



IMAGES IN PAEDIATRICS

Epileptic seizure or parasomnia? From knowledge to recognition. Sleep-related hypermotor epilepsy[☆]



¿Crisis epiléptica o parasomnia? Del conocimiento a la identificación. Epilepsia hipermotora asociada al sueño

Ana Cristóvão Ferreira^{a,*}, Joana Moniz Dionísio^b, Rosário Ferreira^{c,d}, Joana Coelho^e

^a *Servicio de Pediatría, Hospital de Santa Maria, Centro Hospitalar Universitário Lisboa Norte, Lisboa, Portugal*

^b *Servicio de Neurología, Hospital Professor Doutor Fernando Fonseca, Lisboa, Portugal*

^c *Unidad de Pulmonología Pediátrica, Servicio de Pediatría, Hospital Santa Maria, Centro Hospitalar Universitário Lisboa Norte, Lisboa, Portugal*

^d *Facultad de Medicina, Universidade de Lisboa, Lisboa, Portugal*

^e *Unidad de Neuropediatría, Servicio de Pediatría, Hospital Santa Maria, Centro Hospitalar Universitário Lisboa Norte, Lisboa, Portugal*

Available online 12 April 2024

A male patient aged 16 years with no relevant personal or family history presented with nocturnal paroxysmal events (NPEs) characterized by stereotyped hyperkinetic movements, eye opening and production of guttural sounds during sleep, with onset at age 7 years. Due to the increasing frequency of NPEs, he was referred to the sleep laboratory for assessment of suspected parasomnia. An event compatible with an epileptic seizure (stereotyped hyperkinetic movements associated with epileptiform discharges on the electroencephalogram [EEG]) was detected during a video-EEG polysomnography. The findings of the routine electroencephalogram and head MRI were normal. The next-generation sequencing panel for sleep-related hypermotor

epilepsy (SHE) was negative. The NPEs resolved after initiation of treatment with carbamazepine.

Sleep-related hypermotor epilepsy is a rare disease that is more frequent in males and usually with onset in childhood or adolescence.^{1,2} The seizures are brief, hyperkinetic, with possible tonic/dystonic movements, and associated with sleep, occurring less frequently during daytime.^{1,3} Their frequency can range from 1 to 20 episodes per night, affecting performance in daily activities.¹

The diagnosis is based on the anamnesis and the clinical features, since the EEG features are often normal or obscured by muscle artifacts and the findings of neuroimaging may be normal, too, the family history is often negative, and a genetic aetiology is rarely found in sporadic cases.¹ It is frequently misdiagnosed as a non-rapid eye movement parasomnia due to the high prevalence of these parasomnias in childhood.² A sleep video recording is essential for correct diagnosis, which has an impact on the patient's quality of life.

DOI of original article: <https://doi.org/10.1016/j.anpedi.2023.12.013>

[☆] **Previous meeting:** This clinical case was presented as a poster at the XXVII Jornadas de Pediatría, Hospital de Santa Maria, February 24–25, 2022, Lisbon, Portugal.

* Corresponding author.

E-mail address: ana.r.c.ferreira@chln.min-saude.pt
(A. Cristóvão Ferreira).

Description of the video

The video recording obtained during polysomnography showed the patient having one of his typical events on the N3 stage of sleep, characterized by stereotyped, hyperkinetic movements, with eye opening and guttural sounds. We observed epileptiform discharges originating in the right centro-frontal region that propagated to the left frontal region.

Funding

There were no external funding sources for the development of this paper.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.anpedi.2023.12.013>.

References

1. Menghi V, Bisulli F, Tinuper P, Nobili L. Sleep-related hypermotor epilepsy: prevalence, impact and management strategies. *Nat Sci Sleep*. 2018;10:317–26.
2. Tinuper P, Bisulli F, Cross JH, Hesdorffer D, Kahane P, Nobili L, et al. Definition and diagnostic criteria of sleep-related hypermotor epilepsy. *Neurology*. 2016;86:1834–42.
3. Licchetta L, Vignatelli L, Zenesini C, Mostacci B, Ferri L, Provini F, et al. Sleep-related hypermotor epilepsy: a prediction cohort study on sleep/wake patterns of seizures. *Epilepsia*. 2019;00:1–6.