

Case Reports

An Unusual Cause of Camptocormia

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Abstract

Background: Camptocormia is defined as forward flexion of the spine that manifests during walking and standing and disappears in recumbent position. The various etiologies include idiopathic Parkinson's disease, multiple system atrophy, myopathies, degenerative joint disease, and drugs.

Case Report: A 67-year-old diabetic female presented with bradykinesia and camptocormia that started 1 year prior to presentation. Evaluation revealed levosulpiride, a dopamine receptor blocker commonly used for dyspepsia, to be the culprit.

Discussion: It is well known that dopamine receptor blockers cause parkinsonism and tardive syndromes. We report a rare and unusual presentation of camptocormia attributed to this commonly used gastrointestinal drug in the Asian population.

Keywords: Levosulpiride, camptocormia, parkinsonism

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Introduction

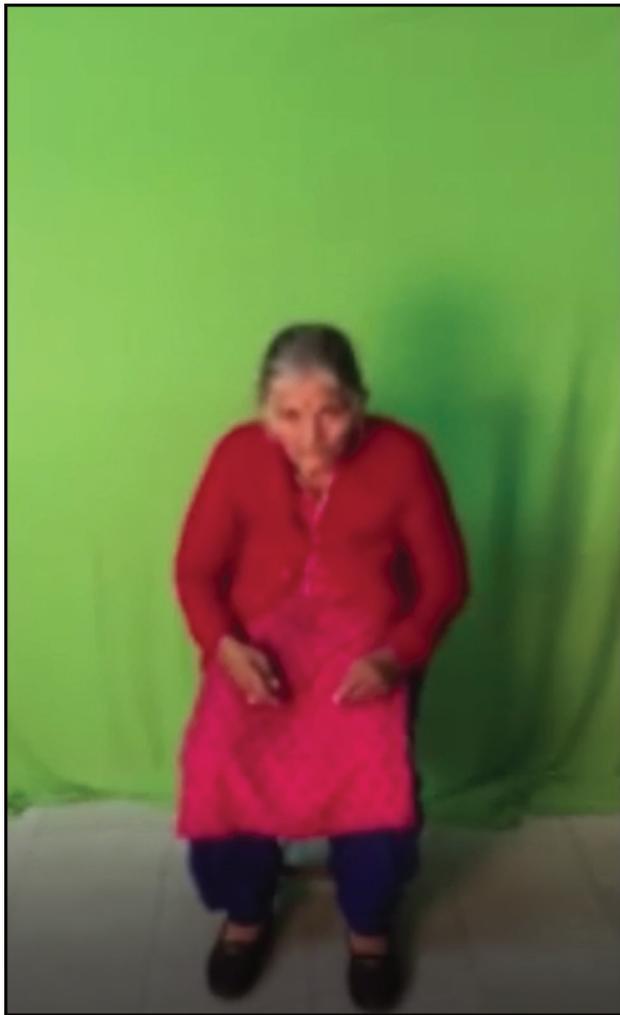
Camptocormia is defined as forward flexion of the spine ($>45^\circ$) that manifests during walking and standing and disappears in recumbent position.¹ It has both neurological and musculoskeletal causes. Various neurological etiologies include idiopathic Parkinson's disease (PD), multiple system atrophy, genetic forms of PD, dystonias, myopathies, and drugs.² We report a patient who presented with camptocormia and evaluation disclosed an unusual cause.

Case report

A 67-year-old female presented with change in speech, slowness while walking, and forward bending of the spine that had started 1 year prior to presentation. Examination revealed slow hypophonic speech, bilateral bradykinesia, and rigidity. Gait was slow and short stepped, with gait initiation difficulty and the presence of camptocormia (Video 1). She was a known diabetic for 3 years and had a history of coronary artery disease with an ejection fraction of 45–50%. Her renal functions were deranged (creatinine 1.9 mg/dL) suggestive of diabetic nephropathy.

There were no red flags in the form of autonomic dysfunction, pyramidal or cerebellar signs, or cognitive problems to suggest atypical parkinsonism. She was evaluated to determine the cause of her symmetrical parkinsonism. Magnetic resonance imaging of the brain was normal. 18 F FLUORODOPA (FDOPA) positron emission tomography showed bilateral symmetrical uptake of dopamine in the striatum, thus ruling out a degenerative cause for parkinsonism (Figure 1A,B). Other relevant investigations such as the thyroid profile and liver function tests were normal. On review of her medical records, it was found that patient had been taking levosulpiride 75 mg/day for the last 18 months for her dyspeptic symptoms. There was a significant improvement in bradykinesia within 10 days of stopping levosulpiride and starting levodopa/carbidopa (125 mg three times daily). However, the improvement in camptocormia was modest and began 2 months after the introduction of levodopa.

