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DIAGNOSTIC APPROACH TO TRANSITORY PEDIATRIC HYPERSOMNOLENCE. CASE REPORT

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Palabras clave: Sueño; Somnolencia; Trastornos intrínsecos del sueño; Trastornos de somnolencia excesiva; Diagnóstico Clínico; Pediatría.

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ABSTRACT

Introduction: Hypersomnolence is a common complaint from parents, caregivers, and patients themselves in pediatric clinical consultations, but it is often underestimated by clinicians. This article aims to review the diagnostic approach to pediatric hypersomnolence and to highlight the gap between knowledge and clinical practice regarding sleep disorders.

Case presentation: An 8-year-old boy presented with an episode of excessive sleepiness that prevented him from attending school and carrying out his daily activities. Anamnesis and family and personal history did not account for the symptoms, and physical and mental examinations showed no significant findings. Evaluation by specialists and results from paraclinical studies performed also did not yield significant findings to identify the cause of these symptoms. Finally, immunological tests showed positive levels for herpes simplex virus type 2 (HSV-2) and Epstein-Barr virus (EBV), suggesting that a resolved viral infection was the most likely cause of the transitory hypersomnolence.

Conclusions: Clinicians should be equipped with the knowledge and tools necessary to properly assess and diagnose hypersomnolence in children and adolescents, as it is a common symptom with potentially serious repercussions on patients and their family environment.

RESUMEN

Introducción. La hipersomnolencia es una queja reiterada por parte de padres, cuidadores y pacientes en las consultas clínicas de población pediátrica; sin embargo, con frecuencia es subvalorada por el personal clínico. El presente artículo tiene como objetivos hacer una revisión del abordaje diagnóstico de la hipersomnolencia pediátrica y evidenciar la brecha que existe entre el conocimiento y la práctica clínica con respecto a los trastornos del sueño.

Presentación del caso. Niño de 8 años quien presentó un episodio de somnolencia excesiva durante el cual dejó de asistir a la escuela y realizar sus actividades cotidianas. La anamnesis y los antecedentes familiares no explicaban la presencia de los síntomas y la exploración física y mental no mostró hallazgos significativos. La evaluación por especialistas y los resultados de los estudios paraclínicos realizados tampoco arrojaron hallazgos significativos para identificar la causa de la somnolencia. Los estudios inmunológicos mostraron niveles positivos para el virus del herpes simple tipo 2 y el virus de Epstein-Barr, lo cual finalmente indicó que una infección viral resuelta era la causa más probable de la hipersomnolencia transitoria.

Conclusiones. Es importante que el personal clínico cuente con los conocimientos y herramientas necesarios para evaluar y diagnosticar adecuadamente la hipersomnolencia en niños y adolescentes, pues es un síntoma frecuente que puede tener repercusiones graves en los pacientes y en su ambiente familiar.

INTRODUCTION

Hypersomnolence or excessive daytime sleepiness is a symptom described as the "inability to remain awake and alert during major waking episodes, resulting in periods of irrepressible need for sleep or unintentional lapses into drowsiness or sleep" (1). However, it may be difficult to determine when daytime sleepiness is excessive enough to be considered a symptom requiring attention and/or treatment. In the absence of specific criteria for children and adolescents, in practical terms, daytime sleepiness becomes problematic when it interferes with school, social, and/or family functioning (2). Hypersomnia, on the other hand, is a different concept, as it specifically refers to a group of disorders whose primary symptom is hypersomnolence and includes conditions such as narcolepsy, idiopathic hypersomnia, and Kleine-Levin Syndrome (recurrent hypersomnia) (1).

Daytime hypersomnolence is a relatively common complaint from parents/caregivers of children and adolescents. According to a survey conducted by the National Sleep Foundation in the United States (3), two out of ten parents (20%) reported that their children under the age of 10 seemed sleepy or tired during the day. Among adolescents, a similar survey conducted by the same foundation indicated that more than a half of the teens surveyed (4), aged 11 to 17, claimed to have had similar symptoms.

The consequences of pediatric hypersomnolence are varied. Excessive daytime sleepiness in school-aged children is associated with a higher risk of poor academic performance and cognitive difficulties, behavioral disorders, peer relationship problems, emotional issues, and depressive symptoms (2,5,6). It has also been reported that 45% of teenagers between the ages of 11 and 17 have experienced at least one problem related to daytime sleepiness, such as being late or missing school due to not waking up on time, falling asleep at school, feeling too tired to exercise or perform any physical activity, feeling too tired or sleepy during the day, and/or falling asleep while studying or doing homework (4).

