OPEN ACCESS Freely available online

Tremor and Other Hyperkinetic Movements

Video Abstracts

Inspiratory Myoclonus

Marieke C. J. Dekker[1*], Kajiru G. Kilonzo[1], William P. Howlett[1], Mark Guttman[2] & Esther Cubo[3]

[1] Department of Internal Medicine, Kilimanjaro Christian Medical Centre, Moshi, TZ, [2] Department of Internal Medicine, Division of Neurology, University of Toronto, Toronto, CA, [3] Neurology Department, Hospital Universitario Burgos, Burgos, SP

Abstract

Background: Movement disorders of respiration are rare and are restricted to a phase of the respiratory cycle.

Phenomenology Shown: The intermittent inspiratory myoclonus in this patient with post-anoxic encephalopathy is likely to be of brainstem origin.

Educational Value: Rare movement disorders can be identified even in remote areas of the world where access to neurological care is limited.

Keywords: Inspiratory, myoclonus, diaphragmatic, post-anoxic encephalopathy, Tanzania

Citation: Dekker MCJ, Kilonzo KG, Howlett WP, Guttman M, Cubo E. Inspiratory myoclonus. Tremor Other Hyperkinet Mov. 2019; 9. doi: 10.7916/3qs5-cv76

*To whom correspondence should be addressed. E-mail: marieke@zwets.com

Editor: Elan D. Louis, Yale University, USA

Received: December 2, 2018 **Accepted:** January 17, 2019 **Published:** February 18, 2019

Funding: None.

Financial Disclosures: E.C. has disclosures to make regarding travel grants from Allergan, Abbvie, and Boston Pharmaceutical. Clinical trials were supported by Roche, Abbvie, Neuroderm, Teva.

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

Ethics Statement: All patients that appear on video have provided written informed consent; authorization for the videotaping and for publication of the videotape was provided.

Three years previously, a 77-year-old Tanzanian male developed post-anoxic encephalopathy after cardiac arrest and pacemaker implantation. After cardiac arrest, there was residual pseudobulbar and cerebellar dysarthria, spastic-dystonic quadriplegia, and largely preserved cognition. Follow-up computed tomography (CT) scans of the brain showed moderate atrophy. He was using the anticoagulant dabigatran. Nine months before admission, without an obvious precipitating event, he developed jerky movements that hindered breathing and eating, and which resolved after 2 weeks. Two days before admission these same movements recurred, and led to respiratory difficulties that occurred during sleep and when awake. Upon examination he had stable vital signs, was alert, and had labored breathing. His oxygen saturation was 98% when on oxygen. When auscultation was performed, basal crepitations were detected. Apart from the pre-existent neurological abnormalities, there were now inspiratory, involuntary jerky movements of the chest, throat, and neck that were not present during expiration. Inspection and palpation of the flanks and abdomen were normal and the patient did not experience hiccups or movements of the abdominal wall (Video 1). Magnetic resonance imaging of the brain was not possible because of a pacemaker. CT scans of the brain, chest, and diaphragm were normal. A chest X-ray showed bilateral infiltrates; however, the laboratory test results were normal. Clonazepam and sodium valproate were associated with mild improvement. Three days after admission, he died from aspiration pneumonia and respiratory failure.

The working diagnosis was intermittent inspiratory myoclonus of brainstem origin in a patient with post-anoxic encephalopathy. It may have recurred after a previous, self-limiting episode of possible brainstem ischemia or hemorrhage undetected by a CT scan of the brain. Myoclonus was less likely to be cortical because the patient was alert throughout. The fatal outcome was likely due to respiratory insufficiency rather than progressive brainstem involvement. Diaphragmatic flutter or dyskinesia was considered, but our patient had a normal expiration phase and was not tachypnoeic.[3] Belly dancer dyskinesia would, as the name suggests, affect the abdominal wall primarily and is a lower frequency and amplitude-movement disorder.[1–3] He did not display Lance Adams (post-anoxic) myoclonus at any time after his cardiac arrest. Truncal spinal myoclonus or a voluntary component is

Video 1. **Jerky, Audible Inspiration Phase.** The expiration phase is largely unaffected.

unlikely. Encephalitis is less likely to be due to the recurrent nature of movement disorders with a symptom-free interval of several months and normal consciousness.

This study's limitations are the short duration of the video and lack of several investigations inherent to the low-resource setting. This is one of few recordings of this phenomenon,[3] stressing the potentially fatal outcome, and originating from an area that is underserved by neurologists.

Acknowledgments

Our sincere gratitude to the late patient and his family for their cooperation and understanding. The partner of this patient and the family furthermore approve of being acknowledged.

References

1. Rigatto M, De Medeiros NP. Diaphragmatic flutter. Report of a case and review of literature. *Am J Med* 1962;32:103–109. doi: 10.1016/0002-9343(62)90186-9

2. Iliceto G, Thompson PD, Day BL, Rothwell JC, Lees AJ, Marsden CD. Diaphragmatic flutter, the moving umbilicus syndrome and 'belly dancer's' dyskinesia. *Mov Disord* 1990;5:15–22. doi: 10.1002/mds.870050105

3. Philips JR, Eldridge FL. Respiratory myoclonus (Leeuwenhoek's disease). *N Engl J Med* 1973;289:1390–1395. doi: 10.1056/NEJM197312272892603