

Total recovery of a patient with psychogenic parkinsonism and the diagnostic utility of contralateral pronation-supination task during EMG recording

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Abstract

Psychogenic movement disorders are commonly seen in neurology clinics which can lead to severe disability. Despite their high prevalence, they constitute a grey area between neurology and psychiatry while neither of the specialists tends to take charge. Psychogenic parkinsonism is an uncommon subtype among psychogenic movement disorders. We report here a man with psychogenic parkinsonism that recovered completely after two years of treatment, showing that an excellent prognosis is possible with appropriate evaluation, diagnosis and treatment. We also discuss the utility of electrophysiological study in differentiating psychogenic tremors from other subtypes and the usefulness of the pronation-supination task.

Keywords: Psychogenic Parkinson' disease, EMG, tremogram, diagnosis

INTRODUCTION

Psychogenic movement disorders include a group of neurological symptoms that cannot be explained by any organic disorder and generally have an underlying psychological or psychiatric basis.¹ Psychogenic parkinsonism (PP) is an uncommon subtype among psychogenic movement disorder and generally manifests as like true parkinsonism.² Clinical features such as fluctuations of tremor, abrupt onset, unresponsiveness to drugs, and absence of accompanying signs of organic neurological disorders may be crucial features in the diagnosis of PP.³ However, distinguishing PP from organic parkinsonism may be a challenging task. Currently, the neurophysiological study of tremor has been emphasized as a crucial paraclinical aid to diagnosis.⁴ Nevertheless, there are still some debates about the utility of tremor analyses in distinguishing psychogenic tremor from organic tremor subtypes. For instance, in a crucial report by Schwingenschuh *et al.*, no single test with adequate sensitivity and specificity for separating the groups of psychogenic tremor and organic tremor was found.⁵ We present here a case of PP where the pronation-supination task was helpful in the differential diagnosis of tremor during EMG recordings. We also draw attention to the dramatic improvement of PP by long-term

psychiatric treatment, thereby emphasizing the importance of the appropriate diagnosis of these patients.

CASE REPORT

A 30-year old man was admitted to our movement disorder clinic due to gait difficulty and tremor. The patient initially had right-sided tremor which was started one year earlier which gradually progressed. There was also progressive slowness of the movements and contractions of lower extremity muscles with rapid deterioration in the last one-month. Five months after the onset of symptoms, the diagnosis of Parkinson's disease (PD) was made in a different center, and ropinirole 3mg and rasagiline 1mg were initiated. However, he continued to deteriorate gradually despite medical therapy and treatments was switched to levodopa/carbidopa/entacapone 5x150/37.5/200 mg daily before admission to our center. Of note, due to the early onset of parkinsonian signs and atypical course, work-up to exclude Wilson disease (serum and urinary copper, ceruloplasmin) neuroacanthocytosis syndromes (peripheral blood smear) were also performed in another center which yielded negative results.

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Date of Submission: 29 June 2020; Date of Acceptance: 29 August 2020

On neurological examination at admission to our clinic, a striking coarse, large-amplitude, 6-7 Hz tremor of the right hand was seen, which was worse on maintenance of posture and at rest. Besides, he had bilateral rigidity which was worse on the right side. However, the rigidity was not at a rhythmic frequency that correspond to the cogwheel sign. Slowness and difficulty of lower extremity movements were also observed. On the other hand, muscle strength evaluations could not be performed due to the excessively increased muscle tone of his lower extremity. He was only able to walk with the aid of a cane held with the right hand exhibiting an usual pattern of gait. His family history was unremarkable and there was no consanguinity of marriage between his parents.

The neurological examination and clinical course of the patient which had developed abruptly were atypical for PD. Additionally, there was no response to the levodopa treatment. At this point, the preliminary diagnosis of psychogenic parkinsonism (PP) or a comorbid psychogenic movement disorder accompanying PD was suspected.

