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Diagnostic challenges and psychosocial impacts of hypersomnia in a Nigerian adolescent: A case report

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Abstract Hypersomnia, a disorder of sleep characterized by excessive quantity of sleep and excessive daytime sleepiness (EDS), could adversely affect health outcomes in childhood. To highlight diagnostic challenges and the impact of childhood hypersomnia in a resource limited setting, a 14 year old Nigerian who presented with a seven year history of hypersomnia was studied. Screening for sleep disorder using BEARS sleep algorithm and assessment for EDS using the Epworth Sleepiness Scale were both positive for EDS. The assessment of sleep hygiene using the Adolescent Sleep Hygiene Scale was normal and the use of Raven's Progressive Matrices to assess

intelligence was also normal. There was no contributory past medical history or demonstrable etiology. Magnetic Resonance Imaging (MRI) of the brain, Electro Encephalogram (EEG), assay of serum electrolytes were all normal while screening test for trypanosomiasis was negative. Poor level of awareness, high cost of evaluation and limited facilities for diagnosis of sleep disorder were the major diagnostic challenges. Depression, poor academic performance, suicidal ideation and stigmatization were all associated with hypersomnia in the patient.

Keywords: Hypersomnia, adolescence, depression, sleep disorders, diagnostic challenges

Introduction

Sleep disorders such as hypersomnias, parasomnias, and insomnias could impair development and result in poor health and social outcomes in childhood^{1,2}. Hypersomnias, characterized by an excessive quantity of sleep and excessive daytime sleepiness (EDS), is one of the major categories of sleep disorders identified by the International Classification of Sleep Disorders (ICSD-2)³⁻⁵. 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^{4,5}. Also in this category is hypersomnia due to substance abuse and medical conditions, such as obesity, obstructive sleep apnea, depression, head trauma and brain tumors^{4,5}.

The estimated prevalence rates for the hypersomnias include: 0.3% for idiopathic hypersomnia, 0.045% for narcolepsy and a range of 1-4% for the behaviorally induced insufficient sleep syndrome⁶. Studies on the

prevalence of sleep disorders in the Nigerian population are lacking⁷. However studies on EDS, the cardinal feature of hypersomnia, from Nigeria indicate a prevalence of 53% among children with neurological disorders, 44.8% among adults with depression and 17% in a school population⁷⁻⁹. An EDS prevalence rate of 15% has also been reported from a school population in the United States while worldwide estimates put the prevalence of EDS as ranging four to 30% in adult populations^{3,6}.

In adolescence the recommended sleep duration is 8.5 to 10.5 hours daily¹⁰. However self-reported nocturnal sleep time declines across the adolescent span just as bed times grow later and rising times earlier¹⁰. In addition adolescents have a higher tendency for daytime sleepiness which is not related to the amount of sleep at night¹⁰. Thus incidence of hypersomnia is susceptible to inappropriate estimation in adolescence. Nevertheless, hypersomnia has ominous implications for cognitive development with impairment in learning, motivation, attention span and academic achievement^{1,2,11}. Further-

more its incidence in adolescence has been associated with increased susceptibility to accidents, and depression^{3,12}. It could also signify presence of a grave medical condition such as a brain tumor⁶. Consequently childhood hypersomnia demands comprehensive assessment and management.

Studies and reports on childhood sleep and sleep related disorders, particularly in an environment like ours with limited services for sleep disorders, could increase awareness and facilitate the development and provision of such services^{7,8}. The studies could achieve these through increasing awareness, and providing data, on the need to provide diagnostic and therapeutic services for sleep disorders.

To promote awareness and highlight diagnostic challenges as well as the impact of sleep disorders in childhood in our environment we studied a 14 year old who presented with hypersomnia of unknown etiology. It was the first of such a case in the first 3000 cases seen at the Child and Adolescent Mental Health (CAMH) Unit of the Federal Neuro Psychiatric Hospital (FNPH) in Kaduna, Northern Nigeria.