At 6 months, there was significant improvement in her symptoms, and the patient is currently on regular doses of levodopa/carbidopa (Video 1). She is able to intermittently keep her posture erect with the reappearance of camptocormia.



Video 1. Segment 1. The presence of camptocormia and short step gait.
Segment 2. The near complete resolution of camptocormia with improvement in slowness.

Discussion

Camptocormia is defined as abnormal and severe flexion ($>45^\circ$) of the thoracolumbar spine that manifests during walking and standing and disappears in recumbent position.¹ It can occur in sporadic PD, post-encephalitic and parkin-related PD as well as in multiple system atrophy.² The pathogenesis of camptocormia in PD is not fully understood. It may represent a PD-related abnormal posture or be attributed either to truncal dystonia or to a severe form of rigidity.³ It usually responds poorly to levodopa but may be alleviated to some extent with botulinum toxin therapy and deep brain stimulation.^{4,5} It can also occur as a tardive syndrome after exposure to dopamine receptor blockers such as levosulpiride.⁶ Focal myopathy involving the paravertebral muscles is another postulate reported in the literature.⁷

Levosulpiride, a commonly used prokinetic in Asian countries, is a substituted benzamide that inhibits D2 receptors in the central nervous system and gastrointestinal tract. It is well known to cause parkinsonism, tremor, and tardive dyskinesia.^{6,8}

Neuronal damage in the striatum via oxidative stress and glutamatergic transmission on prolonged exposure to neuroleptics underlies the pathophysiology of tardive syndromes.⁹ Another school of thought is that hypersensitivity or upregulation of D2 receptors lead to maladaptive plasticity in the synapses and the cortico-striatal transmission, which leads to abnormal involuntary movements.¹⁰ There is also evidence of pharmacogenomics in the pathophysiology of tardive syndromes. Various polymorphisms in the genes encoding D2 and D3 receptors and 5-HT2A receptors, as well as in the *CYP2D6* gene, are known to influence the development of tardive syndromes.^{11,12}

It is well known that drug-induced parkinsonism almost always resolves when the causative agent is stopped.¹³ Tardive dystonias, on the other hand, are usually persistent, and spontaneous remissions are uncommon. Kiriakakis et al.¹⁴ in their study of 107 patients of tardive dystonia found that only 14% of patients showed remission after a mean duration of 8.5 years. The abnormal flexion of the spine in our patient took more than 2 months to resolve, and is therefore more likely to be

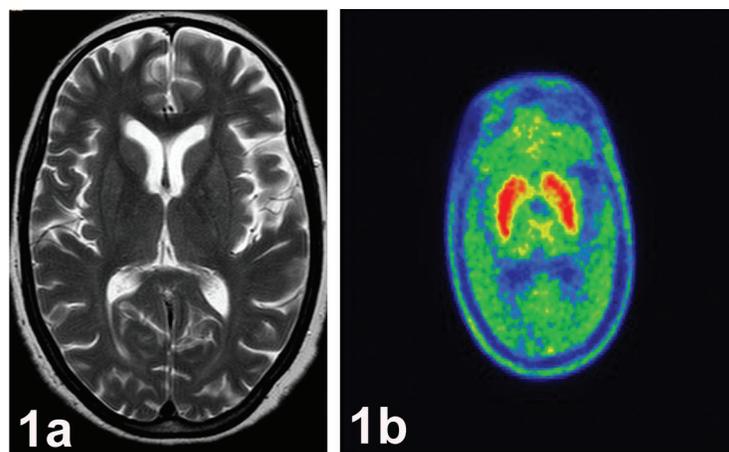


Figure 1. Structural and Functional Imaging of the Brain. (A) T2 weighted magnetic resonance imaging brain shows normal appearing bilateral basal ganglia. (B) 18 F FLUORODOPA (FDOPA) positron emission tomography revealed adequate tracer uptake in bilateral striatum.

due to tardive dystonia. Bradykinesia that resolves in less than 2 weeks suggests it to be the result of drug-induced parkinsonism.

Presentation with camptocormia was slightly unusual in our patient. In our patient, the strong temporal relationship of occurrence of camptocormia with levosulpiride intake in the presence of normal structural and functional imaging indicates a causative association. To the best of our knowledge, levosulpiride-induced camptocormia has not been previously reported in the literature.

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