Further studies with neurophysiologic tremor analysis were performed. He was instructed to tap rhythmically at an imposed frequency of 2 Hz, 3 Hz, and 5 Hz while performing the polygraphic recording of the left hand (abductor digiti minimi) during which there was no recorded interruption of the tremor of the left hand. After that, tremor recording was performed during the cognitive tasks of counting backward from 100 by serial 7's which also did not lead to a significant interference. Finally, he was instructed to perform pronation-supination movement of the right hand, and during this maneuver decrease of tremor amplitude and interruptions of tremor (distractibility) as well as mild shifting of tremor frequency to the rhythm of contralateral hand pronation-supination (entrainment) was recorded (Figure 1).

Taken together, the diagnosis of PP was established and he was referred to the psychiatry department for consultation. The psychiatric evaluation suggested that the symptomatology could be a manifestation of conversion disorder. There was also depressive symptoms. Sertraline was started and he underwent a cognitive behavioral therapy program. On the other hand, medications for PD were tapered off over a two week period which did not lead to any deterioration. At two years of follow up, his depressive symptoms had improved, and

neurological symptoms recovered completely such that he could maintain a full-time employment as a worker in a textile factory over the last three months.

DISCUSSION

PP which constitutes about 10% of all psychogenic movement disorders is considered to be a complex diagnosis requiring a very detailed clinical examination by a movement disorder specialist and confirmation with paraclinical methods.^{6,7} Most patients with PP manifest with tremors rather than other parkinsonian symptoms⁸ and the characteristics of the tremor is similar to that of other psychogenic tremor.⁸ Currently, EMG recordings of tremor during specific maneuvers such as tapping, pronation-supination of contralateral hand or cognitive distraction tasks can be useful to differentiate psychogenic tremor (PT) from organic tremor subtypes.^{9,10} In a study, executing tapping maneuvers as a distraction task was found to be useful in the differentiation of PT from other tremor subtypes.¹¹ However, in another report, no difference among types of distraction tasks was found, and the authors found that a combination of electrophysiological tests was able to distinguish psychogenic and organic tremors with excellent sensitivity and specificity.⁵ However, in the report, tasks including tapping and ballistic movements of the contralateral hand were performed, whereas the effect of pronation-supination task was not investigated.⁵ Another study by Kenney *et al.* found that the sensitivity of moderate-to-marked distractibility in distinguishing psychogenic and essential tremor was higher with alternate finger tapping, while specificity was slightly higher with serial 7 s. However, the utility of pronation-supination task was not evaluated in the study.¹²

The remarkable feature of our patient was that the cognitive distraction tests and simultaneous contralateral tapping tests during electrophysiological tremor analyses were nonsignificant, however, during contralateral pronation-supination task, dramatic distractibility and entrainment in ipsilateral tremor were recorded in simultaneous EMG recording which led to the final diagnosis of PT. As far as we are aware, in differentiating PT from organic tremor, a systematic study comparing the utility of the various distraction tasks including pronation-supination task has not been conducted. However, the usefulness of this task in the diagnosis of PT as shown in our patient will need to be confirmed in future large-scale studies.

