

Video Abstracts

A Case of Peripherally Induced Task-Specific “Lipstick Dystonic Tremor”

Francesco Cavallieri^{1,2,3*}, Franco Valzania¹, Laurent Vercueil^{4,5}, Elena Moro^{3,4} & Valérie Fraix^{3,4}

¹Neurology Unit, Neuromotor & Rehabilitation Department, Azienda USL-IRCCS di Reggio Emilia, Reggio Emilia, IT, ²Clinical and Experimental Medicine PhD Program, University of Modena and Reggio Emilia, Modena, IT, ³Movement Disorders Unit, Neurology Department, Grenoble Alpes University Hospital, Grenoble, FR, ⁴Université Grenoble Alpes, Inserm, Grenoble Institut Neurosciences, Grenoble, FR, ⁵Neurophysiology Unit, Neurology Department, Grenoble Alpes University Hospital, Grenoble, FR

Abstract

Background: Peripherally induced movement disorders (PIMDs) represent a rare and debated complication of peripheral trauma.

Phenomenology Shown: We report a case of task-specific “lipstick” jerky dystonic tremor as a consequence of traumatic shoulder injury, successfully treated with EMG-guided botulinum toxin injections.

Educational Value: This case expands the phenotypic spectrum of PIMDs, with a visual example of a task-specific dystonic tremor after peripheral trauma, and the efficacy of EMG-guided botulinum toxin treatment in the setting of posttraumatic dystonic tremor.

Keywords: Dystonic tremor, lipstick, posttraumatic, task specific, botulinum toxin, movement disorders, electromyography, peripherally induced

Citation: Cavallieri F, Valzania F, Vercueil L, Moro E, Fraix V. A Case of Peripherally Induced Task-Specific “Lipstick Dystonic Tremor”. Tremor Other Hyperkinet Mov. 2019; 9. doi: 10.7916/tohm.v0.689

*To whom correspondence should be addressed. E-mail: francesco.cavallieri@unimore.it

Editor: Elan D. Louis, Yale University, USA

Received: June 24, 2019; **Accepted:** September 5, 2019; **Published:** October 1, 2019

Copyright: © 2019 Cavallieri et al. This is an open-access article distributed under the terms of the Creative Commons Attribution–Noncommercial–No Derivatives License, which permits the user to copy, distribute, and transmit the work provided that the original authors and source are credited; that no commercial use is made of the work; and that the work is not altered or transformed.

Funding: The authors have not declared a specific grant for this research from any funding agency in the public, commercial, or not-for-profit sectors.

Financial Disclosures: F. Cavallieri, F. Valzania, and L. Vercueil report no disclosures. E. Moro has received honorarium from Medtronic for lecturing, and research funding from Merz. V. Fraix received reimbursement of travel expenses from Merz, honoraria for scientific counselling from AbbVie, and from UCB for lecturing.

Conflicts of Interest: The authors report no conflicts of interest.

Ethics Statement: This study was performed in accordance with the ethical standards detailed in the Declaration of Helsinki. All patients that appear on video have provided written informed consent; authorization for the videotaping and for publication of the videotape was provided.

Peripherally induced movement disorders (PIMDs) represent a rare and debated complication of peripheral trauma. The phenomenology of PIMDs comprises several hyperkinetic and hypokinetic movement disorders (MDs), including dystonia, tremor, myoclonus, tics, and parkinsonian features. Cardoso and Jankovic proposed diagnostic criteria in 1995, based on the severity of the trauma, the anatomical relation to the site of the PIMDs, and the interval between trauma and onset of movement disorders.¹ Here we report a case of task-specific dystonic tremor as a consequence of traumatic shoulder injury.

A right-handed 67-year-old woman, with no neurological family history, was referred to our department with a 5-year history of involuntary movements of the right upper limb. The involuntary movements were present only when the patient put on lipstick and not while eating soup, using a knife and fork or drinking from a glass. The movements started

6 months after right shoulder injury due to an accidental fall with direct shoulder trauma with pain, tenderness and bruises in the injured area, and swelling and stiffness of the glenohumeral joint. The X-ray study performed in the emergency department did not show bone damages. The patient was sent home in an arm slings shoulder immobilizer and the symptoms disappeared in 3 weeks. Subsequently, the patient had a brief physical therapy for rehabilitation. Neurological examination revealed the presence of brief, involuntary, and repetitive movements of right arm and shoulder triggered by the action of putting on lipstick associated with a mild dystonic posturing of the right hand (Video 1–3). The involuntary movements were not influenced by distraction with motor tasks and suggestibility was not present. Electromyography (EMG) recordings showed pseudorhythmic bursts (mean burst frequency: 5 Hz, mean burst duration: 78 ms) superimposed on voluntary contractions of the right



