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A case of multiple system atrophy-parkinsonian type with stuttering- and palilalia-like dysfluencies and putaminal atrophy

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ABSTRACT

Both developmental and acquired stuttering are related to the function of the basal ganglia-thalamocortical loop, which includes the putamen. Here, we present a case of stuttering- and palilalia-like dysfluencies that manifested as an early symptom of multiple system atrophy-parkinsonian type (MSA-P) and bilateral atrophy of the putamen. The patient was a 72-year-old man with no history of developmental stuttering who presented with a stutter for consultation with our otorhinolaryngology department. The patient was diagnosed with MSA-P based on parkinsonism, autonomic dysfunction, and bilateral putaminal atrophy revealed by T2-weighted magnetic resonance imaging. Treatment with levodopa improved both the motor functional deficits related to MSA-P and stuttering-like dysfluencies while reading; however, the palilalia-like dysfluencies were much less responsive to levodopa therapy. The patient died of aspiration pneumonia two years after his first consultation at our hospital. In conclusion, adult-onset stuttering- and palilalia-like dysfluencies warrant careful examination of the basal ganglia-thalamocortical loop, and especially the putamen, using neuroimaging techniques. Acquired stuttering may be related to deficits in dopaminergic function.

1. Introduction

Neurogenic stuttering is an acquired speech disorder that typically affects adults with neurological disease; such stuttering is most often associated with cerebrovascular accident (CVA) (Ardila & Lopez 1986; Grant, Biousse, Cook, & Newman, 1999; Helm-Estabrooks, Yeo, Geschwind, Freedman, & Weinstein, 1986; Helm, Butler, & Benson, 1978; Jokel, De Nil, & Sharpe, 2007; Rosenfield, 1972; Sakai 2010, 2011; ; Theys, van Wieringen, Sunaert, Thijs, & De Nil, 2011; Theys, De Nil, Thijs, van Wieringen, & Sunaert, 2013), traumatic brain injury (TBI) (Helm-Estabrooks & Hotz, 1998; Jokel et al., 2007; Ludlow, Rosenberg, Salazar, Grafman, &

Abbreviations: BGTC loop, basal ganglia-thalamocortical loop; CVA, cerebrovascular accident; MRI, magnetic resonance imaging; MSA-P, multiple system atrophy Parkinsonian type; PD, Parkinson's disease; SLD, stuttering-like dysfluency; T2WI, T2-weighted imaging; TBI, traumatic brain injury; UPDRS, Unified Parkinson's Disease Rating Scale

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Smutok, 1987; Strasberg, Johnson, & Parry, 2016), and neurodegenerative disease (Koller, 1983; Leder, 1996). TBI and CVA are easily diagnosed as causes of neurogenic stuttering; TBI is normally reported after a traumatic event, and CVAs such as brain infarction can be diagnosed in early stages using diffusion-weighted imaging (Minematsu et al., 1992), while brain hemorrhages can be diagnosed with computed tomography (Tohgi et al., 1981). In contrast, stuttering accompanying neurodegenerative disease is frequently overlooked or misdiagnosed, especially in early stages of the disease.