Case Report

S.J. is a 14 year old male who presented to the CAMH Unit with a seven year history of excessive sleep during the day. The informants were the patient and his parents. In the 18 months prior to presentation the duration of his day time sleep had increased to an average of 45 minutes with a frequency of 4 times daily. His sleep at night was described as normal lasting an average of eight hours per night. There was no history of sleep related hallucination, weakness, snoring or that suggestive of difficulty with breathing while at sleep. The patient was not on any routine medication nor was there a history of substance abuse. There was no past history of trauma to the head, that which was suggestive of meningitis or encephalitis. The patient's EDS was initially attributed to laziness by parents and school teachers. However progression in the duration and frequency EDS necessitated several visits to the family clinic and other secondary health facilities where parents were reassured the patient would overcome his sleepiness. His growth is said to be normal and comparable to that of his siblings and peers. His academic performance was described as initially good but had gradually deteriorated in the past three years. This culminated in his being asked to repeat his last class grade in school. The patient complained about taunts from his peers in the school concerning his sleepiness. He was also unhappy about his uncontrollable propensity to sleep. The patient is the second of three children in a monogamous setting. His father is a 50 year old self-employed engineer and mother, a 43 year old unemployed university graduate. His siblings were said to be doing well academically and there no family history of hypersomnia or any mental disorder.

At presentation he was well kempt, not pale, had no cervical lymphadenopathy. His Body Mass Index (BMI) was 22.1kg/m² (Normal) while his Sexual Maturity Rate

(SMR) staging was Stage 3 (Mid- Adolescence). The initial screening assessment for sleep using BEARS sleep screening algorithm indicated that EDS was the only affected domain.¹³The Adolescent Sleep Hygiene Scale (ASHS) score was 63% of the maximum score and indicative of normal sleep hygiene.¹⁴ The Epworth Sleepiness Scale (ESS) score was 19 (Table 1), with maximal scores in all sedentary parameters, indicating EDS^{7,15}. The examination of the central nervous system was normal except for the findings of: loss of interest, sad mood, impaired concentration and suicidal ideation while using the Hospital Anxiety and Depression Scale (HADS) instrument to assess for depression¹⁶. His HADS score was 12 which is greater than the cutoff for depression. Assessment of intelligence using the Raven's Progressive Matrices indicated an average intellectual capacity and performance. The Card Agglutination Test for Trypanosomiasis (CATT) was negative. The Electro Encephalo Gram (EEG) and serum electrolytes and urea assay showed no abnormality. The Magnetic Resonance Image (MRI) scan of the brain (Fig 1) showed mild prominence of the sulci which was reported as a normal variation. In the absence of structural abnormalities and considering the fact that childhood peaks of gray matter development is followed by a decline in adolescence, the MRI was reviewed as normal by the authors¹⁷. Due to lack of facilities for sleep studies in the CAMH unit and in our region of practice polysomnography and multiple sleep latency test (MSLT) were not done. Despite subsidization the estimated cost of the investigations was a hundred thousand naira (\$ 625.00) as against the eighty thousand naira (\$500) average monthly income of the family.

Table 1: Patient's Epworth Sleepiness Scale (ESS)^{8,15} score

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

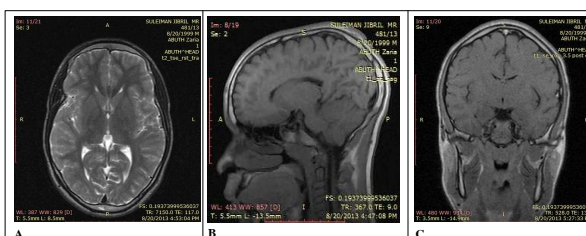


Fig 1. (A) Axial cut showing normal cerebral mantle and ventricles. (B) Sagittal and (C) Coronal cuts showing mildly prominent sulci.

