

“A disorder which occurs on standing”: The Earliest Account of Orthostatic Tremor by Pazzaglia

Roberto Erro, RE, MD,^{1,2,*} Elena Antelmi, EA, MD,^{1,3} Kailash P. Bhatia, KPB, MD, FRCP¹

Abstract: The earliest account of orthostatic tremor (OT) dates back to 1970, when Pazzaglia and colleagues reported on three patients with a peculiar disorder only occurring on standing. Although Pazzaglia et al. did not use the term OT, they first provided both the clinical and neurophysiological features of such a condition.

“The farther backward you can look, the farther forward you are likely to see.” Winston Churchill

Although the term “orthostatic tremor” (OT) first entered the medical literature in 1984, when Heilman described 3 patients with a movement disorder characterized by tremor of the legs upon standing,¹ the earliest account of OT dates back to 1970.² Pazzaglia et al. reported on, in the Italian journal *Rivista Sperimentale di Freniatria* (Fig. 1), 3 patients affected with an “unusual disorder of erect standing position” featuring



Figure 1 Cover of the issue containing the paper by Pazzaglia et al.

tremor of the legs, which started with a short latency when they stood.²

When Pazzaglia et al. saw the first of these 3 patients, in January 1968, they were left perplexed (translated from Italian):

This observation left us deeply puzzled, since we were unable to find any pathophysiological explanations for such a peculiar disorder of erect standing position; so puzzled to doubt about its true organic nature.

This patient had no other neurological disturbances apart from an extreme difficulty to stand:

There were fast and regular oscillations of the antigravitary muscles, exclusively present on standing. The subject put effort into avoiding falls by widening of his feet, bending over the trunk, or by putting weight onto one leg. [...] After a few seconds, he had to lean onto something or to walk: In this way, everything stopped immediately, with his obvious relief. This was the only symptom.

They also provided the electrophysiological features of OT, although the first description in this regard has been generally attributed to Thompson, who, in 1986, described 1 OT patient noting a 16-Hz tremor of the legs when standing.³ Pazzaglia et al. found a 14-to 16-Hz tremor of the legs on standing, which was abolished when the subject leaned onto something (Fig. 2).² Notably, they also picked up a postural tremor of the arms at a lower frequency (Fig. 2),² which has been subse-

¹Sobell Department of Motor Neuroscience and Movement Disorders, University College London (UCL) Institute of Neurology, London, United Kingdom; ²Dipartimento di Scienze Neurologiche e del Movimento, Università di Verona, Verona, Italy; ³Dipartimento di Scienze Mediche e Chirurgiche, DIMEC, University of Bologna, Bologna, Italy

*Correspondence to: Dr. Roberto Erro, National Hospital for Neurology and Neurosurgery, Queen Square, London WC1N 3BG, United Kingdom; E-mail: erro.roberto@gmail.com

Keywords: orthostatic tremor, tremor, standing.

Relevant disclosures and conflicts of interest are listed at the end of this article.

Received 16 October 2014; accepted 2 November 2014.

Published online 24 February 2015 in Wiley InterScience (www.interscience.wiley.com). DOI:10.1002/mdc3.12127

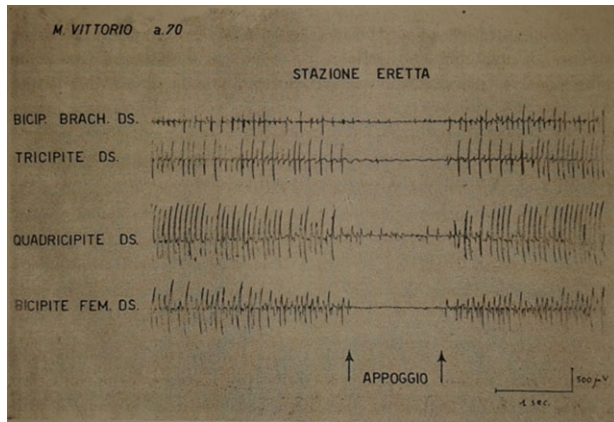


Figure 2 Original picture of the article by Pazzaglia et al. (with permission). Up to down, surface electromyography recording of the right biceps, triceps, rectus femoris, and biceps femoris muscles. In the leg, a 14- to 16-Hz tremor can be seen, which stops when the patient leans onto something (arrows). In the arm, a tremor at a lower frequency is seen.

quently recognized as part of the condition in the majority of OT patients.⁴

The other 2 patients in their series had similar symptoms in the context of another neurological disorder: tabes dorsalis (syphilitic myelopathy) and flaccid paralysis likely resulting from a spinal stroke, respectively. On electrophysiology, the 2 patients had less regular contractions of the leg muscles at a lower frequency of approximately 10 to 12 Hz.² Currently, we would diagnose these 2 patients with pseudo-OT.⁴

Very interestingly, Pazzaglia et al. wondered whether such a peculiar condition was associated with inappropriate postural responses:

With the aim to establish whether such hyperkinesias were triggered by postural responses on standing or were simply related to the vertical position of the body, the first patient underwent tilt table testing. We hence observed that the vertical position of the body in the absence of postural loading did not elicit any symptoms.

They therefore speculated that the disorder was a result of the dysfunction of the regulating system of the postural tonus,

involving, at the same time, the spinal and supraspinal level.² Thirty-six years later, Spiegel provided similar evidence using a slightly more sophisticated technology,⁵ but such a hypothesis has not yet been entirely settled.

Author Roles

(1) Conception; (2) Manuscript: A. Writing of the First Draft; B. Review and Critique.

R.E.: 1, 2A

E.A.: 2B

K.P.B.: 2B

Acknowledgments

The authors are grateful to Luigi Tagliabue, editor of *Rivista Sperimentale di Freniatria*, who kindly allowed the publication of the original picture of the article published in 1970 by Pazzaglia et al.

Disclosures

Funding Sources and Conflicts of Interest: The authors report no sources of funding and no conflicts of interest.

Financial Disclosures for previous 12 months: R.E. received funding for travel from Ipsen. K.P.B. receives royalties from Oxford University Press and received funding for travel from GlaxoSmithKline, Orion Corporation, Ipsen, and Merz Pharmaceuticals.

References

1. Heilman KM. Orthostatic tremor. *Arch Neurol* 1984;41:880–881.
2. Pazzaglia P, Sabatini L, Lugaesi E. Su di un singolare disturbo della stazione eretta (osservazione di tre casi) [On an unusual disorder of erect standing position (observation of three cases)]. *Riv Freniatr* 1970;96:450–457.
3. Thompson PD, Rothwell JC, Day BL, Berardelli A, Dick JP, Kachi T, Marsden CD. The physiology of orthostatic tremor. *Arch Neurol* 1986;43:584–587.
4. Erro R, Bhatia KP, Cordivari C. Shaking on standing: a critical review. *Mov Disord Clin Pract* 2014;1:173–179.
5. Spiegel J, Krick C, Fuss G, Sood D, Becker G, Dillmann U. Orthostatic tremor during modification of standing. *Mov Disord* 2006;21:173–178.