The negative effects of hypersomnolence are also evident at home, since having a child with excessive daytime sleepiness in the family environment has a negative impact on the parents' quality of life. In a study carried out in children aged 2 to 12 years, a significant correlation was found between daytime sleepiness in children and daytime sleepiness among their parents, especially their mothers (7). Moreover, parents whose children have excessive daytime sleepiness tend to sleep fewer hours than parents whose children do not (3).

In Colombia, although some research has been carried out on the prevalence of sleep disorders in children (8), no studies on hypersomnolence or hypersomnia in the pediatric population have been reported. As a result, the personnel in charge of providing healthcare services to children and adolescents must be knowledgeable about how to clinically approach hypersomnolence and be able to make an accurate and timely diagnosis to establish a treatment plan. With this

purpose in mind, this clinical case will serve as a guide through the diagnostic approach to hypersomnolence in the pediatric population.

CASE PRESENTATION

An 8-year-old boy from Calarcá, Quindío, Colombia, residing in Montenegro, Quindío, Colombia, and studying in the fourth grade, was taken to an outpatient secondary care institution for a child and adolescent psychiatry medical consultation. He and his family did not identify with any particular ethnicity. His mother reported an episode of excessive daytime sleepiness that began 15 days earlier, and that these symptoms had been present for two weeks but had subsided spontaneously over the two days preceding the consultation.

According to the mother, the child began to present daytime drowsiness, which intensified to a point where "he slept all day and night". During this time, the child stopped attending school and carrying out his daily activities, and also exhibited emotional and behavioral disturbances, such as emotional instability, hostility, defiance, and impulsivity. Other symptoms, such as alterations in his nighttime sleep routine, were not reported, and behaviors such as hyperphagia and sexualized behavior were ruled out during the clinical interview. The child had age-appropriate sleep patterns, falling asleep at 8:00 pm and waking up at 6:30 am, for a total of 10.5 hours of sleep. Some days, he woke up a bit later, at 09:00 am, for a total of 13 hours of sleep, and he was not used to taking naps during the day.

No personal history that could explain the symptoms described was found in the anamnesis. The child had no history of pharmacological treatment, and there were no signs that suggested substance use. Family medical history included reports of hypothyroidism (maternal grandaunt), a suicide attempt (second maternal cousin), and drug dependence (maternal granduncles). Physical examination showed no relevant findings. Mental examination revealed an oppositional and uncooperative attitude and mutism, which limited the extent of the evaluation. A review of his medical records revealed that he had been referred to a child psychiatrist due to the presence of hypersomnolence and behavioral symptoms, leading to a diagnostic suspicion of Kleine–Levin syndrome.

Based on the information available and the clinical findings made up to this point, an initial diagnosis of transitory hypersomnolence was made. Even though emotional and behavioral symptoms were reported, their sudden onset related to the sleep disturbances described was not compatible with the natural course of an affective disorder. Therefore, treatment initially focused on providing sleep hygiene recommendations and tracking sleeping habits. Psychotherapy was prescribed to treat the behavioral and affective symptoms described. No specific treatment for hypersomnolence was administered.

At follow-up visits over the next few months, the mother reported that there had been no similar episodes of hypersomnolence after the first episode, with occasional episodes of hypersomnolence lasting less than a day, and that the behavioral and affective symptoms subsided along with the excessive sleepiness. She also reported sporadic febrile episodes, which resolved with the administration of paracetamol and warm baths, and also that when the child received medical care for such episodes at other health centers, laboratory tests failed to identify a specific cause.

During the six months of diagnostic testing, the child was evaluated by specialists in different areas: Pediatrics, Child and Adolescent Psychiatry, Pediatric Neurology, Infectious Diseases, and Genetics. Since the first consultation, several paraclinical studies were requested and performed, including a polysomnogram (PSG), which showed results within normal parameters for his age; thyroid function, also showing normal values; an electroencephalogram (EEG) that showed no evidence of abnormal brain activity; and an immunological study with positive results for antibodies against herpes simplex virus type 2 and Epstein Barr Virus (Table 1), leading to propose this resolved viral infection as the most likely cause of transitory hypersomnolence. The multiple sleep latencies test (MSLT) was also requested during the first consultation, but it was never performed due to insurance issues and administrative delays.

