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Case report

Mirror movements or functional tremor masking organic tremor

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ABSTRACT

Background: Functional tremors can be diagnosed based on clinical and physiologic criteria such as entrainment, suggestibility, distractibility, variable nature with the associated clinical history of psychosomatic co-morbidities. The current case report highlights the underrecognized utility of neurophysiology in the correct diagnosis of tremors, providing useful clinical and neurophysiologic insights into clinical and physiological assessment of tremors.
Case report: A 62-year-old woman with a past medical history of polio was referred by a movement disorders neurologist for evaluation of tremor with concerns of a likely functional etiology. On first assessment there were findings notable for a possible organic etiology, but upon subsequent evaluation the tremor was noted to be variable and entrainable, suggestive of a functional etiology. Neurophysiological tremor study could identify an underlying organic tremor (likely of multi-factorial etiology). Tremor entrainment with contralateral hand tapping could be mirror movements or functional movements, as the underlying organic tremor was not entrained. The amplitude of mirrored movement was commensurate with the tapping amplitude.
Discussion: Functional tremors may mask an underlying organic tremor. Additionally, motor overflow which may happen especially with large amplitude movements may masquerade as mirror movements, which can be difficult to differentiate from an entrained functional tremor. Objective physiology and refinement of the current clinical and physiologic tremor evaluation techniques may help identify an underlying organic etiology.

1. Introduction

Functional tremors are the most common type of functional movement disorders, but the etiopathogenesis is poorly understood (Edwards and Bhatia, 2012; Edwards et al., 2014). Clinical and physiologic diagnostic criteria have been proposed for these disorders which can be difficult to diagnose (Hallett, 2016, 2018; Schwingenschuh et al., 2011, 2016; Fahn and Williams, 1988). Clinical characteristics such as entrainment, suggestibility, distractibility, variable nature, presence of co-contraction sign taken together with the history of psychosomatic co-morbidities and response to treatment, aid in the diagnostic certainty (Shill and Gerber, 2006). Clinical neurophysiology can further aid in the diagnosis; however, its utility in identifying an underlying organic tremor in a diagnosed case of functional tremor is not well recognized.

2. Case report

We report a 62-year-old woman who was referred to us by a movement disorders neurologist for evaluation of bilateral upper extremity tremors with concerns for a likely functional etiology. She presented with a history of bilateral upper extremity and voice tremor notably worse over the last 2–3 years, which waxes and wanes based on her mood. She endorsed a dissociative psychiatric disorder, PTSD with concomitant depression, reported her tremor being worse when she feels 'not quite like herself', and acknowledged that her mood disorder led to bouts of worsening tremor. Of note, she reported a history of polio, contracted at the age of 10 months with worse affliction of her right arm and left leg. Her relevant neuropsychiatric medications included Modafinil and Fluoxetine for alertness and mood disorder, respectively. She was also taking Clonazepam 0.5 mg at bedtime for insomnia. She had undergone extensive diagnostic evaluation for tremor; relevant positive findings included presence of motor neuron disease per EMG/NCS (evidence for motor polyneuropathy consistent with patient's known history of poliomyelitis affecting primarily the