In view of the indeterminate findings and duration of sleep at night (less than 10 hours) a clinical diagnosis of hypersomnia of unknown etiology (idiopathic hypersomnia) without long sleep time, with complicating depression, was made. The patient was commenced on psychotherapy, oral amitriptyline (50mg b.i.d.) for depression and given clinic appointments to monitor his management. Depression and the confounding expression of suicidal ideation necessitated our prescription of an anti-depressant. The prescription of other effective anti-depressant drug such as fluoxetine, of the selective serotonin re uptake inhibitors' class, was not considered because of the history of suicidal ideation in the patient which itself is a side effect of the drug. Furthermore, the effectiveness of the use of a wake-promoting drug such as modanafil or that of a stimulant such as methyl phenidate that has been demonstrated to provide variable successes in the treatment of idiopathic hypersomnia could not be ascertained because the drugs are not available in our practice.^{3,18} He was introduced to a series of absorbing leisure time activities while counseling sessions were organized for him and other members of his family. Informative interactive sessions were also undertaken with his class teachers and school authority.

Discussion

Hypersomnia of unknown etiology in this Nigerian adolescent was characterized by a seven year delay in diagnosis, high cost of investigations and limited facilities for diagnosis of sleep disorders. Its impact included a poor academic performance, depression, felt stigma, suicidal ideation and increased family expenditure. Psycho social implications of hypersomnia have been reported from other regions of the world as well as the dearth of facilities for sleep studies in the country.^{1-3,7,8} It took seven years and a significant progressive deterioration in academic performance before a concerted effort was undertaken by his caregivers to unravel the disorder. It underscores the poor degree of awareness about sleep disorders in the patient's immediate environment. The perception of EDS as a form of laziness and that he will outgrow it further buttresses the assertion.

The presentation of hypersomnia in adolescence could have significant implications.¹⁻⁴ It could reflect a past history of head injury, encephalitis or, indicate the presence of substance abuse.⁴ It could also signify the presence of a potentially ominous condition such as a brain tumor.⁷ Furthermore, it could be indicative of more specific sleep disorders such as narcolepsy, which is additionally characterized by cataplexy and hallucinations, and Klein-Levin syndrome which has megaphagia and increased sexuality as complimenting features.^{18,19} These enumerated features that have been associated with hypersomnia were absent in our patient. However he did

present with depressive symptoms. Depression is often debated as etiological or an outcome in hypersomnia.³ Depressive symptoms have been noted in 15-25% of patients with idiopathic hypersomnia.³ In our patient depression was considered as an outcome because of the development of depressive symptoms after onset, and with increasing severity, of EDS. The implication of depression is particularly worrisome in the adolescent as it reduces adolescent productivity, increases the risk of substance abuse and suicidality, and that of involvement with the juvenile justice system.²⁰

The management of hypersomnia due to medical conditions focuses on the treatment of the etiological factors. However there is currently no cure for idiopathic hypersomnia.^{18,19} Its contemporary scope of management include the use of wake promoting medication such as modanafil and stimulants such as methyl phenidate even though they have been reported not to be very effective in the management of idiopathic hypersomnia.^{18,19} Behavioral approaches and sleep hygiene techniques have also been employed with overall little beneficial effects.¹⁹ Spontaneous improvement in symptoms has been reported in a minority among a cohort of patients with idiopathic hypersomnia.¹⁸

The relative high cost of accessing health care for hypersomnia in this study highlights the challenge that could confront accessing health care for uncommon or complex health disorders in our environment with resultant defaults in management. This is even more remarkable in settings where there are no supportive or limited health care funds to access. Additionally, as it is with most chronic disorders,²¹ hypersomnia could impact negatively on the physical, economic and psychosocial wellbeing of other family members.

Lack of facilities for conducting sleep studies and management of sleep disorders were limitations in the study. The weakness in the health system's ability to provide readily accessible and available services for sleep disorders, as observed in this study and other studies from the country,^{7,8} is probably influenced by the health systems need to address other more common childhood disorders. Improving the health system's capacity to manage sleep disorder would enhance awareness, promote favorable outcomes and strengthen health care delivery in general.

Hypersomnia was associated with diagnostic challenges and detrimental psycho social outcomes in this study. Improved awareness and a better outcome with hypersomnia, and other sleep disorders, could be achieved through communal enlightenment campaigns, instructional school health initiatives, provision of diagnostic facilities in the health system, training of teachers and child health practitioners in the management of sleep disorders.

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