



OROFACIAL MOVEMENT DISORDER CAUSED BY PRAMIPEXOLE ABUSE. A CASE REPORT.

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ABSTRACT

Orofacial movement disorders are a group of hyperkinetic extrapyramidal movements presenting dysfunctional activities on the masticatory, facial mimic, or tongue musculatures. The most common cause of acquired orofacial movement disorders is drug-induced dyskinesias. Our report describes a rare case of pramipexole-induced orofacial movement disorders in a patient with restless legs syndrome.

KEYWORDS: *Dyskinesia, pramipexole, movement disorders, restless leg syndrome*

INTRODUCTION

Orofacial movement disorders (OMD) are a group of hyperkinetic extrapyramidal movements presenting as isolated or combined dysfunctional activation of the masticatory, facial mimic, or tongue muscles (1). The aetiology of OMD may be genetic, idiopathic, or acquired (2). However, the most common cause of acquired OMD is acute or tardive drug-induced dyskinesias, mainly caused by antidepressants, antiemetics, neuroleptics, or levodopa (3).

The main treatment of dyskinetic/dystonic symptoms is the slow tapering of the offending drug. Pharmacological treatment is mostly empirically based on using tetrabenazine, clonazepam, amantadine, or piracetam. In addition, anticholinergic drugs are useful for associated dystonic symptoms (4). Besides conventional therapies, levodopa-induced dyskinesias (LID) may also benefit from non-ergot dopamine-agonist (DA), clozapine, or antiepileptics (5).

Case presentation

Here we report a case of acute OMD caused by chronic DA abuse in a non-Parkinson's disease (PD) patient under treatment with pramipexole for restless legs syndrome (RLS). A 72-year-old woman was admitted to the emergency room for the appearance, during the last week, of intermittent, involuntary muscle contractions, causing repetitive eye movements, grimacing, pursing of the mouth and lips, and writhing of the tongue with stereotyped vocalizations and sustained neck dystonia.

Neurological examination was otherwise normal. The patient's medical history included RLS, low back pain, diabetes mellitus type 2, and surgical removal of a foot acral melanoma with inguinal lymphadenectomy. Her medical therapy consisted of 1,000 mg metformin daily, 150-200 mg tapentadol daily, and pramipexole prescribed at a 0.18 mg daily dose to control RLS, but arbitrarily taken at 0.9 mg daily dose for 2 months due to a self-assessed lack of efficacy. No past or current exposure to other relevant drugs was reported.

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The patient underwent a computed tomography scan of the brain and a dopamine transporter single photon emission tomography (DAT SPECT) scan, with normal results. In addition, the patient was prescribed a reduction of pramipexole to 0.18 mg daily, which resulted in the complete resolution of OMD in 4 days.

DISCUSSION

Our report describes a rare case of DA-induced OMD in a patient with RLS due to drug abuse. The mechanism underlying LID is attributed to a specific enhancement of the direct striatopallidal pathway and the inhibition of the indirect striatopallidal pathway, which may be found in PD patients (6). The resulting decreased output of the internal globus pallidus may lead to increased activity in the motor nuclei of the thalamus (7).

A chronically increased dopaminergic tone in RLS may induce a postsynaptic receptor down-regulation, mainly of the indirect pathway (8). Thus, in RLS patients, the relative impairment of dopamine transmission becomes clinically evident in the late hours since circadian dopamine activity is physiologically lower in the evening. Accordingly, a low dose of DA at bedtime is the first-line therapy for RLS (9).

RLS association with PD did not receive sufficient evidence in the literature (10), and our patient did not show any PD-related motor or non-motor symptoms, as often happens in RLS cases (10). Nevertheless, after excessive intake of DA, she developed symptoms that resembled LID, a typical PD complication.

CONCLUSIONS

This report is the first on an OMD following excessive intake of DA in non-PD patients. Follow-up of this patient will address the possible future development of Parkinson's disease.

Statement of Ethics

The authors confirm that the approval of an institutional review board was not required for this work. Written informed consent was obtained from the patient's caregiver to publish this case report. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

Conflict of interest

The authors declare that they have no conflict of interest.

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Author Contributions

Conceptualization: DC and LB; methodology: LB, CC, and GN; writing-original draft preparation: DC and MADR; writing-review and editing: DC, MADR, and LB; visualization: DD and LB; supervision: MO, FC, and LB.

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