Table 1. Immunology test results

Test	Results	Reference value
Antistreptolysine O	<200	<200
HSV-2 IgG	4.11	Up to 1
EBV VCA IgM	16.9	Up to 20
EBV VCA IgG	107.9	Up to 25
EBNA IgG	>800	Up to 25
EBNA IgM	<1/20	<1/20
HIV-1 and HIV-2 antibodies	Non-reactive	- ¹
HBsAg	Negative	-

HSV-2: Herpes Simplex Virus 2; IgG: Immunoglobulin G; EBV VCA: Epstein Barr Virus - Virus Capside Antigen; IgM: Immunoglobulin M; EBNA: Epstein Barr Nuclear Antigen; HIV: Human Immunodeficiency Virus; HBsAg: Hepatitis B Surface Antigen. Source: Own elaboration.

DISCUSSION

The diagnostic approach to hypersomnolence, as well as to any other symptom or clinical entity, begins with a good clinical history. In the case of hypersomnolence, the author's recommendation is to focus on the following aspects, which will allow the clinician to elucidate the possible cause, or causes, of this symptom:

- 1. Characteristics of hypersomnolence: Is it a persistent or episodic condition? If episodic, are the episodes recurrent?
- 2. Circumstances in which the hypersomnolence occurs: Does it occur under certain circumstances, such as in the middle of a conversation? Does it occur even after the child has slept for an adequate number of hours the night before?
- 3. Impact on overall functioning: Is excessive daytime sleepiness severe enough to have a negative impact on the individual's social, school, or family functioning?
- 4. Sleep schedules: Is the person getting enough sleep at night, considering his/her age and stage of development? What are his/her sleep patterns? Is his/her sleep uninterrupted? Does he/she take naps during the day?
- 5. Presence of associated symptoms: Are there coexisting symptoms associated with the onset of hypersomnolence? Have there been behavioral changes such as hyperphagia or hypersexuality? Have there been episodes of cataplexy or hallucinations?
- 6. Presence of risk factors: Is the child obese? Are there respiratory symptoms such as snoring, apnea, or mouth breathing?
- 7. Presence of psychiatric symptoms: Are depressive symptoms suspected? Are there reports of behavioral problems such as refusal to go to sleep at bedtime?
- 8. Presence of parasomnias: Are there nightmares that cause repeated awakenings?
- 9. Personal history: Is there any coexisting disease that may be associated with excessive daytime sleepiness or other sleep disorders?
- 10. Pharmacological and toxicological history: Does the patient consume any medication or psychoactive substance that may cause drowsiness during the day?

The use of clinical instruments such as sleep diaries, or the administration of scales and questionnaires, may be useful to obtain complementary information. These tools will provide data about the environment in which the individual sleeps, their rest periods, sleep behavior, sleep patterns, circadian rhythms, emotional state, school performance, and drowsiness (9). Many instruments are available worldwide, but, in Colombia, two have been validated to assess drowsiness in the pediatric population, namely, the Pediatric Drowsiness Scale (EPS-VC, as per its Spanish acronym in the Colombian version) and the Cleveland Adolescent Sleepiness Questionnaire (CCSA-VC, as per its Spanish acronym in the Colombian version) (10). Medical personnel should then use the information gathered through these instruments to establish the possible cause(s) of the child's hypersomnolence.

Kothare *et al.* (11) divides the causes of hypersomnolence into three categories: those related to insufficient sleep, those related to fragmented sleep, and those related to an increased need for sleep (Table 2). Once the possible causes have been established, the relevant paraclinical studies should be carried out, and the intervention of other specialists or other disciplines, for example, psychology, will be determined.

Table 2. Causes of pediatric hypersomnolence

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Insufficient sleep	Fragmented sleep	Increased need for sleep
Insomnia Insufficient sleep syndrome Circadian-rhythm disorders Sleep movement disorders (RLS)	Behavioral insomnia Sleep breathing disorders Sleep movement disorders (PLMD) Parasomnia (e.g., night- mares) Medical conditions (e.g., asthma, rhinitis) Environmental causes (e.g., excessive noise)	Transitory hypersomnolence Medical conditions (e.g., metabolic disorders) Neurological diseases (e.g., epilepsy) Side effects of medications Recurrent hypersomnolence (e.g., KLS) Persistent hypersomnia (e.g., narco- lepsy and idiopathic hypersomnia)

RLS: restless leg syndrome; PLMD: periodic limb movement disorder; KLS: Kleine-Levin syndrome.