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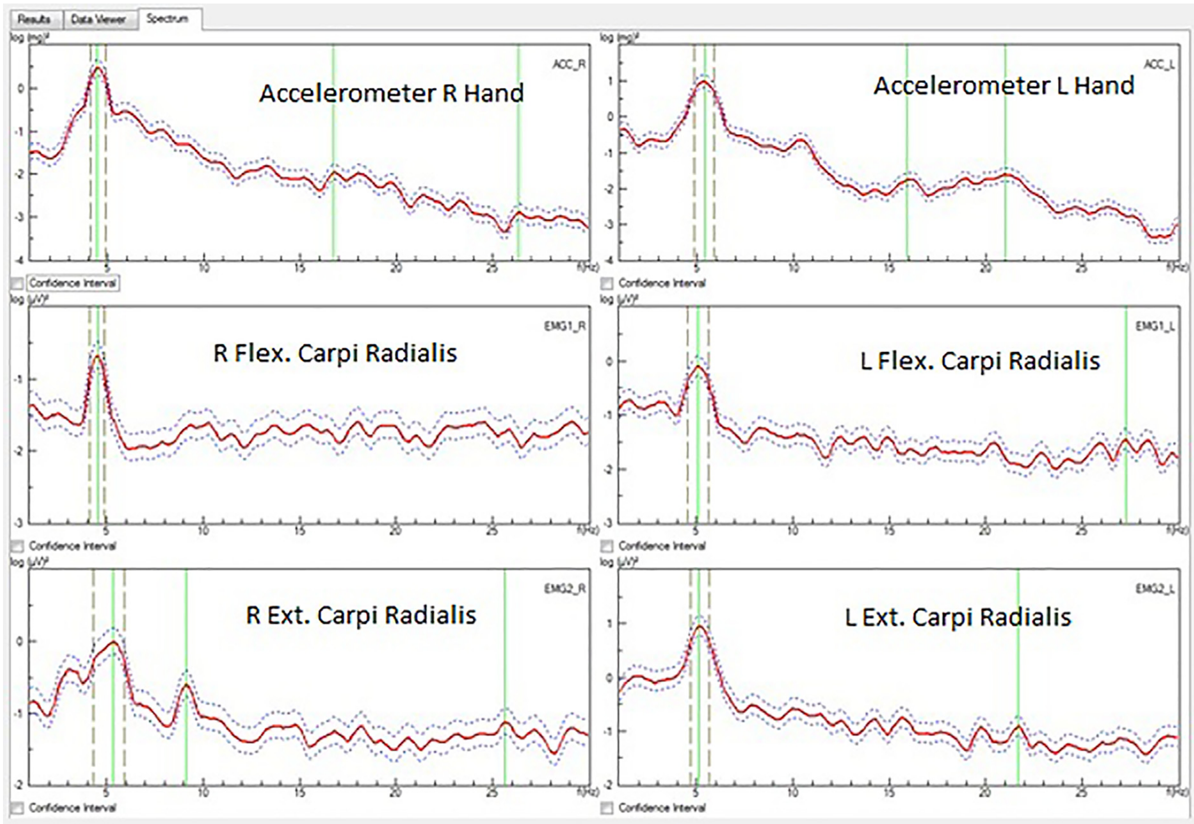

Fig. 1. Frequency spectrum of rest tremor in both hands with the spectral power on the y axis in a log scale.

