

Adult-onset Re-emergent Stuttering as a Presentation of Parkinson's Disease

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Abstract

Introduction: The basal ganglia-thalamocortical motor circuits are postulated to play a key role in the aetiopathogenesis of stuttering. The main dysfunction is thought to be an impairment in the ability of the basal ganglia to produce timing cues for the initiation of the next motor segment of speech, explaining the association of acquired and re-emergent stuttering with diseases such as dystonia and Parkinson's disease. **Clinical Picture:** We describe a 61-year-old man presenting with re-emergent stuttering and mild hypomimia, only to develop unilateral rest tremors, hypo- and bradykinesia, rigidity and gait difficulties one year later. **Treatment and Outcome:** His parkinsonism responded well to treatment with bromocriptine, but he continued to stutter. **Discussion:** This case illustrates the association between acquired or re-emergent stuttering and basal ganglia disorders, and highlights the need to assess such patients for an underlying aetiology.

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Key words: Parkinson's disease, Re-emergent, Stuttering

Case Report

A 61-year-old right-handed man with no past medical history presented to our movement disorders clinic with a history of stuttering which had begun 5 years earlier, and appeared to worsen in the preceding year. He had stuttered briefly in childhood, from the age of 5 to 11, but had been able to speak fluently thereafter, until the age of 56, when he suddenly found himself once again having difficulty with fluent speech. He noticed that the stuttering was worse when he was stressed or anxious, and when he spoke on the telephone. He stuttered less when speaking to close friends or family, but fared badly when speaking to a stranger. He had no family history of tremors or Parkinson's disease (PD), had no exposure to neuroleptic medications, and denied brady- or hypokinesia, postural difficulties, tremors, autonomic symptomatology such as orthostasis, erectile impotence or constipation, change in handwriting, sialorrhoea or limb stiffness. He denied cognitive or bulbar dysfunction, and had no postural difficulties. Clinical examination was unremarkable, except for a barely discernible hypomimia, which could have passed for normal, and a tendency to stutter, in that he demonstrated repetitions

and prolongations on initial syllables. This occurred mainly in self-formulated speech. He had less difficulty speaking when he read from a prepared text or when he quoted a well-remembered speech, such as the pledge of allegiance. He had no hypophonia, brady- or hypokinesia, tremors or cogwheel rigidity, and was scored at 1 (for hypomimia) on the motor scale of the Unified Parkinson's Disease Rating Scale (UPDRS). He walked with normal stride length, had good arm swing and did not retropulse on the pull test. Cognition was assessed with the abbreviated mini-mental examination, and was within normal limits. Neuroimaging with computed tomography scan of the brain and the electroencephalographic examination were within normal limits. Thyroid dysfunction and Wilson's disease were excluded, and he had no acanthocytosis. He was referred to a speech therapist and underwent intensive smooth speech therapy, but defaulted in view of his perceived poor response. He felt that his stuttering was less marked when he spoke slowly, and when he chorus-spoke. He was reviewed on a six-monthly basis in view of the mild hypomimia.

One year after his initial consultation, he was noted to have mild cogwheel rigidity in the right upper and lower

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limbs. He had hypomimia, was tachypneic and moderately hypophonic and was noted to have right sided hypo- and bradykinesia, demonstrated when he performed repetitive movements such as heel and finger taps, hand grip and pronation-supination movements of the forearm. He had occasional rest tremors in the right hand and walked with diminution of right armswing. The stutter had not worsened. Extraocular movements were full, and the saccadic and pursuit eye movements were well within normal limits. There was no demonstrable orthostasis, and he had no cerebellar dysfunction. He scored 9 on the motor scale of the UPDRS, and was diagnosed to have PD. He was commenced on bromocriptine 2.5 mg thrice daily and selegiline 5 mg twice daily, and reviewed 3 months later, during which time there was improvement in motor score to 1 (for hypomimia). There was complete resolution of his rest tremors, brady- and hypokinesia, and there was no trace of cogwheel rigidity, even with the performance of the Jendrassék manoeuvre, during the prolonged follow-up visit. His hypophonia improved, although he still tended to stutter, especially when he was agitated or when he spoke quickly.