Video 1. Video Recorded 5 months after the Last Botulinum Toxin Injection. The action of putting on lipstick causes the appearance of brief, involuntary, and repetitive movements of right arm and shoulder associated with a mild dystonic posturing of the right hand in particular of the fourth and fifth fingers. Moreover, it is interesting to note that the involuntary movements are more evident when the hand is moving from left- to right-hand side, and less on the opposite direction; for instance, dystonic tremor is enhanced when the body part moves in the opposite direction compared to the dystonia and decreases at the so-called null point. During the video, V. F. is injecting botulinum toxin type A (BTX-A) into teres major muscle using EMG guidance.



Video 2. Imitation of the action of putting on lipstick. While the Patient Is Mimicking the Action of Putting on Lipstick, No Myoclonic Jerks Appear.

pectoralis major, pectoralis minor, deltoid, and teres major muscles, triggered by the action of putting on lipstick. Brain MRI revealed only age-related white matter hyperintensities, while cervical MRI was normal. EMG-guided botulinum toxin type A (BTX-A) injections into the clavicular head of the pectoralis major were performed. After 3 weeks, involuntary movements were reduced. Then, EMG-guided BTX-A treatment was repeated every 5 months with 100 Botox® units injected into the pectoralis major, deltoid, and teres major muscles, leading to persistent functional improvement (Video 4).

PIMDs represent a controversial and not yet completely understood issue in the field of MDs, particularly regarding their pathophysiological mechanisms. The relationship between the trauma and movement

disorder onset may be difficult to ascertain due to the interval between both events and a coincidence may be difficult to exclude. In addition, the heterogeneity of phenomenology of PIMDs also increases the challenges in the understanding of PIMDs pathophysiology. The rarity of PIMDs makes even more difficult the investigation of this debated condition. In our case, it seems unlikely that the movement disorder was coincidental: the severity of the trauma, and the anatomical and temporal relation between shoulder injury and the onset of involuntary movements fulfill the proposed diagnostic criteria by Cardoso and Jankovic.¹ Moreover, the lack of family history and the absence of other possible obvious causes allow us to assume that our patient has developed a PIMD.



Video 3. Action of drinking in a plastic glass. The Action of Drinking in a Plastic Glass Does Not Cause the Appearance of Myoclonic Jerks.



Video 4. Video Recorded 2 Months after EMG-Guided Botulinum Toxin Type A (BTX-A) Injection into the Pectoralis Major, Deltoid, and Teres Major Muscles. BTX-A treatment has led to an almost complete resolution of the involuntary movements.

It has been previously reported that peripheral trauma can lead rarely to the development of focal task-specific dystonia very similar to that seen in primary dystonia.^{1,2,3} In our patient, putting on lipstick was the only specific actions which triggered the involuntary movements that we defined as task-specific dystonic tremor. There are examples of exquisitely position specific tremors that are regular, about 5–6 Hz in frequency, and can be only triggered by a specific position with a good response to botulinum toxin treatment. However, in our case the involuntary movements did not appear while the patient was mimicking the action of putting on lipstick even if the posture assumed by the right arm was exactly the same assumed during the action of putting on lipstick. This suggests that the tremor was not position specific but task specific. The literature data suggest that botulinum toxin symptomatic treatment was found to be successful in only 20% of PIMDs cases, as seen in our patient.³ In conclusion, this case expands the phenotypic

spectrum of PIMDs, with a visual example of a task-specific dystonic tremor after peripheral trauma. Moreover, it shows the efficacy of EMG-guided botulinum toxin treatment in the setting of posttraumatic dystonic tremor.

References

1. Cardoso F, Jankovic J. Peripherally induced tremor and parkinsonism. *Arch Neurol* 1995;52:263–270. doi: 10.1001/archneur.1995.00540270055019
2. Frucht S, Fahn S, Ford B. Focal task-specific dystonia induced by peripheral trauma. *Mov Disord* 2000;15:348–350. doi: 10.1002/1531-8257(200003)15:2%3C348::AID-MDS1028%3E3.0.CO;2-7
3. van Rooijen DE, Geraedts EJ, Marinus J, Jankovic J, van Hilten JJ. Peripheral trauma and movement disorders: a systematic review of reported cases. *J Neurol Neurosurg Psychiatry* 2011;82:892–898. doi: 10.1136/jnnp.2010.232504