

JACTATIO CAPITIS NOCTURNA WITH PERSISTENCE IN ADULTHOOD

CASE REPORT

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ABSTRACT - Rhythmic movement disorder, also known as jactatio capitis nocturna, is an infancy and childhood sleep-related disorder characterized by repetitive movements occurring immediately prior to sleep onset and sustained into light sleep. We report a 19-year-old man with a history of headbanging and repetitive bodyrocking since infancy, occurring on a daily basis at sleep onset. He was born a premature baby but psychomotor milestones were unremarkable. Physical and neurological diagnostic workups were unremarkable. A hospital-based sleep study showed: total sleep time: 178 min; sleep efficiency index 35.8; sleep latency 65 min; REM latency 189 min. There were no respiratory events and head movements occurred at 4/min during wakefulness, stages 1 and 2 NREM sleep. No tonic or phasic electromyographic abnormalities were recorded during REM sleep. A clinical diagnosis of rhythmic movement disorder was performed on the basis of the clinical and sleep studies data. Clonazepam (0.5 mg/day) and midazolam (15 mg/day) yielded no clinical improvement. Imipramine (10 mg/day) produced good clinical outcome. In summary, we report a RMD case with atypical clinical and therapeutical features.

KEY WORDS: rhythmic movement disorder, parasomnia, jactatio capitis nocturna, headbanging.

Jactatio capitis nocturna com persistência na vida adulta: relato de caso

RESUMO - O distúrbio rítmico do movimento, também chamado jactatio capitis nocturna, é considerado um distúrbio do sono da infância e caracteriza-se por movimentos estereotipados que ocorrem no início ou no começo do sono. Relatamos o caso de um rapaz de 19 anos com história de movimentos rítmicos de cabeça e de balanceio do tronco desde lactente, sempre associados ao início do sono e de ocorrência diária. Tem antecedente de prematuridade mas com desenvolvimento neuropsicomotor normal. Seu exame físico geral e neurológico foram normais. O estudo polissonográfico hospitalar mostrou: tempo total de sono de 178 minutos, com eficiência de sono de 35,8%; latência de sono 65 minutos; e latência do sono REM 189 minutos. Não se observou apnéia e os movimentos de balanceio de cabeça ocorreram em torno de 4/minuto em vigília e nos estágios 1 e 2. Não foram observadas alterações eletromiográficas tônicas ou fásicas durante o sono REM. O paciente foi medicado com clonazepam (0,5 mg/dia) por 3 semanas sem melhora dos movimentos e a seguir recebeu midazolam (15 mg/dia) por 2 semanas, também sem resposta. Cloridrato de imipramina (10 mg/dia) produziu melhora significativa do quadro. Em suma, relatamos um caso de distúrbio rítmico do movimento com aspectos clínicos e terapêuticos atípicos.

PALAVRAS-CHAVE: distúrbio rítmico do movimento, jactatio capitis nocturna, parasomnia.

The first description of rhythmic movement disorder dates back to Wepfer in 1727. Several years later, Zappert (1905) reported on six children presenting headbanging during sleep and proposed

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the denomination *jactatio capitis nocturna*. During this century, several reports describing the various clinical features of headbanging became available. However, it was only in 1979 that a full description of a polysomnographic study of a patient revealing headbanging during sleep stages 1 and 2 was published¹.

Motor parasomnias are purposeful motor behavior occurring during REM, non-REM sleep or transitional states. The International Classification of the Sleep Disorders² tabulates them in arousal disorders, sleep-wake transition disorders, parasomnias associated with REM sleep and other parasomnias. Rhythmic movement disorder (RMD), also known as *jactatio capitis nocturna*, is a infancy and childhood sleep-wake transition parasomnia characterized by repetitive movements. These movements involve the head and neck, typically occurring at sleep onset and sustained into light sleep. Nearly 75 to 80% of the episodes occur at sleep onset; about 6% of patients have episodes during the night, and 10% have them both at bedtime and upon waking in the morning. The episodes usually last 15 minutes or less. It may be familial in some cases. RMD is classified as a sleep transition parasomnia². Headbanging usually occurs during pre-sleep drowsiness and early NREM sleep. RMD usually begin around 9 months of age and rarely persists after the four years of age. Persistence of these movements into older ages may be associated severe brain dysfunction as autism, mental retardation and other development abnormalities.