Discussion

PD is a neurodegenerative disease diagnosed by the presence of 2 out of 3 cardinal features of tremors, rigidity and bradykinesia.¹ Diagnosis is known to be difficult in the early stages of PD.¹ Indeed, diagnosis of PD based on the above criteria alone led to incorrect diagnoses in 25% of cases that had postmortem studies performed at the London Brain Bank.¹ Asymmetry, with one side being more affected than the other, resting tremor and good response to levodopa are thought to be more reliable criteria for the diagnosis of idiopathic PD, whereas early-onset postural instability, autonomic dysfunction, dysphagia, poor response to levodopa, rigidity that is more axial than appendicular and prominent speech dysfunction are thought to indicate the possibility of atypical parkinsonism.¹

Speech is a dynamic motor function process, requiring a highly synchronised and adaptive neural network in order to function seamlessly. Different regions of the brain have been shown to be involved in the production of speech.² Wingate³ defined stuttering as “a disruption in the fluency of verbal expression, which is characterised by involuntary, audible or silent, repetitions or prolongations in the utterance of short speech elements, namely, sounds, syllables, and words of one syllable. These disruptions usually occur frequently or are marked in character and are not readily controllable. Accessory bodily activities and altered emotional state may be present”.

Stuttering is most commonly observed in young children, usually transiently in boys aged 2 to 5, when it is called developmental stuttering.⁴ When stuttering develops in the

adult, it is called acquired stuttering. It is rare, and is usually associated with multifocal hemispheric damage.⁵ Developmental stuttering is often associated with anxiety, embarrassment and even fear, because of the emotional and psychological factors which result from stuttering during the formative years.²

The aetiopathogenesis of stuttering is not well understood. For centuries, it was a widely held belief that it arose from abnormalities of the larynx or tongue.⁶ It is now thought that abnormalities in the central control of speech are not due to disturbances in any one brain region, but are due to a systems dysfunction that interferes with rapid and dynamic speech processing.²

Acquired stuttering, which involves repetitions, vowel prolongations and occasional blocks, can be distinguished from palilalia, in which there is uncontrolled rapid festinating syllabic repetitions.² Acquired stuttering has been described in patients bearing lesions in locations which exert influence on multiple brain regions, such as the basal ganglia, particularly the putamen,² or which support and facilitate rapid communication between brain regions, such as the corpus callosum.² It is interesting that the lesions associated with acquired stuttering rarely involve the primary speech and language regions of the left hemisphere, which tend, instead, to cause aphasia. These patients do not stutter even upon recovery from the aphasia, which is an argument against the postulate that stuttering is masked by the aphasia.⁷

In 1983, Koller⁵ reported stuttering in extrapyramidal disease. Stuttering dysfluency has been reported in cases of PD, progressive supranuclear gaze palsy⁸ and parkinsonian syndrome. More recently, Moretti et al⁹ reported a case of speech initiation hesitation in a patient with no past history of stuttering, 1 month after subthalamic nucleus deep brain stimulation surgery for PD. It has also been reported in other disorders thought to involve the basal ganglia, such as dystonia¹⁰ and Tourette's syndrome.^{2,11}

Management of stuttering has ranged from speech therapy (comprising chorus speech, smooth speech and delayed auditory feedback), injections of botulinum toxin,¹² anticonvulsants,¹³ transcranial electromagnetic field application,¹⁴ to dopamine antagonists such as haloperidol, olanzapine and risperidone,¹⁵ in line with the “excess dopamine theory of stuttering” postulated by Wu et al.¹⁶ However, the role of dopamine agonists and antagonists in the management of stuttering is not definite. Just as there have been reports of improvement with dopamine antagonists, dopamine agonists such as apomorphine¹⁷ and pramipexole¹⁸ have been likewise found to improve stuttering, while dopamine antagonists like clozapine have been found to induce stuttering.¹⁷

The association between PD and stuttering is intriguing. PD is well known to be associated with repetitive speech phenomena,¹⁹ including stuttering. It has been proposed that the basal ganglia-thalamocortical motor circuits through the putamen are responsible for the symptoms of stuttering, explaining the association of stuttering with dystonia and PD.²⁰ The main dysfunction in stuttering is thought to be the impaired ability of the basal ganglia to produce timing cues for the initiation of the next motor segment of speech.²⁰ Recurrence of childhood stuttering in 12 patients, which had resolved by the age of 16, has been described by Shahed and Jankovic¹⁷ as re-emergent stuttering. They hypothesised that the same mechanisms may be involved in developmental and recurrent stuttering. We postulate that it may be possible in their patients (and in ours) that the developmental stuttering seen in childhood may have predisposed the patients to develop acquired stuttering later on, i.e., a *locus minoris resistentiae*. This case illustrates a little-known feature of the myriad of presentations possible with PD, and highlights the need to assess patients with acquired or re-emergent stuttering for basal ganglial disorders.

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