To some degree, the phenomenology of stuttering resembles that of a gait disorder in Parkinson's disease (PD). PD is a disorder that involves the basal ganglia, and gait in patients with PD is typically characterized by small steps (i.e., reduced stride length) and lower cadence associated with reduced gait speed, together with festination and freezing (i.e., difficulty in gait initiation or stopping when turning or approaching an obstacle) (Giladi et al., 1992). Visual (e.g., floor markers) or auditory (e.g., metronome) cueing can improve gait performance in patients with PD (Lim et al., 2005; Suteerawattananon, Morris, Etnyre, Jankovic, & Protas, 2004). Similarly, stuttering is also improved by use of a metronome or other external auditory cueing (Brady, 1969; Toyomura, Fujii, & Kuriki, 2011). Recently, the cortico-basal ganglia-cortical network was implicated as a neural substrate of both acquired stuttering (Theys et al., 2013) and developmental stuttering (Chang, Chow, Wieland, & McAuley, 2016; Chang & Zhu, 2013; Craig-McQuaide, Akram, Zrinzo, & Tripoliti, 2014; Ingham, Wang, Ingham, Bothe, & Grafton, 2013; Sitek et al., 2016; Toyomura, Fujii, & Kuriki, 2015; Yang, Jia, Siok, & Tan, 2016). In developmental stuttering, putaminal neuropathology appears to underlie the disorder (Alm, 2004; Beal, Gracco, Brettschneider, Kroll, & De Nil, 2013; Chang, Kenney, Loucks, & Ludlow, 2009; Ingham et al., 2013; Jiang, Lu, Peng, Zhu, & Howell, 2012; Lu et al., 2010; Toyomura et al., 2011, 2015). Putaminal atrophy has been reported in Parkinson-plus syndromes such as multiple system atrophy-parkinsonian type (MSA-P) (Feng et al., 2015; Schrag et al., 1998). MSA-P is characterized by parkinsonism (bradykinesia, rigidity, irregular jerky tremor, and postural instability), as well as autonomic failure in the form of bladder dysfunction (including early urinary incontinence) and/or orthostatic hypotension (Gilman et al., 2008). Here, we describe a case of adult-onset stuttering associated with MSA-P and bilateral putaminal atrophy diagnosed using magnetic resonance imaging (MRI).

2. Case presentation

The patient was a 72-year-old right-handed man with no history of stuttering. At the age of 70 years, the patient's wife reported that he was repeating his speech and that this was the time at which he first noticed his stuttering. He went to a hospital complaining of only the stutter and underwent diagnostic MRI, but no abnormalities were detected. However, at the age of 71 years, he noticed a tremor of the hands and abnormal gait (short steps and difficulty changing direction). Thereafter, he went to another hospital to receive a second opinion, but, once again, there were no abnormal findings on MRI. At the age of 72 years, the patient read a newspaper advertisement about consultation for stuttering and visited our department. His speech was characterized by repetitions and blocks. He also presented with hand and tongue tremors, and it was suggested that he undergo MRI for suspected basal ganglia abnormalities.

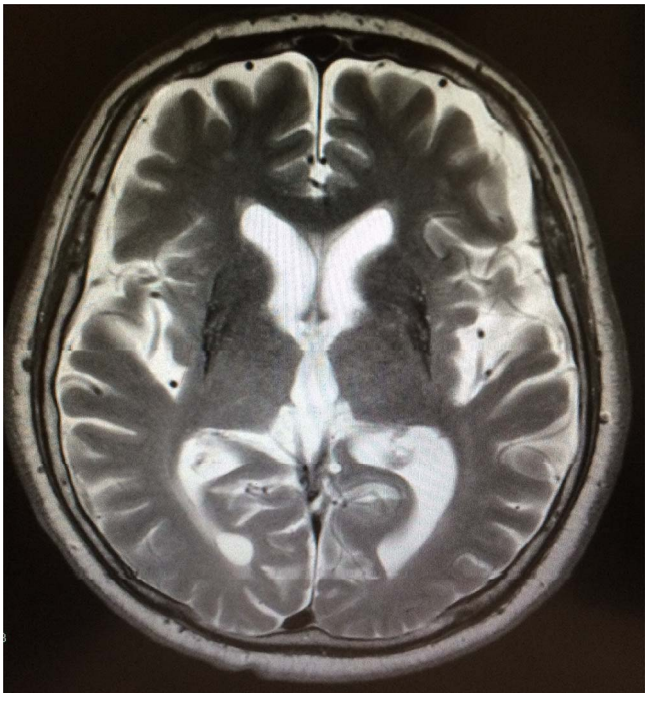

Fig. 1. Bilateral putaminal atrophy and rim hyperintensity on T2WI.