RMD does not usually require drug therapy. Nevertheless, there are literature reports of traumatic cataract and subdural hematoma associated with this disorder^{3,4}. Persistence in adulthood warrants drug therapy and the medication of choice is clonazepam whereas other benzodiazepines (BZD) and tricyclic antidepressive drugs (TCD) have been reported to produce a good therapeutical outcome⁵.

CASE REPORT

We report a 19-year-old young male with a history of nightly sleep onset headbanging and bodyrocking since infancy. He was born a premature baby but psychomotor development was unremarkable. The patient was a senior high school student at the time of the first consultation. There was no family history of mental and neurological disorder and his personal history was unremarkable. Physical and neurological diagnostic workups were also unremarkable.

The patient reported no daytime sleepiness. A hospital-based polysomnography showed: total sleep time 178 min; sleep efficiency index 35.8; sleep latency* : 65 min; REM latency* 189 min; stage 1 14.6%; stage 2 59.5%; stage 3 10.1%; stage 4 10.1%; REM sleep 5.6%. [*The relatively long sleep latency and REM latency can be attributed to a "first night effect"]. There were no respiratory abnormalities. Headbanging movements at 4/minute rate during wakefulness, NREM stages 1 and 2 were recorded. No tonic or phasic electromyographic abnormalities were recorded during REM sleep. There were no other parasomnic behaviors documented during NREM-REM sleep. A definitive diagnosis of RMD was performed on the basis of the clinical and sleep study data.

He was put on 0.5 mg/day of clonazepam for three weeks with no clinical improvement. Midazolam 15 mg/day for two weeks was initiated with no improvement thereafter. He finally was put on imipramine 10 mg/day, with an unequivocal subjective reduction of the rhythmic movements.

DISCUSSION

This case reported herein portrays an young male with persistence of RMD into adulthood and with a BZD-failure and a tricyclic antidepressant agent response.

RMD is sleep-wake transition parasomnia with usually a good prognosis not requiring drug treatment^{1,6}. The medication of choice is clonazepam with the best outcome whereas other short-acting BZD produce variable responses^{5,7}. There are several reports of RMD persisting into adulthood where BZDs were employed. Chisholm and Morehouse⁸ have described two patients with persistence of RMD into young adulthood (ages 19 and 24 years). Both have obtained good response to clonazepam. Walsh et al.⁷ have used oxazepam (10-20 mg) in a 8 year-old girl with a good outcome.

TCDs stand as a second-choice drug with good therapeutical outcome in cases where there is an initial failure to BZDs. This failure is associated with brain lesion or with persistence of clinical manifestations into adulthood. It might be that the some cases with persistence of clinical manifestations failing BZDs have an undocumented underlying brain injury which makes the clinical manifestations less responsive to BZDs. Our patient had no clinical features of brain injury. No signs of cognitive impairment were made evident despite a premature delivery. Yet, there are no clinical reports available in the literature showing that TCDs produce a good outcome as a first choice drug.

Drake reported a case of RMD responsive to imipramine, similarly as the case reported herein with no signs of brain injury⁹.

A differential diagnosis with REM sleep behavior disorder (RBD) is warranted. RBD usually begin after the 40th decade, and the movements occur exclusively during REM sleep. It consists of often injurious dream-enactment motor activity associated with vivid dreaming. Chronic RBD is most common in elderly men and in some cases there is a familial predisposition. Sleep studies often disclose atypical REM sleep with intermittent loss of the typical REM atonia. Recently, a subgroup of RBD patients with NREM parasomnias (sleepwalking and sleep terrors) was described and named as parasomnia overlap syndrome¹⁰. The patient reported here showed no clinical features compatible with RBD.

Other sleep-related movement-related disorders, as nocturnal frontal lobe originating from supplementar motor area, cingule girus and orbito-frontal or dorsolateral areas the frontal lobe epilepsy may produce motor activity during sleep. There are two clinical forms¹¹: one sporadic and other which is autosomal dominant, both beginning during adolescence. Seizures activity usually includes prominent motor and behavioral manifestations arising from NREM sleep. An unremarkable surface EEG have ruled out these conditions in these patients.

In summary, we report a RMD with atypical clinical and therapeutical features.

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