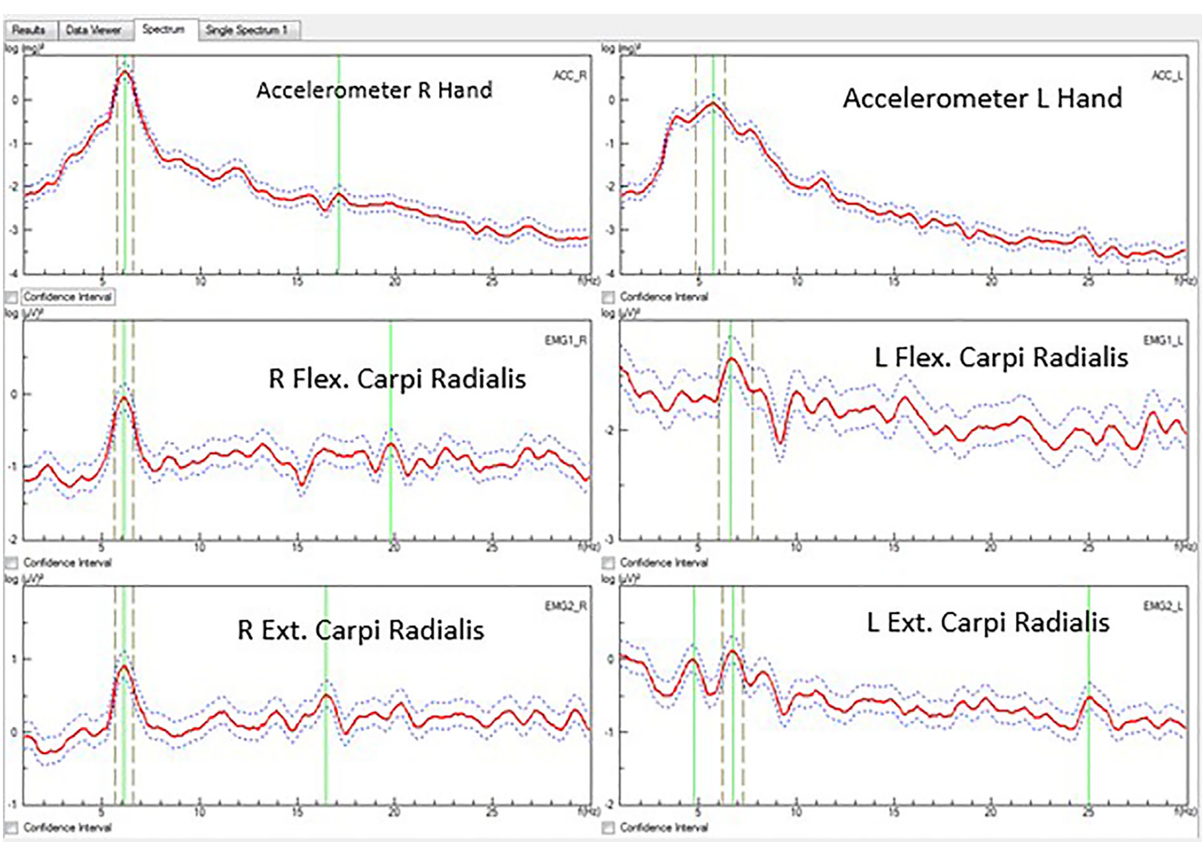

Fig. 2. Frequency spectrum of postural tremor in both hands with the spectral power noted on the y axis in a log scale.