Source: Own elaboration based on Kothare et al. (11).

In this case, in view of the presence of febrile episodes of unknown etiology, it was considered that an evaluation by the Infectious Diseases Department was required. Their findings suggested that an infection had occurred at least 6 months prior to these tests, as registered by the infectious diseases specialist in the medical chart. It should be noted that, due to the procedures for the scheduling appointments with the specialist and conducting the aforementioned tests, more than 6 months had already elapsed since the onset of the symptoms.

On the other hand, his family history of hypothyroidism, added to the described symptomatology, made it necessary to perform thyroid function tests, which were found to be within normal values. Likewise, due to the relationship between epilepsy and excessive daytime sleepiness (12), an EEG was requested, which was also within normal parameters.

Furthermore, PSG with oximetry allows to objectively obtain sleep data such as: total sleep time; sleep efficiency; sleep latency; rapid eye movement (REM) sleep latency; arousal index; apnea-hypopnea index (AHI); periodic limb movement index; arousal index related to periodic limb movement; minimum, maximum, and average oximetry; end-tidal PCO2; and abnormalities in EEG, electrocardiogram, and non-REM and REM sleep stages (13). The relevance of PSG should be assessed by the medical personnel considering the respiratory and non-respiratory indications for its performance (14–15). In this case, the PSG report was within normal parameters for the child's age.

The multiple sleep latency test (MSLT) is a study that allows to objectify the hypersomnolence complaint, as it measures the tendency to fall asleep (11). MSLT is recommended for patients with suspected narcolepsy, idiopathic hypersomnia, or other hypersomnias (e.g., Kleine–Levin syndrome) (16–17), but not for the routine evaluation of sleepiness in medical or neurological disorders, insomnia, or circadian rhythm disorders (16). It may also be useful when medical history, self–report or parental report, and PSG are inconclusive in determining the

severity of hypersomnolence (2). It is recommended that the MSLT be preceded by a PSG from the previous night to rule out other sleep disorders and to ensure that there is no sleep deprivation (9).

In the case under study, an MSLT was requested because of an initial suspicion of Kleine–Levin syndrome, which is the same reason why a genetic assessment was requested. It should be noted that, in some settings, the logistics for performing a PSG followed by an MSLT may be challenging, as they are requested, authorized, and billed as separate studies, making it difficult to schedule them appropriately. As mentioned above, an MSLT was never performed, but it was hypothesized that since the symptom of hypersomnolence had disappeared at the time of the first consultation with the child and adolescent psychiatry service and no subsequent episodes were described, the MSLT would not have provided information relevant to the case study.

In view of the information obtained from the clinical history and paraclinical studies, the possible etiologies of hypersomnolence described in Table 2, and having ruled out possible medical and neurological causes, the probable causes of the symptoms described above are related to transitory hypersomnolence. In the pediatric population, transitory hypersomnolence may be secondary to viral infections and other medical illnesses, use of certain medications, and consumption of psychoactive substances (18). Since no evidence of the latter two were found in the exposed clinical case, it was established that the most probable cause of this condition was secondary to a viral infection. This diagnostic hypothesis is supported by the findings of the immunological study, in which the results suggested a resolved Epstein–Barr virus infection.

CONCLUSIONS

Sleep medicine is a growing field, but there are still gaps between the knowledge of sleep disorders in the pediatric population and the application of this knowledge in clinical practice. One of the strengths of this case report is that many clinicians were aware that a sleep disorder could be suspected, however, the progression seen in this case reflects the limitations encountered, including the delay in diagnosis, the difficulty in integrating all the clinical work done by each specialist, and the barriers imposed by the health care system.

Considering the frequency of complaints related to hypersomnolence in children and adolescents, as well as the severity of the impact it may have on the child and their family environment, clinicians must have the necessary knowledge and tools to assess this symptom and undertake an appropriate diagnostic approach. This article represents a modest effort to make this possible, but this effort must be multiplied and extended to undergraduate and postgraduate university medical training, as well as to continuing education programs.

ETHICAL CONSIDERATIONS

The mother's express consent was obtained for the preparation of this article, which was documented by filling out and signing an informed consent form for the publication of information contained in the medical records for academic purposes.

CONFLICT OF INTERESTS

None stated by the author.

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