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Tardive Dystonia or Tardive Myoclonus

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Classical tardive dyskinesia (TD) is a disorder with perioral (orobuccolinguo-masticatory) choreiform or stereotypical movements that occur as a late complication of long-term treatment with dopamine receptor blocking drugs (1). Variants of TD include tardive dystonia (TDt), tardive myoclonus (TM), tardive tremor, tardive tics, and tardive parkinsonism (1, 2).

The term TDt, introduced in 1973, is used to describe a movement disorder fulfilling the following criteria proposed by Dressler et al. (3): 1- presence of chronic dystonia, 2- history of antipsychotic drug treatment preceding or concurrent with the onset of dystonia, 3- exclusion of known causes of secondary dystonia by appropriate clinical and laboratory investigation, 4- no family history of dystonia. Other important clinical characteristics of TDt are that the dystonia is not aggravated by action, it often spares the lower extremities, retrocollis is a frequent feature, and it affects young males most frequently (3-5). TDt is often an incapacitating disorder, affecting 1.5-2% of all patients receiving antipsychotic medication (3).

However, myoclonus is rarely reported as a manifestation of tardive syndromes, and the presence of TM is still controversial.

Case Report

In August 1997, a 25-year-old man presented with involuntary, abrupt, brief, rapid jerks most prominent in the shoulders, but also appearing around the mouth, back and the arms. Involuntary movements never involved the lower extremities. They disappeared during sleep and they were not aggravated by action. He had a diagnosis of bipolar disorder and had used haloperidol, biperiden, and thioridazine in 1990, 1991, 1993, 1994, and 1996. He was also on clomipramine in 1990 and 1991; on pimozide in 1993 and 1994; on fluoxetine in 1993 and 1994; on lithium and chlorpromazine in 1995, 1996, and 1997; and on carbamazepine in 1997.

The onset of the involuntary movements was in the summer of 1996, first in the right and then in the left shoulder, spreading over the body parts mentioned above within a year. The patient had been severely disabled by the disorder since May 1997. At the time of the first consultation, he was on carbamazepine, 600 mg daily, for bipolar disorder, and diazepam, 15 mg daily, had been added to his regimen for the movement disorder three months previously. The neurological examination was normal except for the involuntary movements. The electromyographic (EMG) activity of the left trapezius, biceps brachii and extensor digitorum communis muscles exhibited bursts with durations of 800-1000 milliseconds

simultaneous with the involuntary movements (Figure). Although there were shorter bursts, which could be compatible with the duration of myoclonus, they were not associated with any movement. Cranial magnetic resonance imaging, serum biochemistry, blood count, thyroid tests, serum and urine copper and serum ceruloplasmin levels were all normal.



Figure 1. Electromyographic recordings of the involuntary movements.

L.: Left.

Ext. Digitorum Com.: Extensor digitorum communis.

The diagnosis of TDt was supported by the dramatic improvement of the movement disorder following IM injection of 5 mg biperiden. Consequently, the patient was put on 18 mg biperiden po, daily. This treatment ameliorated the dystonia, but the drug had to be discontinued because of a central anticholinergic

syndrome causing confusion and visual hallucinations. Then the patient was given tetrabenazine and the dose was titrated up to 200 mg daily until the resolution of symptoms without any side effects.

The confirmation of the diagnosis of TDt was made on the basis of the EMG pattern of the movement disorder as well as the dramatic improvement with an anticholinergic drug. The presence of bursts with a duration of 800-1000 milliseconds was compatible with dystonia but not myoclonus, where the duration of the bursts would be expected to be between 50 and 300 milliseconds (6). The duration of dystonic bursts is longer than that of myoclonic bursts (200 milliseconds and more) (7). Although TM is included among the tardive syndromes, we could find only two specific clinical reports of this movement disorder in the literature (8, 9). The fact that the patient reported by Tominaga et al. (9) had postural myoclonus makes the diagnosis of TM questionable, since tardive movement disorders characteristically decrease with posture or action (4). This report was also criticized by Little and Jankovic (8), authors of the second report of TM, who said that the movement disorder was probably not tardive, because it was not specifically noted whether the disorder was aggravated or at least remained the same following withdrawal of the typical dopamine receptor blocking agent. Therefore it seems unlikely that their patients had a tardive movement disorder.

Little and Jankovic (8), in discussing their patient, stated that "while the jerk-like movements resemble rapid dystonic movements, we believe that these movements represent myoclonus." We think that an EMG study could also be compatible with dystonia in their patient. The clinical differentiation between dystonia and myoclonus was difficult in our patient as well and only after the EMG could we exclude tardive myoclonus.

It is sometimes difficult to differentiate between dystonia and myoclonus clinically and with the criticisms outlined above it appears that the previously reported cases of TM is questionable. Cases giving the clinical impression of TM should be evaluated by EMG. This approach would put an end to the debate about whether such an entity as TM exists or not.

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