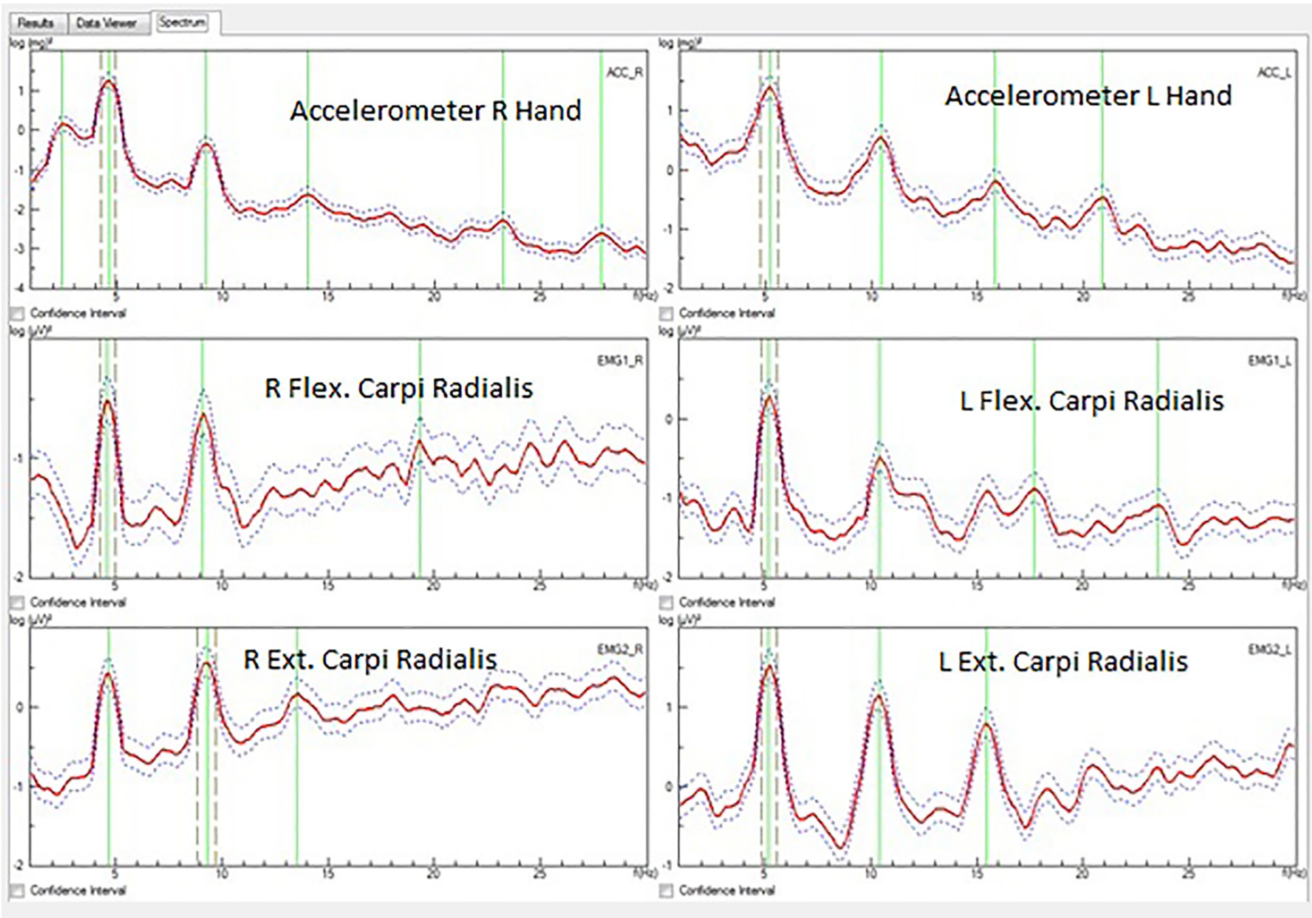

Fig. 3. Frequency spectrum of postural tremor with 2 pounds.weight loading in both hands with the spectral power noted on the y axis in a log scale.

right arm/hand) and MRI brain suggestive of periventricular white matter disease.

Clinical examination was evaluated twice the same day. At first, she was noted to have a tremor at rest and posture being slightly worse in the right upper limb compared to the left. She also had an action tremor without any clear terminal intentional worsening; however, reaching towards the target, there was jerky dysmetria which was noted with finger-nose-finger, as well as with dot approximation task and while drawing spirals. She also had associated minipolymyoclonus like movements of the fingers only in the right hand. On the second assessment, the postural tremor was noted to have a variable intensity and amplitude. We could distract the tremor to some extent, and it appeared entrainable with contralateral tapping. Of note, the patient reported that when she was evaluated the second time, she felt like she was dissociated from herself, and the tremor which came out was noticeably different and worse from what she was having when she was initially evaluated. Considering the ambiguities in the clinical examination; findings of a suggestible and entrainable tremor along with clinical findings suggestive of a potential underlying organic etiology, a tremor analysis was performed. Video 1 demonstrates the clinical examination of the 'entrainable' postural tremor during the second examination.

3. Tremor recording methods and data processing

The setup included 4-channel surface EMG and 2-channel uniaxial accelerometry; surface EMG recorded from the wrist flexors/extensors and accelerometers (Kistler. sensitivity 20 mV/g) placed at the back of the hands. Recording conditions were 30 s each and involved rest (sitting upright, arms resting on armrest, hands resting unsupported), posture with hands outstretched and arms supported, posture with hands outstretched and arms supported plus the addition of incremental weights (0.5, 1 and 2 lbs.) to each hand. Additionally, we also performed entrainment maneuvers which involved tapping of the left hand at 2 Hz and 3.5 Hz, triggered by a metronome. We also conducted an additional recording with tapping the right hand at 2 Hz and recording the postural tremor from the left hand. Free runs of postural tremor of the right hand were recorded with the performance of ballistic movements with the left hand to look for any significant decrease in tremor amplitude or arrests in ongoing tremor, and also to identify any contralateral mirror movements (Kumru et al., 2007). EMG data were rectified, and spectral analysis was performed using a stochastic time series analysis method (Timmer et al., 1998). The accelerometer and EMG peak frequencies were calculated and the total and half-width spectral power as a measure of tremor amplitude. Coherence analysis was performed between the accelerometer and EMG channels.

