



DIAGNOSTIC CHALLENGES OF HYPERSOMNIA IN AN INDIAN CHILD: A CASE REPORT

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KEYWORDS :

INTRODUCTION

Hypersomnia is a type of sleep disorder, characterized by an excessive quantity of sleep and excessive daytime sleepiness (EDS). The International Classification of Sleep Disorders 2nd edition (ICSD-2) defines EDS as “the inability to stay awake and alert during major waking episodes of the day, resulting into unintended lapses into drowsiness or sleep” [1]. Hypersomnia can contribute to poor general health, social outcomes and learning difficulties in children (2). Sleep disorders in this category include narcolepsy and its variants, idiopathic hypersomnia with and without long sleep time, behaviorally induced insufficient sleep syndrome, and recurrent hypersomnia typified by Klein-Levin syndrome. This category also includes hypersomnia due to substance abuse and medical conditions, such as obesity, obstructive sleep apnea, epilepsy, depression, head trauma and brain tumors (3). Since hypersomnia has a subjective interpretation, it could be mistaken as tiredness or fatigue, thus presenting a diagnostic challenge.

The normal sleep physiology changes with growing age. Children of 5–13 years require about 10 hours sleep per day (5). From a subjective point of view, structured questionnaires and scales that are validated in the general population are commonly used. The Epworth Sleepiness Scale (ESS) is the most common instrument used in sleep research [4]. It is a quick self-applied scale with 8 typical dozing situations with a 0 to 3 point score for each question. With a maximum of 24 points, scores >10 are considered abnormal for most authors (4).

The Case

XYZ, a 8 years old boy studying in 3rd standard brought by mother and uncle to a tertiary care hospital with complaints of excessive day time sleepiness and lethargy since last 3 months. He was apparently asymptomatic 3 months back when his mother started receiving complaints from his class teacher regarding frequently falling asleep in school. Gradually his mother noticed an increase in total sleep time. After sleeping for 9-10 hours at night he had great difficulty to wake up next morning. He was frequently falling asleep in school or in social gatherings. He even found it difficult to remain awake while eating food, taking a bath or using the toilet. He was spending 14-15 hours per day sleeping. The sleep was non refreshing and he always wanted to sleep more. Since last 3 months his scholastic performance has deteriorated and he also complained of poor attention and concentration.

The patient was a 2nd child out of a non-consanguineous marriage. Pregnancy and delivery period was uneventful. There was no history of any developmental delay, other physical complains, sleep related breathing complains, behavioral disturbance, substance abuse or any stressor. On specific asking mother reported of occasional jerky movements of both the upper limbs while sleeping at night for last 2 months. No history of similar complains in other two siblings. His mother had multiple nodules over face neck and hands looking like neurofibromatous nodules, for which she never sought any medical advice. Patient's 4 years old younger brother was a diagnosed case of cerebral palsy.

At presentation he was well kempt, well behaved, cheerful child appearing smaller than his age. His body mass index (BMI) was 17.6 kg/m². General physical examination and all the systemic examinations including central nervous system were within normal limits. The Epworth Sleepiness Scale (ESS) score was 19 (Table 1), with maximal scores in all sedentary parameters, indicating EDS. A

pediatric review was done to rule out organic causes of EDS, which was within normal limits. All routine blood investigations including CBC, LFT, RFT, RBS, serum ammonia, serum electrolytes, routine urine and stool examinations were within normal limits. MRI brain was within normal limits. His natural sleep EEG showed a symmetric, synchronous background activity of 7-8 Hz. Intermittent sharp and slow waves were recorded in bilateral centro-parietal region at many places. Due to lack of facilities for sleep studies in our institution and financial constraints of the patient's family, polysomnography and multiple sleep latency test (MSLT) were not done. The test for trypanosomiasis was not done as it was not endemic in the locality and there were no other clinical features suggesting towards the diagnosis. A Neuro physician reference was done and the diagnosis of myoclonic epilepsy with bilateral centro-parietal focus was made.

The patient was prescribed syrup Divalproex sodium 250 mg BD and was advised to maintain a sleep diary. On follow up after 15 days mother reported 20-30% improvement in symptoms. On follow up after 1 month he was sleeping 11-12 hours per day and his scholastic performance also improved. He could maintain his daily routines with much lesser struggle to remain awake.

