Neurological Sciences (2019) 40:2205–2207
https://doi.org/10.1007/s10072-019-03922-3

LETTER TO THE EDITOR

Pseudo-orthostatic tremor: description of a not typical case

Aniello Iovino[1] · Silvio Peluso[1] · Fiore Manganelli[1] · Stefano Tozza[1] · Marcello Esposito[1]

Received: 8 December 2018 / Accepted: 4 May 2019 / Published online: 12 June 2019

Dear Sir,

Pseudo-orthostatic tremor (PsOT) is a rare movement disorder recently included in the latest classification of tremors [1]. It shares notable clinical features with primary orthostatic tremor (OT); in fact, it presents at lower limbs mainly during standing, and it causes unbalance that improves during walking.

Actually, PsOT shows a lower frequency in comparison to the primary form because PsOT frequency is around 10 Hz whereas primary OT typically presents a frequency higher than 13 Hz [2].

Here, we describe a case of pseudo-OT not associated with other neurologic disorders.

Case report

A 23-year-old man was admitted to our hospital because of a severe leg tremor. Patient reported a mild leg tremor since he was 14, and only recently, after an unexplained weight loss, the movement disorder worsened and became disabling. Clinical examination showed severe legs shaking in upright position that was almost not evident during walking. Postural tremor was also present only at lower limbs. There was no family history of tremor or parkinsonism. Patient underwent many investigations: brain MRI and DAT Spect scan, colonoscopy were normal, blood tests for infectious, and autoimmune diseases were negative [3]. Patient was also studied with evoked potentials [4] and transcranial magnetic stimulation exploring the central motor pathway according to the published procedure [5]. Findings of TMS and evoked potentials studies did not show abnormalities. A neurophysiological assessment of the tremor was performed by a video multichannel EMG recording in upright position and kneeling on all fours. Surface electrodes were used to record electrical muscle activity, and limbs oscillation was detected by accelerometers measuring tremor frequency as the peak of the power spectrum determined by Fourier transform. During standing, EMG were recorded from rectus and biceps femoris, and the accelerometer was attached at the thigh, on all four position EMG surface electrodes were placed on biceps and triceps and the accelerometer on the arm. EMG bursts were almost regularly alternating in antagonist muscles with a duration of 80–200 msec and amplitude of 100–400 μV (Fig. 1). Tremor analysis showed a fixed 10.5 Hz tremor that was detectable at lower limbs only when standing and at upper limbs when patient was on all fours position. The evidence of a tremor at the same frequency only in those conditions was suggestive of an orthostatic tremor that we defined as pseudo-OT because of the lower frequency comparing with primary forms. A 7 Hz postural tremor was also recorded only at lower limbs. The use of distractive maneuvers during the tremor analysis showed no change of frequency and suppression of the tremor excluding a functional origin of the movement disorder (video).

A treatment with gabapentin was started with poor tremor reduction, also other medications were tried, propranolol and clonazepam, without benefits.

Patient is currently under follow-up assessment every 6 months with no evidence of clinical changes. No other neurologic or non-neurologic disorder occurred during this period. Two years after the onset of the tremor patient underwent again a brain MRI scan that was still normal. There was also no change at the follow-up tremor analysis which was conducted with a video EMG multichannel recording following the same procedure of the previous one.

Electronic supplementary material The online version of this article (https://doi.org/10.1007/s10072-019-03922-3) contains supplementary material, which is available to authorized users.

✉ Marcello Esposito
marcelloesposito@live.it

[1] Department of Neurosciences, Reproductive Sciences and Odontostomatology, University of Naples Federico II, Via Pansini, 5, Naples, Italy