4. Tremor analysis results

During rest, there was a clear peak noted in EMG/accelerometer channels in both hands with peak frequency noted around 4.5 Hz on the right and around 5 Hz on the left (Fig. 1). With posture, the peak frequencies noted were slightly higher, around 6 Hz on

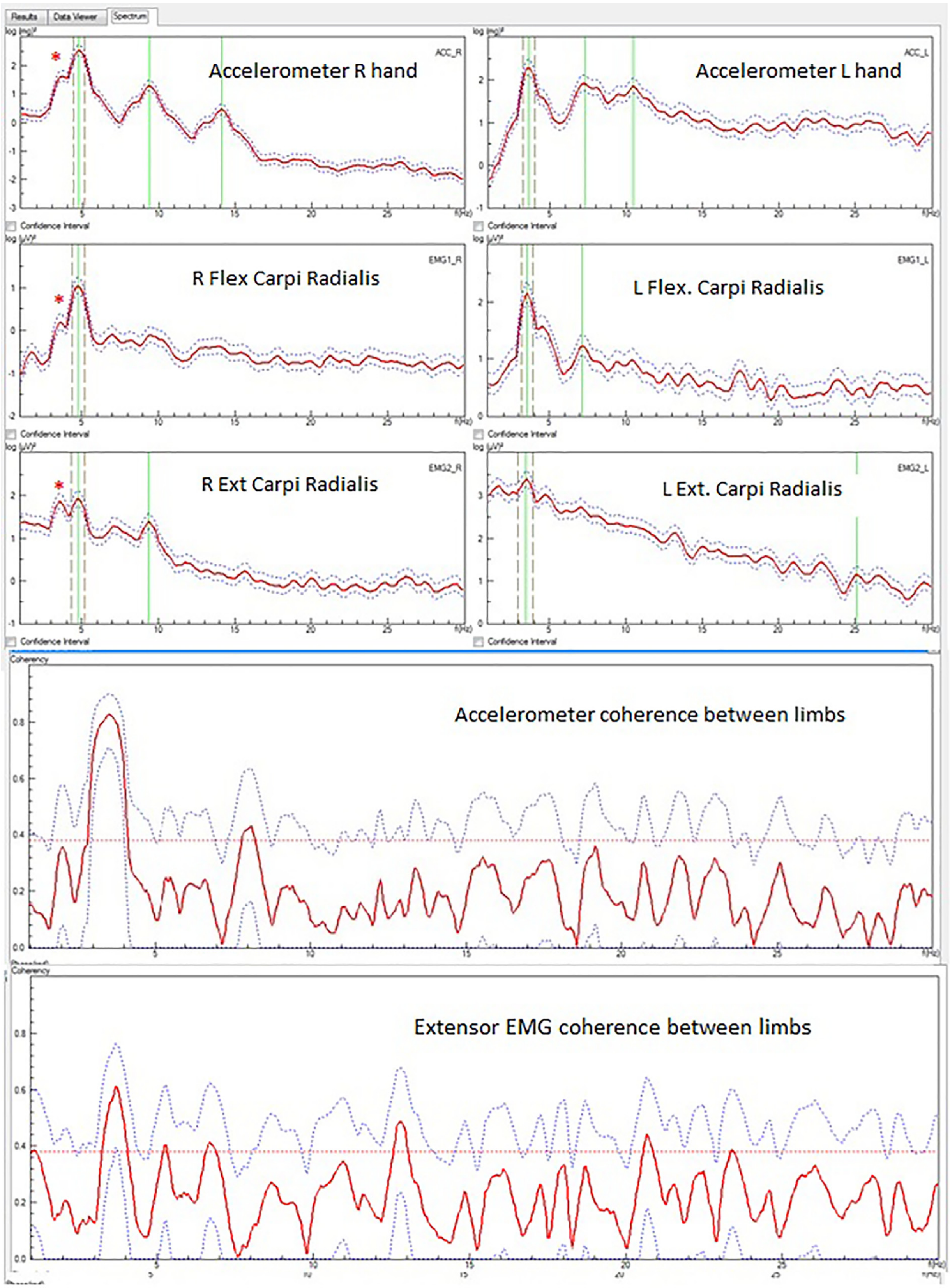

Fig. 4. Frequency spectrum of postural tremor in right hand with 3.5 Hz tapping with left hand with the spectral power noted on the y axis in a log scale in the top part of the trace. A small peak at the tapping frequency of 3.5 Hz is noted by an asterisk prior to the central tremor peak at 5 Hz in the right accelerometer and EMG traces. The bottom 2 traces demonstrate accelerometry and EMG coherence between the limbs illustrating the magnitude of coherence on the y axis and frequency spectrum on the x axis.

