A Wilted Flower: Lotus Position Dystonia

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Adult-onset axial dystonia is a rare disorder and may be idiopathic or secondary to another systemic condition. It may, moreover, be under-recognized or misdiagnosed because of the unusual phenomenology. The pathophysiological mechanisms are poorly understood. Task specificity, common to other forms of dystonia, is moreover difficult to demonstrate in axial dystonia. We present herein a case of adult-onset axial dystonia related to a partial “lotus” position, with task specificity and sensory trick.

Case Report

A 58-year-old Punjabi man presented with a primary complaint of altered posture while sitting with legs crossed, in a partial lotus position. Within seconds of sitting in that specific position, he would develop progressive stiffness and a lean to the left. This was not present if seated in other positions, including with legs extended, or if kneeling. Furthermore, it was not present even in the lotus position if he leaned against a wall. His symptoms evolved subacutely over a few months, then progressed more slowly over 2 years. Movements initially only occurred when sitting in the cross-legged position, though after some months he developed a similar, though lesser, lean while sitting in a chair (Video 1). He had taken up yoga in the past few years and would sit in this position for hours at a time. There were no other movement abnormalities or neurological symptoms. There was no significant past medical history, though, as seen in the video, the patient had an old right elbow injury with a resultant fixed deformity. He had not been exposed to neuroleptics or antiemetics. There was no trauma. There was no family history of dystonia. Investigations, including MRI brain, MRI spine, and cerebrospinal fluid (CSF) analysis, were all normal. N-methyl-D-aspartate receptor antibodies, measured using an enzyme-linked immunosorbent assay assay, were positive in serum once, but were negative on repeat testing with cell-based assay and were also negative in CSF. Glutamic acid decarboxylase antibodies were negative. A diagnosis was made of isolated, task-specific focal dystonia of the neck and trunk triggered by

sitting cross-legged. A course of botulinum toxin to the paraspinal muscles provided no relief. Various medications have been trialled with little effect.

Discussion

Task specificity is a well-known feature of dystonia and can, at times, be bizarrely specific. It is thought to relate to overspecialization of a task and is often observed after repetitive, highly skilled, or unaccustomed tasks. However, it can also develop with a relatively simple repetitive task, such as walking downstairs.¹ Pathophysiology may be related to altered neuroplasticity and disordered sensory integration.² Minor secondary segmental spread or generalization after onset is not uncommon, as observed in this patient, and may be related to alteration in neural networks.²

Dystonia can present in any body part, though onset in adulthood is typically focal and most commonly involving the craniocervical region. Axial onset dystonia in adulthood is uncommon. The largest series of such patients was reported in 1997 and included 18 patients.³ The majority had flexion dystonia, with most getting worse with action (i.e., walking). The phenotype, however, was more severe than that noted here and was not isolated to a particular task. Other causes of adult-onset axial dystonia have been described, including neurodegeneration with brain iron accumulation,⁴ chorea acanthocytosis,⁵ ataxia telangiectasia,⁶ and pseudo-axial dystonia in stiff-person syndrome.⁷ There are typically more systemic features, however, and, again, movements are not typically task specific. Axial dystonia, often taking the form of forward (camptocormia) or lateral (“Pisa” syndrome) trunk flexion is also a well-known feature of parkinsonian syndromes, both typical and atypical, and may occur at onset or present later on in the disease course.⁸ However, the axial dystonia is typically not task specific.

Sitting cross-legged is common in many non-Western cultures. The related lotus position, in which each foot is placed on the opposite thigh, is commonly assumed in yoga and is often maintained for prolonged periods. This position may be

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considered a “specialized” task and therefore analogous to other known triggers of task-specific dystonia.

The features noted here are consistent with an adult-onset, idiopathic axial dystonia and, interestingly, task specific to sitting cross-legged in a partial lotus position. We present this case for its unique phenomenology. Moreover, adult-onset axial dystonia is uncommon, with task specificity rarely, if ever, reported (walking could be considered either a specific task or nonspecific action in terms of control of truncal activity). Given the ubiquity of this position worldwide, we wonder whether this wilted flower syndrome may be under-recognized.

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Author Roles

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Supporting Information

Supporting information may be found in the online version of this article.

Video 1. Patient demonstrates axial dystonia, which is present immediately on assuming the partial lotus position. The dystonia is not present if seated in other positions or with a sensory trick of leaning against a wall. There is mild axial dystonia when seated on a chair. There is mild right shoulder elevation on walking, but gait is otherwise normal.