Table 1: Patient's Epworth Sleepiness Scale (ESS)

Sr. no.	Activity	Situation Chance of Dozing (Score 0-3)*
1.	Sitting and reading	2
2.	Watching TV	2
3.	Sitting, inactive in a public place (e.g. a theatre or a meeting)	3
4.	As a passenger in a car for an hour without a break	3
5.	Lying down to rest in the afternoon when circumstances permit	3
6.	Sitting and talking to someone	1
7.	Sitting quietly after a lunch without alcohol	3
8.	In a car, while stopped for a few minutes in the traffic	2
	Total	19

*Scoring Scale: 0 = would never doze, 1 = slight chance of dozing, 2 = moderate chance of dozing, 3 = high chance of dozing

DISCUSSION

Relationship between epilepsy and sleep disorder are well studied. Epilepsy can promote sleep disruption and significantly affect both the quality, quantity, and the architecture of sleep (6). Patients with epilepsy generally have macro-structure sleep abnormalities such as increased number and duration of awakenings during sleep, reduced sleep efficiency, reduced or abnormal K complexes and sleep spindles, reduced and fragmented REM sleep, and increased stage shifts (7). Seizures may acutely disrupt the sleep-wake state, with the clinical consequence of nocturnal insomnia and EDS (8), (9). There are many causes of sleep disruption in epilepsy patients, including inadequate sleep hygiene, coexisting sleep disorders, and circadian rhythm disturbances (10). The end result of sleep disruption is EDS, worsening seizures, and poor quality of life.

Patients with epilepsy in general appear to have a greater prevalence of sleep disturbance than normal controls (6). In children with epilepsy

questionnaire-based studies reveal that they are more likely to have sleep complaints than the general pediatric population (11), (12). In Table 2, a summary of the studies addressing the relationship between epilepsy and EDS is displayed.

Table 2: Children-based Studies On Epilepsy and EDS.

Author/ place/ year	Study design	Population	Instruments	Findings on EDS
Larson et al. USA, 2012 (13)	Cross-sectional	105 households with a child with epilepsy 79 controls	E-Chess; CSHQ; PSQI; Iowa Fatigue Scale.	Children with epilepsy with more daytime sleepiness, parasomnias, bedtime resistance, and sleep onset delay.
Tang et al. UK, 2011 (14)	Cross-sectional	43 patients 494 controls	CSHQ	CHSQ was higher in the epilepsy group and parents reported shorter sleep duration, parasomnias, and increased daytime sleepiness in the rolandic epilepsy group.
Ong et al. Malaysia, 2010 (15)	Cross-sectional	92 cases; 92 healthy siblings as controls	Medical record; SDSC; EISS; ESSS-C.	Higher SDSC score in the epilepsy group in disorders of initiating and maintaining sleep, sleep-wake transition, sleep-disordered breathing, and EDS.
Maganti et al. USA, 2006 (16)	Cross-sectional	26 cases, 26 controls	Pediatric Sleep Questionnaire; Pediatric Daytime Sleepiness Scale.	Breathing was more reported in the epilepsy group. In PDSS, EDS scores were worse in the epilepsy group.
Becker et al. USA, 2003	Cross-sectional	14 cases, 14 controls	Pediatric Sleep Questionnaire; CPRS-R:L; ECBI; Children's Depression Inventory; Revised Child Manifest Anxiety Scale; PSG	More than 50% of children with epilepsy had behavioral problems. No difference in snoring, EDS, and restless sleep.

CSHQ: Children's sleep habits questionnaire; PSQI: Pittsburgh sleep quality index; E-Chess: early childhood epilepsy severity scale; PSG: polysomnography; EDS: excessive daytime sleepiness; SDSC: sleep disturbance scale for children; ECBI: Eyberg child behavior inventory; EISS: epilepsy illness severity score; ESSS-C: Epilepsy syndrome severity score-child.

Only a few Indian studies are available on childhood sleep disorder. A questionnaire based survey by Suri J.C., et al. was performed to determine the prevalence of sleep related disorders in Indian school-going children residing in Delhi. It was of great concern that the findings suggested that no effort was made on the part of parents to seek medical help in significantly large number of children with sleep disorders. This scenario indicates a total lack of awareness amongst the general population about the larger implications of sleep disorders in children (18).

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