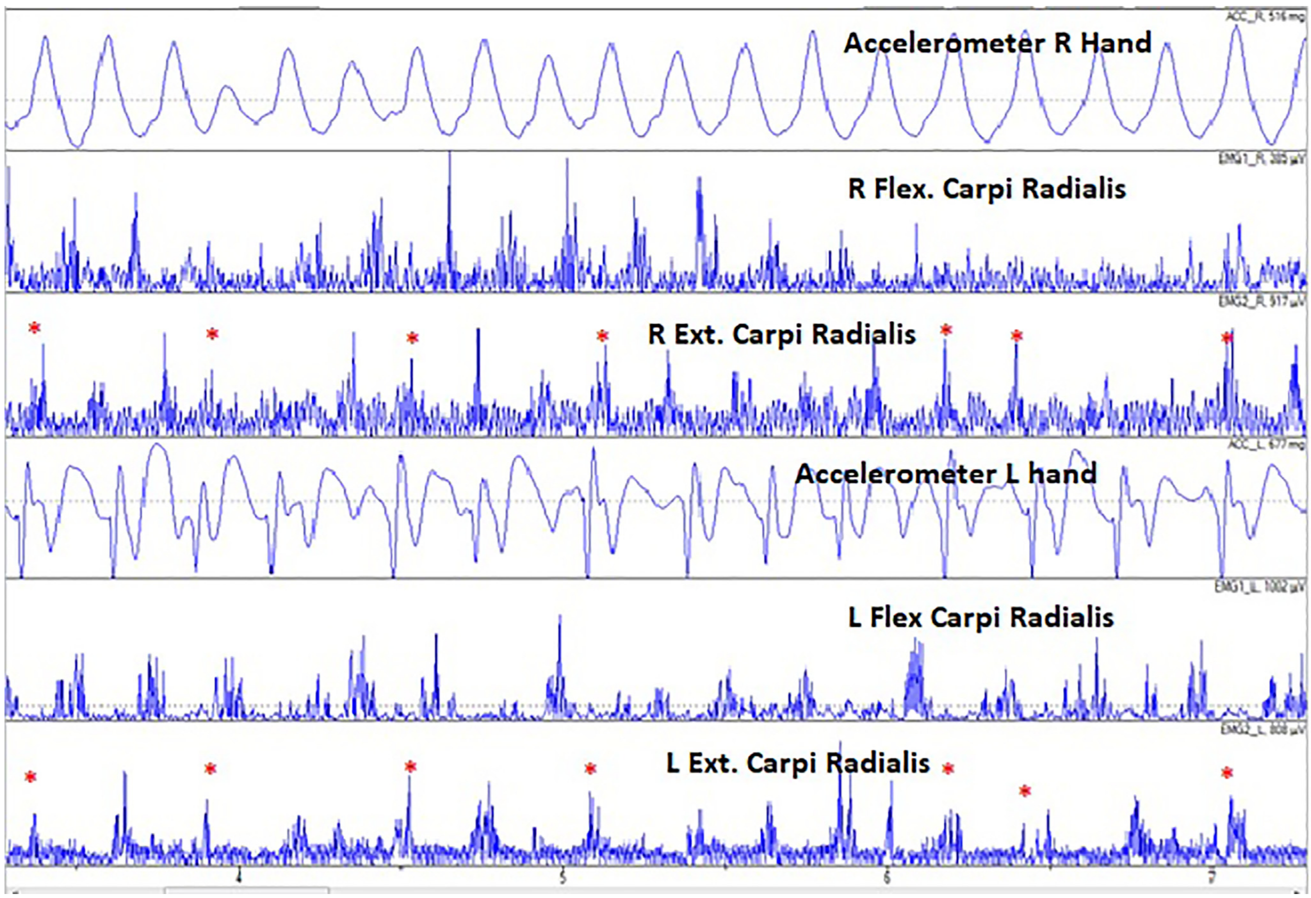

Fig. 5. 4 s recording in time domain with postural tremor recording of the right hand and 3.5 Hz tapping with left hand showing 3.5 Hz mirror movements with red asterisks in EMG channels together with the persistent central tremor component between 4 and 5 Hz as clearly noted in the right-hand accelerometer trace.

