairs, and posture improvement with resting. The leg on the affected side faced outwards, straight leg raise test was 30 degrees positive, hand to floor distance was 30 cm, pace test and Freiberg test were positive, and taut band (feeling of pain by deep palpation of the muscles) was present around the PM. A diagnostic piriformis block was performed at the point of a “twitch response” on the taut band, and posture distortion was corrected. Hence the patient was diagnosed with PS. Two sessions of piriformis block, one week apart, were performed with a combination of 10...
mL 0.25% bupivacaine and 80 mg triamcinolone. Posture and walking were normal at the three-month check-up.

The patient has last been on 1500 mg lithium and 100 mg clozapine, and he has been on the same treatment for the last 1.5 years. He did not experience any mood episodes in this period.

**DISCUSSION**

Since physicians generally concentrate only on their own area of expertise, they may fail to consider other etiologies, especially rare situations. The neurology and physical therapy departments interested in movement disorders also failed to consider PS in our case. They attempted to treat the patient for dystonia. Consequently, the patient was considered to have chronic dystonia, and he was administered almost all antidystonic treatments: biperiden, benzodiazepine, baclofen, tetrabenazine, ECT, and botox; however, he remained irresponsible and was hospitalized for 172 days.

The algologist treating the patient, when asked what can be used to improve dystonia, explained that the patient suffered from PS requiring an intervention, as an anteflexion position was observed due to his back pain. Thus the patient was not diagnosed with dystonia, but with PS, and his posture improved as he recovered from PS. Actually, PS had developed in the patient, consequently he had taken anteflexion position while using antipsychotics. This condition was inadvertently considered to be dystonia associated with antipsychotics. The algologist explained that the patient had fallen, resulting in a stretched PM and compression of the sciatic nerve, thereby causing PS.

The literature says that PS frequently goes unrecognized or is misdiagnosed in clinical settings. PS symptoms are similar to other common somatic dysfunctions, such as intervertebral discitis, lumbar radiculopathy, primary sacral dysfunction, sacroiliitis, sciatica, and trochanteric bursitis. Further, through compensatory or facilitative mechanisms, piriformis syndrome may contribute to cervical, thoracic, and lumbosacral pain, as well as to gastrointestinal disorders and headache. Our patient was standing in the anteflexion position to get rid of pain, while we thought that this postures was caused by antipsychotic dystonia.

The PS diagnosis was reached by the algologist’s special examination as mentioned in the case presentation, and anyway anti-dystonic treatment did not work, whereas the patient improved with specific treatment of PS.

In the treatment of PS, hot applications, ultrasound treatment, non-steroidal anti-inflammatory drugs (NSAIs), and muscle relaxants are used; if these are ineffective, local anesthetics, steroids or botulinum toxin type A (BTX-A) are injected into the PM or the perisciatic region. Stretching of the PM and strengthening of the abductor and adductor muscles should also be included in patient treatment plans. If all of the pharmacologic and other therapies fail, the final treatment option is surgical decompression.

Psychiatrists may not know PS well nor be aware of these situations requiring algology consultation. This case shows that advanced diagnostic examinations should be carefully performed in psychiatric clinical practice, especially in therapy-resistant cases, in conjunction with consultations from related clinical departments. Additionally, psychiatrists should keep up with advances in general medicine and follow newly developed disciplines such as algology.
References:


