Levodopa-induced belly dancer’s dyskinesia: Case report

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ARTICLE INFO

Article history:
Received 12 April 2020
Received in revised form 5 July 2020
Accepted 7 August 2020
Available online 14 August 2020

Keywords:
Belly dancer dyskinesia
Levodopa, drug-induced movement disorder
Parkinsonism

ABSTRACT

The first report of an abdominal wall dyskinesia syndrome was published in 1990 by Illiceto et al. (1990), who were the first to use the “belly dancer dyskinesia” (BDD) nomenclature. BDD is a rare syndrome involving repetitive, involuntary, and continuous movements of the abdominal wall muscles (Illiceto et al., 1990). We describe the case of a 79-year-old, right-handed woman who showed symptoms of BDD syndrome after using levodopa due to Parkinson’s disease (PD). In this report, BDD was treated by stopping levodopa. After 15 days without levodopa, a positive outcome was achieved. The patient no longer exhibited BDD at the six-month and one-year follow-ups.

1. Introduction

Historically, the first report of an abdominal wall dyskinesia syndrome was published in 1990 by Illiceto et al. [1], who were the first to use the “belly dancer dyskinesia” (BDD) nomenclature. BDD is a rare syndrome involving repetitive, involuntary, and continuous movements of the abdominal wall muscles, which have been reported to cause pain [1]. Several etiologies have already been linked to this phenomenon, including drug-induced onset (prolopa, biperiden, domperidone, quetiapine), post-abdominal surgery, diaphragmatic flutter, functional movement disorders, and tumors of the spinal cord. In these cases, the only clinical option is management of symptoms, and usually, recovery is incomplete.

2. Case report

We describe the case of a 79-year-old, right-handed woman presenting with constipation associated with bradykinesia. The patient previously exhibited hypomimia for approximately 10 years. According to family members, the patient experienced a significant worsening of gait and some episodes of muscle stiffness over the years, in addition to a tendency to fall over in recent months. In view of the reported symptoms, the patient was initially evaluated by a general practitioner, who referred her for consultation with a neurologist.

Upon initial neurological examination, the patient presented with hypomimia, a small-stepped gait, and trunk anteroflexion. Motricity was used to measure muscle strength, with a result of grade 5/5 in all limbs. Plastic rigidity was present in the upper limbs, along with cogwheel phenomenon in both upper limbs and a positive Myerson’s sign. There was no evidence of tremors. The patient was also taking 50 mg losartan two times per day and 25 mg hydrochlorothiazide once per day to control systemic arterial hypertension. The patient’s brain MRI was normal. Since the findings were strongly suggestive of rigid akinetic Parkinson’s disease (PD). We chose to start the patient on levodopa plus benzerazide 200/50 three times per day.

Following the PD medication regimen, the patient demonstrated improvements in rigidity and postural instability, and was walking more safely, with no report of the “wearing off” phenomenon common in levodopa. After four months of PD treatment, the patient returned to the doctor with complaints of involuntary movements restricted to the abdomen, which began approximately 20 min after taking levodopa and lasted for about 4 h. Family members reported that these symptoms ceased during sleep.

Neurological examination indicated involuntary, continuous and undulating movements restricted to the abdominal wall (Videos 1 and 2). Such findings strongly suggested levodopa-induced BDD. As a result, levodopa administration was suspended, and after 15 days the patient returned to the doctor no longer exhibiting the movements that she had previously presented with (Videos 3 and 4). Following this, the patient was started on 1 mg Rasagiline once per day to treat the symptoms of PD. Following this change in medication, the patient no longer exhibited BDD at the six-month and one-year follow-ups.

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Available online 14 August 2020
Accepted 7 August 2020
Received 12 April 2020
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Clinical Parkinsonism & Related Disorders 3 (2020) 100068
Contents lists available at ScienceDirect
Clinical Parkinsonism & Related Disorders
journal homepage: www.elsevier.com/locate/prdoa

http://dx.doi.org/10.1016/j.prdoa.2020.100068
2590-1125/http://dx.doi.org/10.1016/j.prdoa.2020.100068
3. Discussion

The exact etiology of dyskinesia induced by levodopa is still unknown. Following the reduction of dopamine in PD, resulting dopamine receptor supersensitivity is thought to contribute to the development of dyskinesias. According to Olanow et al. [2], the short half-life of levodopa and pulsatile release of dopamine, which occur once the buffering capacity of the dopamine transporter is lost, are considered to be the major risk factors in the development of levodopa-induced motor complications.

BDD syndrome is a rare form of dyskinesia that has been linked to the use of levodopa by another case report [3]. A previous study suggested that BDD arises from either a dysfunction in the inhibitory control of spinal interneurons or a change in the organization of neuronal circuits [1]. Other studies have postulated that the BDD movements are related to dopaminergic receptors of non-physiological postsynaptic stimulation, thus generating an imbalance in motor control [4].

In order to properly assess potential BDD syndrome cases, a detailed investigation with a clinical history and a thorough neurological physical examination is essential. Although several approaches have been detailed in literature to address this syndrome, BDD remains a challenge even for the most experienced doctors. It is important to check for history of recent surgery or trauma in the abdominal region in potential BDD patients, as well as the presence of psychogenic determinants, as these can influence symptoms.

The presently described case is somewhat similar to the case report by Carecchio et al. [3], who presented a patient in a similar age group (>70 years) and of the same sex (female). In addition, this case report also described a similar time of symptom onset following administration of levodopa (around 30 min), and similar duration of symptoms (3–5 h). Like our case report, the report described by Carecchio et al. [3] described an interruption of BDD symptoms after elimination of levodopa. Finally, another similarity between the cases was the administration of rasagiline at a dose of 1 mg per day as a replacement for levodopa in the treatment of PD, resulting in no new BDD symptoms in both cases.

Although rasagiline, like levodopa, is a drug that acts on dopaminergic neurotransmission, dyskinesia is not a common side effect. Generally, patients with adverse symptoms complain of nausea, headaches, dizziness and drowsiness. The severity of PD greatly affects the ability to achieve an adequate response to treatment, where this response decreases as the disease progresses. In addition to the drug’s already known effects on PD, the neuroprotective properties of this drug are also being investigated [5].

In addition to levodopa, other medications have been reported to cause BDD syndrome, such as biperiden, clebopride, and domperidone [6]. The treatment approach for BDD consists of firstly removing the possible drug involved and then observing the patient. The expected therapeutic response is the improvement of dyskinetic symptoms. However, this response is variable, where the patient may either remain symptomatic or present with great improvement in symptoms, such as in the present case.

To date, no clinical study has studied the treatment options for BDD. Therefore, recommended treatments are based on the experience of doctors who have dealt with BDD cases. A variety of medicated and non-medicated treatment options have already been used in cases where suspension of the drug that potentially caused the syndrome did not eliminate symptoms. Some drugs reported to have caused BDD include diphenylhydantoin, haloperidol, clonazepam, and aripiprazole [7].

4. Conclusion

In conclusion, treating BDD syndrome is challenging, and little information is known. In the case described in this report, BDD was treated by stopping levodopa. After 15 days without levodopa, a positive outcome was achieved. The patient no longer exhibited BDD at the six-month and one-year follow-ups.

Supplementary data to this article can be found online at https://doi.org/10.1016/j.prdoa.2020.100068.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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