

Uneven focal shoe deterioration in Tourette syndrome

Andrea E Cavanna^{1,2}
 Francesco Monaco¹
 Marco Mula¹
 Mary M Robertson²
 Hugo D Critchley³

¹Department of Neurology, Amedeo Avogadro University, Novara, Italy;

²UCL Institute of Neurology, London, UK; ³Department of Psychiatry, Brighton and Sussex Medical School, Brighton, UK

A 31-year-old single man (AB) sought neuropsychiatric consultation for treatment-resistant motor and vocal tics. He described himself expressing a total of 24 different tics, mainly facial twitches (eye blinking, raising eyebrows, mouth opening, lips licking, stereotyped grimacing) and inappropriate utterances (grunting, throat clearing, sniffing), since the age of 7. There appeared to be no family history of tic disorder. He reported occasional utterance of swear words in contextually inappropriate situations (coprolalia), and the urge to copy other people's movements (echopraxia). Other tic-associated symptoms included self-injurious behaviours and forced touching of objects. A.B. met both DSM-IV-tr and ICD-10 criteria for Tourette syndrome, and also DSM-IV-tr criteria for attention deficit hyperactivity disorder (combined type) in childhood. His YGTSS score was 51%. Quite interestingly, he did not report any distressing ritual or obsessional thought. On the other hand, the clinical examination revealed, among other signs, a complex, non-purposeful motor tic characterized by repeated tapping of the tip of the right foot against the floor, with consequent focal deterioration of the tip of the right shoe (Shapiro et al 1987). The patient described this tic as "one of the most longstanding and annoying" he had ever experienced. Following pharmacological treatment with neuroleptics (flupenthixol 20 mg/day for 8 years), AB reported that most tics had decreased in frequency, but right foot tapping was still obvious.

Tourette syndrome is a neurodevelopmental disorder characterized by multiple motor and vocal tics with onset at childhood. Motor tics are most commonly localized to the face and neck/shoulders, followed by the upper limbs (Shapiro et al 1987). Tics involving the feet are less frequent, but not rare, and usually present with a more complex pattern, often resembling voluntary movements such as kicking or stamping (Jankovic and Fahn 1986). This patient's shoes provided evidence of asymmetrical foot tapping causing marked psychosocial distress and potential self injury through repeated microtrauma at the interphalangeal joints of the first and second toe (Robertson et al 1989).

Bilateral presentation of shoe semiology is suggestive of habitual behaviours, and restlessness associated with restless leg syndrome/akathisia or ADHD. On the other hand, asymmetrical shoe deterioration can be encountered in a few pathological conditions, including hemiparesis, foot dystonia, and rheumatoid arthritis. Moreover, unevenly worn shoes have been described as important clues to be taken into account beyond more obvious symptoms in peripheral neuropathies (Willison and Winer 2003). Recently, Liu et al (2005) defined the "Machado-Joseph shoe" (MJS) as a shoe/slipper worn for less than 2 years that shows a deep hallux imprint at its base, in the absence of any other signs of shoe abrasion. The suggested explanation for the formation of MJS is the excessive motion of the hallux during walking to compensate ataxia. According to Liu et al (2005), the MJS plus hyperostosis/osteoma toes may be

Correspondence: Andrea Eugenio Cavanna, Department of Neurology, Amedeo Avogadro University, Corso Mazzini 18, 28100 Novara, Italy
 Tel +39 03 21373 3371
 Fax +39 03 21373 3298
 Email a.cavanna@ion.ucl.ac.uk

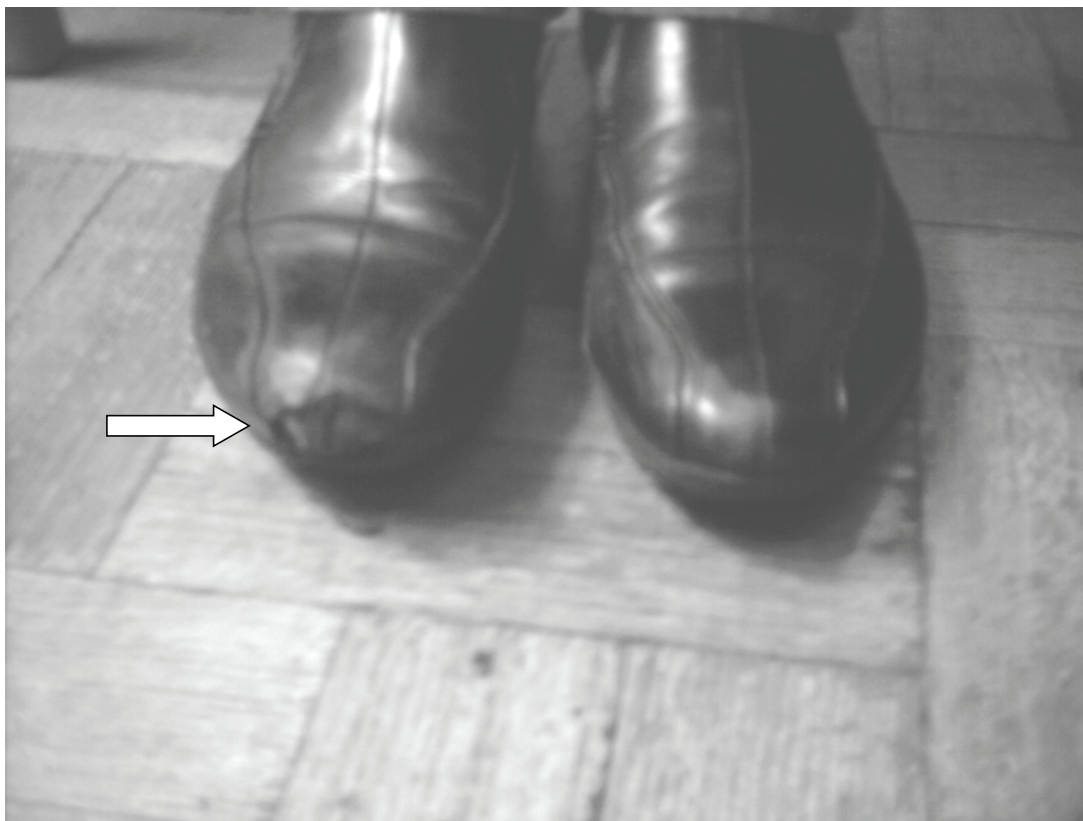


Figure 1 Deterioration of the tip of the right shoe. Reproduced with permission.

helpful in the diagnosis of Machado-Joseph disease as early ataxic signs before the occurrence of truncal ataxia. Likewise, we show that uneven focal deterioration of the shoe tip as a result of longstanding foot tapping tic, could represent a pathognomonic, albeit unusual, manifestation of Tourette syndrome.

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