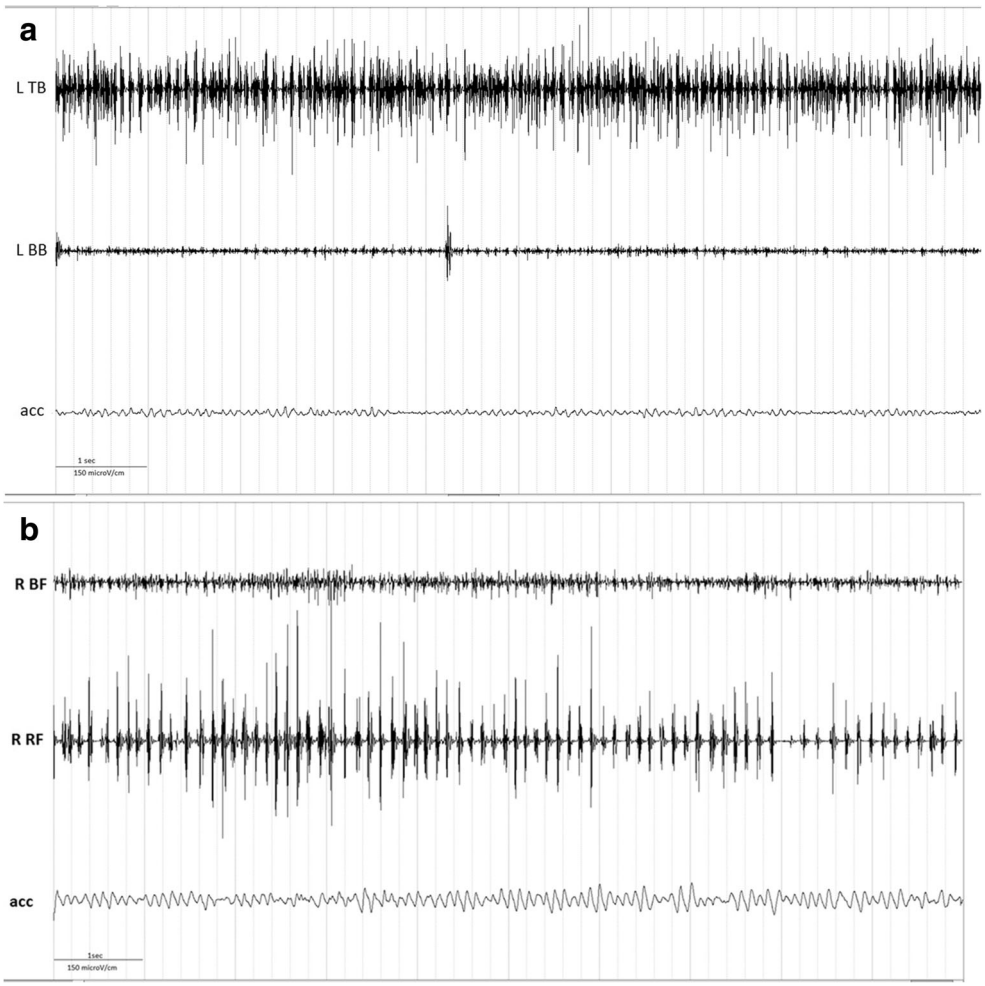

Fig. 1 Multichannel EMG recording. **a** Recording from the upper limb when the patient was on all fours position, accelerometer was attached to the arm. **b** Recording from the lower limb in upright position, accelerometer was attached to the thigh. L TB: left triceps brachii; L BB: left biceps brachii; R BF: right biceps femoris; R RF: right rectus femoris; acc: accelerometer

Discussion

The description of this case would be consistent with an idiopathic not progressive tremor with major clinical features suggestive of a PsOT. Electrophysiology study revealed a tremor occurring at lower or upper limbs only when loaded by body weight proving the orthostatic nature of the tremor. Moreover, upper and lower limbs shared the same tremor frequency showing a common drive of the tremor. On this base, the patient could have an idiopathic low-frequency OT, that is indeed a PsOT, complicated with a postural leg tremor. Actually, the combination of postural tremor and OT was already reported in a previous study although the former was described to occur only at the upper limbs and in association with primary forms of OT [6].

OT and PsOT should be distinguished by another weight-bearing hyperkinetic movement disorder that is the orthostatic myoclonus (OM). There are only few clinical differences between OM and OT, and neurophysiological assessment is necessary to confirm the diagnosis. The multichannel EMG recording performed in our case showed findings consistent with tremor because EMG bursts were alternating and rhythmic with no evidence of spread of jerks from proximal to distal limb muscles. Furthermore, we found a tremor at the arm when the patient was on all fours position that is a condition inducing tremor reported only in descriptions of OT and not of OM. Finally, there was no evidence of negative myoclonus [7].

Based on those considerations, the movement disorder of the present case could be defined as an atypical PsOT. The combination of a postural tremor at lower limbs with a PsOT was not previously reported, moreover, that movement disorder can be considered atypical especially because the tremor was not associated to any other neurological disorder, and there was no evidence of progression. This would be the first description of an idiopathic pseudo-OT in fact previous studies found other neurological conditions, like multiple sclerosis or parkinsonism, in addition to the tremor [8, 9].

The description of a new case of PsOT with such atypical features would be of interest because it may extend the clinical spectrum of this rare movement disorder.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

References

1. Bhatia KP, Bain P, Bajaj N, Elble RJ, Hallett M, Louis ED, Raethjen J, Stamelou M, Testa CM, Deuschl G (2018) Tremor task force of the International Parkinson and Movement Disorder Society. Consensus statement on the classification of tremors. From the task force on tremor of the International Parkinson and Movement Disorder Society. Mov Disord 33(1):75–87
2. Panida P, Qiping P, Yu (2005) Clinical and neurophysiologic spectrum of orthostatic tremor: case series of 26 subjects. Mov Disorder Soc 20(11):1455–1461
3. Dubbioso R, Pappatà S, Quarantelli M, D'Arco F, Manganelli F, Esposito M, Santoro L (2013) Atypical clinical and radiological presentation of cryptococcal choroid plexitis in an immunocompetent woman. J Neurol Sci 334(1–2):180–182
4. Iodice R, Dubbioso R, Topa A, Ruggiero L, Pisciotta C, Esposito M, Tozza S, Santoro L, Manganelli F (2015) Electrophysiological characterization of adult-onset Niemann-pick type C disease. J Neurol Sci 348(1–2):262–265
5. Dubbioso R, Ranucci G, Esposito M, Di Dato F, Topa A, Quarantelli M, Matarazzo M, Santoro L, Manganelli F, Iorio R (2016) Subclinical neurological involvement does not develop if Wilson's disease is treated early. Parkinsonism Relat Disord 24:15–19. https://doi.org/10.1016/j.parkreldis.2016.01.024
6. Yaltho TC, Ondo WG (2014) Orthostatic tremor: a review of 45 cases. Parkinsonism Relat Disord 20:723–725. https://doi.org/10.1016/j.parkreldis.2014.03.013
7. Hassan A, van Gerpen JA (2016) Orthostatic tremor and orthostatic myoclonus: weight-bearing hyperkinetic disorders: a systematic review, new insights, and unresolved questions. Tremor Other Hyperkinet Mov (N Y) 6:417
8. Baker MR, Fisher KM, Lai HM, Duddy ME, Baker SN (2009) Slow orthostatic tremor in multiple sclerosis. Moviment Disorders 24(10):1550–1553. https://doi.org/10.1002/mds.22630
9. Infante J, Berciano J, Sánchez-Juan P, García A, Di Fonzo A, Breedveld G, Oostra B, Bonifati V (2009) Pseudo-orthostatic and resting leg tremor in a large Spanish family with homozygous truncating parkin mutation. Mov Disorders. 24(1):144–147