Matters arising

Nocturnal paroxysmal dystonia

Sir: Recently Lugaresi, et al\(^1\) described twelve patients with a remarkable and uniform clinical syndrome: nocturnal seizures characterised by stereotyped dystonic and/or ballistic movements which do not fit into any of the usual clinical categories of sleep-related motor attacks. In their cases the duration of the attacks varied between 15 and 50 seconds. Seizures recurred almost every night, sometimes several times per night, during slow wave sleep. The normal and interictal EEG recordings during both sleep and wakefulness prevent definite confirmation of the epileptic nature of the attacks. A particular form within the category of parasomnias, for example paroxysmal nocturnus, is also unlikely because of the high frequency and the short duration of the seizures.

Are we dealing with a new nosological entity whose pathophysiology is not yet understood?\(^2\)

At the time of this publication we were consulted by a 35-year-old woman referred to us with a nearly identical type of complaints.

About ten years ago the seizures abruptly occurred for the first time. Almost every night during sleep, usually between 4 and 5 am she shows one or more attacks of motor agitation. She suddenly sits upright in her bed, her head is postured in retroflexion and her eyes are fixed in a stare. Subsequently violent and disordered movements of limbs and trunk appear mostly accompanied by loud vocalisation. After about 40 seconds the attack subsides at once, the contractions disappear, the howling respiration is followed by apnoea and she lies motionless and deeply relaxed in bed. She is completely unresponsive to any form of communication in this episode that may take about 20 minutes. Afterwards she reports tiredness, nausea and aching muscles and is unable to give a detailed account. She appears exhausted and confused. Attacks never occur during the daytime. Repeatedly, neurological examination and laboratory investigations do not reveal any positive findings at all. EEG recordings during both sleep and wakefulness are normal. Treatment by medication, in particular hydantoin and barbiturates, has not any effect on the seizures.

Our clinical findings are in accordance with those described by Lugaresi et al.\(^1\) Contrary to them however we postulate that nocturnal paroxysmal dystonia might be an unusual type of conversion behaviour: symptoms as tactics in human relationships.\(^3\)\(^4\)

Family and personal history seem to be irrelevant in our case: so are the results of neuropsychologic testing and personality inventories, for example the MMPI, within the normal range. Other findings however are interesting from the psychological point of view: The onset of the seizures coincided with the beginning of her marriage: The attacks have far-reaching consequences for patients family life: the nights are without intimacy, but full of fear; the husband sleeps in another room to avoid being disturbed; the patient is restless and spiritless during the day; the couple is unable to have visitors or to sleep elsewhere because of the noise caused by the patient; the patient's behaviour is haughty and selfdefensive. If so asked she attempts to imitate the characteristic and efficient attitude of her husband.

The husband turns out to be a cold and cerebral man who describes himself in only rational terms. Nobody likes to have a nice chat with him. He knows that his wife feels inferior to him, but he as well thinks he is much more efficient than she is. “The last time, however, my wife showed some mental growth”. He reports very proudly that their marriage had meant “an escape” to his wife, who came from a socially isolated parental family. He sends his wife from one physician to another, “if need be to a mental hospital, for she must get rid of it”.

In view of these intake data we have no reason for the present to categorise the patient’s disturbance in a new nosological diagnosis. On the contrary: there are many indications that the nocturnal paroxysmal dystonia can be understood as functional behaviour, a way of dealing with the husband.

Maybe the syndrome gives the patient the advantage in gaining control of the conflictual relationship? With her nocturnal spasms she drives her self-compliant husband to despair. She does not conduct the struggle by open battle because she is sure to be a loser.

In order to test our hypothesis we instructed the patient to note down all the conflicts which will occur between her and her husband during the next four weeks. Especially she was asked to draw up an inventory of:

- what is the script of the conflict,
- who runs the show and who conforms,
- who is the first to compose the quarrel.

After this month her notes enable us to give the following answers:

- conflicts degenerate into slanging matches.

—the husband wins by means of his arguments and she climbs down,
—she takes the first step towards reconciliation.

She accomplishes this task with enthusiasm and her seizures stay away. In four psychotherapeutic sessions we teach the patient by using paradoxical and assertive instructions how to derange the stereotyped patterns of marital interaction.

After a lapse of two years she is ringing up to make an appointment. The nocturnal episodes have completely disappeared. Her behaviour has become much more open and assertive. She reports to be able now to oppose her husband in a direct way.

Is hypnogenic paroxysmal dystonia a new nosological entity? We consider the seizures of our patient to be an unusual example of an old diagnostic category in psycho-pathology: conversion behaviour. We do not think that the polysomnographic findings reported by Lugaresi et al\(^1\) give definite evidence against such an opinion.

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References


Lugaresi et al reply:
In the case of a 35 year old woman described by Bergen et al\(^1\) as affected with short lasting nocturnal paroxysmal dystonia\(^2\) several clinical features were not typical. We have never observed ictal non responsiveness for up to 20 min in our patients, who were usually able to give a fairly accurate account of the abnormal movements during the night. Moreover, we are not told whether carbamazepine was tried and found effective or whether polygraphic and audiovisual recordings of the attacks were obtained.

In over 20 years of sleep recordings we
Nocturnal paroxysmal dystonia.

H J Berger, T M Berendsen-Versteeg and E M Joosten

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