the right and between 6 and 7 Hz on the left (Fig. 2). After the addition of weights, a persistent central component at approx. 4–5 Hz was noted on the right and around 5 Hz on the left (Fig. 3). The higher frequency peaks (between 9–10 Hz and 13–14 Hz on the right; between 10–11 Hz and 15–16 Hz on the left) are harmonics of the central tremor component. Accelerometer and EMG recordings were coherent within limbs, but not between the limbs. With attempted entrainment, tapping with the left hand at 2 Hz and 3.5 Hz, 2 different frequency peaks were noted in the right hand per accelerometer and surface EMG; one at the tapping frequency with coherence between the limbs and the other being a persistent peak close to 5 Hz which was not coherent between the limbs (Fig. 4). Both accelerometer and EMG coherence are present at the tapping frequency; the accelerometer coherence between sides being noticeably more significant. Evaluating the traces in the time domain showed that there were synchronous EMG bursts with the voluntary taps (noted by red asterisks in the extensor EMG traces) that co-existed with the central tremor component with associated EMG bursts (Fig. 5). Most free runs of postural tremor recordings from the right hand while performing left wrist ballistic movements demonstrated no significant reduction in tremor amplitude of the right hand and no change in tremor frequency or tremor arrest (Fig. 6). However, occasionally when mirror movements were noted in the right hand synchronous with left hand ballistic movements a slight pause/arrest in the ongoing tremor activity was noted, concurrent with the mirror movements. Video 2 demonstrates the persistent organic tremor in the right hand with low amplitude tapping cued to a metronome and the persistent tremor with left wrist ballistic movements; however, occasional pause in tremor noted concurrent with a mirror movement.

5. Discussion

Based on history, phenomenology and the patient's own acknowledgment that her psychiatric dissociative disorder had an association with the variability in her tremor, she fits well with the clinical diagnosis of a functional tremor based on the published criteria, which were satisfied during the clinic exam (Fahn and Williams, 1988; Shill and Gerber, 2006). However, based on the tremor study we identified a clear central component in each limb, which remained consistent and non-coherent between the limbs as characteristic of an organic tremor. Additionally, this central component persisted and was clearly noted even when the tremor was modified with contralateral limb tapping during physiologic tremor analysis. She also satisfied the physiologic criteria proposed for the diagnosis of functional tremor, considering her tremor was "entrainable" (Schwingenschuh et al., 2011, 2016).

Mirror movements are more easily noticeable with larger amplitude movements which facilitate motor overflow, as also more clearly noted in the current case when performing large amplitude tapping movements during the second examination (Beck et al., 2009). The physiology reveals that the apparent "entrained tremor" could in fact be the intrusion of mirror movements. The presence of concurrent EMG bursts noted with contralateral hand tapping for tremor entrainment and similar contralateral EMG bursts noted with ballistic movements favor