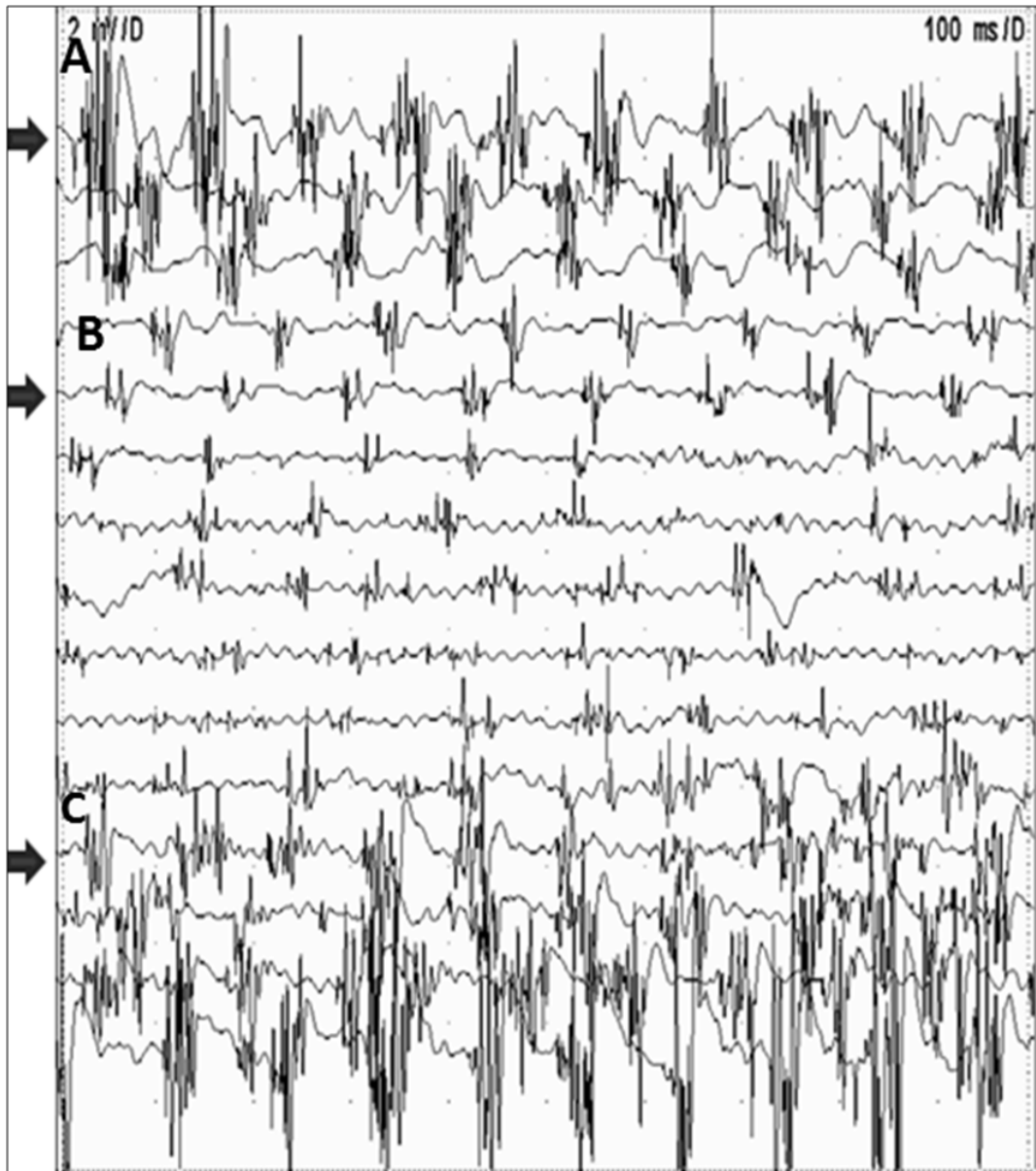


Figure 1. A, Polygraphic recording of the left hand (abd digiti min) during rest. B, Rhythmic pronation-supination of the right hand is instructed. Note the decrease of tremor amplitude and interruptions of tremor (distractibility) as well as mild shifting of tremor frequency to the rhythm of pronation-supination of the contralateral hand (entrainment). C, Recording again during rest

In conclusion, we think that this case represents a rare example of the dramatic recovery of a patient with PP after psychiatric treatments. In our opinion, this report may illustrate the importance of the accurate diagnosis of these patients with a life-changing improvement by appropriate treatment. Besides, during tremor analyses, we emphasize the importance of applying full distraction tasks including pronation-supination maneuvers for achieving optimal diagnostic

accuracy. We think that the differentiating effect of pronation-supination task from other tasks may be because it is a rather more complex task requiring the processing of more extensive supraspinal control in comparison to tapping and cognitive tasks. Keeping in mind the insufficiency of literature in this regard, we hypothesize that this task may be a practical method to be used during tremor in movement disorders clinics.

DISCLOSURE

Financial support: None

Conflict of interest: None.

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