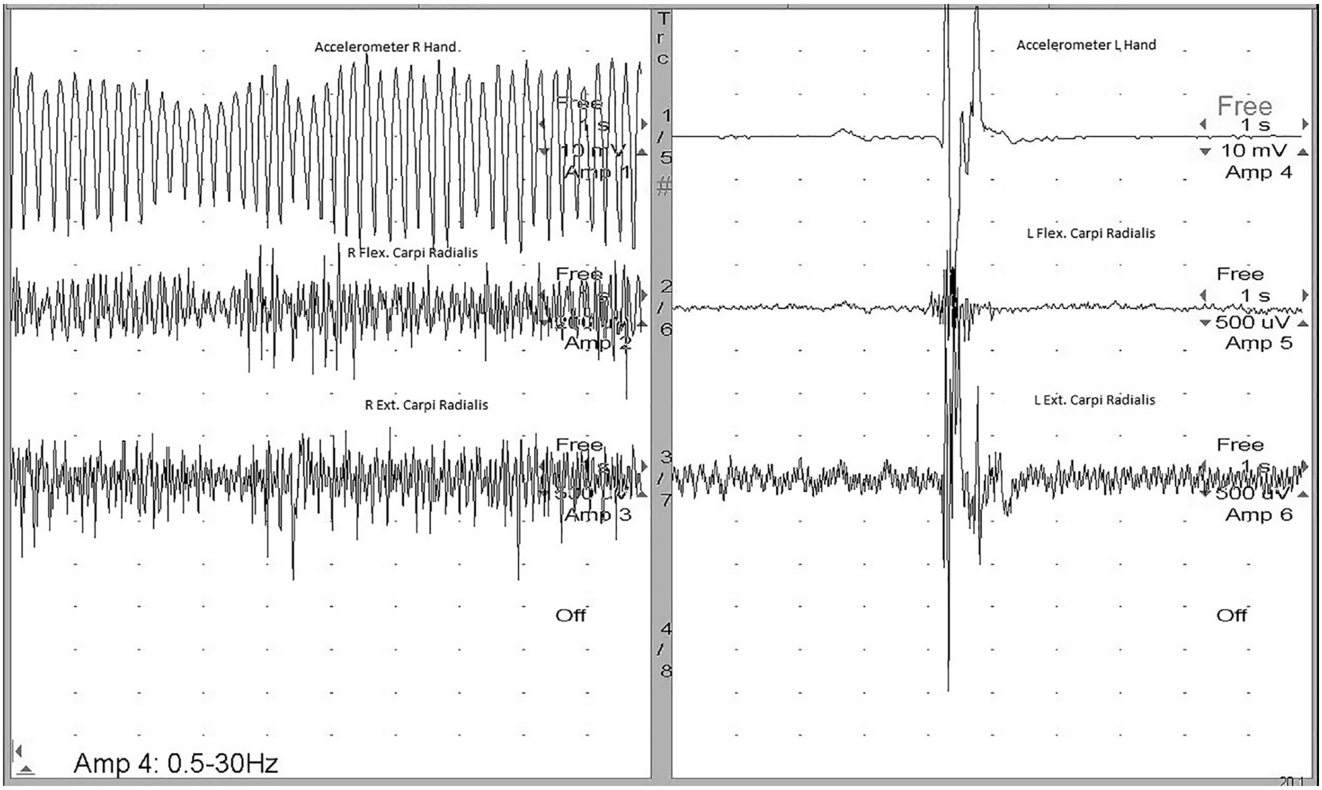

Fig. 6. Ballistic left wrist movement with continuous tremor recording from the right hand showing no tremor interruption or significant change in tremor amplitude.

mirroring. The co-existence of an ''entrained frequency'' with the central tremor component noted in the physiology study shows that the underlying tremor is likely not being entrained. However, it remains to be determined if what we note physiologically as entrained tremor can be clearly distinguished from mirror movements either clinically or physiologically, based on the findings noted on tremor analysis.

The etiology of the central component in the current case is not clear and could be multifactorial which may include essential tremor, tremor due to her lower motor neuron disease (polio) or drug induced considering her history of neuropsychiatric drug use. The lower frequency could likely be explained by her lower motor neuron disease and age (Elble, 2003; Gawronska, 1969; Hanajima et al., 2009). The minipolymyoclonus of the right hand is a residual of the polio and reflects an unfused tetanus of large motor units (Spiro, 1970). This movement is clearly separate from the tremors under consideration here due to its irregular nature.

The current case highlights the utility of objective physiology in identifying the nature of a tremor when functional tremor is suspected, but not completely clear on physical examination. The diagnosis of functional tremor can be biased in cases with psychiatric comorbidities and the severity of the functional tremor can vary based on the clinical setting, which may further mask the underlying organic tremors. Mirror movements may be mistaken for tremor entrainment which is one of the core clinical and physiologic criteria for the diagnosis of functional movement disorders (Schwingenschuh et al., 2011, 2016; Fahn and Williams, 1988; Shill and Gerber, 2006). Using low amplitude movements for tremor entrainment during clinical and physiologic testing may help reduce the likelihood of motor overflow. Additionally, EMG coherence should be used for evaluation of tremor entrainment since the accelerometers may erroneously report volume conduction. Organic tremors which can be potentially treatable, may be a major source of disability and perhaps an underrecognized comorbidity in diagnosed functional tremor patients.

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7. Conflict of interest

None.

Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.cnp.2018.05.001.

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