Case Report

Workplace Implications of Idiopathic Hypersomnia in the Setting of Occupational Sleep Medicine: A Case Report and Literature Review

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Abstract

Background and Objective: Idiopathic hypersomnia (IH) manifests with excessive daytime sleepiness (EDS), cognitive and autonomic symptoms, debilitating sleep inertia, difficulty awakening, prolonged sleep duration, and non-restorative sleep. Here, we present a case with IH who was a shift worker in an automobile factory. Workplace implications and fitness for work in safety-sensitive jobs are especially important for patients with hypersomnolence, because rates of accidents increase in these situations.

Case Report: The patient was a 34-year-old man referred to Baharloo Sleep Clinic, Tehran, Iran, because of EDS. He was a shift worker in an automobile factory. The subject underwent multiple sleep latency test (MSLT) after overnight polysomnography (PSG). He had no sleep onset rapid eye movement (REM) periods in MSLT and the mean sleep latency (MSL) was 5.75 minutes. According to PSG and MSLT report, the diagnosis of narcolepsy was ruled out. According to his sleep log, the patient slept more than 11 hours per 24 hours and had MSL lower than eight minutes. Our patient used modafinil for improvement of his symptoms and he was unfit for working in night shifts.

Conclusion: Here, we reported a case of IH. Attention to past medical history, occupational history, sleep log or actigraphy, PSG, and MSLT findings is very helpful for diagnosis and confirmation of IH. Pre-employment and periodic evaluation of sleepiness, accident analysis, and tracking in an integrative sleepiness or fatigue management system would be very useful.

Keywords: Idiopathic hypersomnia; Sleepiness; Workplace

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Introduction

Idiopathic hypersomnia (IH), first introduced by Roth in 1956, manifests with excessive daytime sleepiness (EDS), cognitive and autonomic symptoms, debilitating sleep inertia, difficulty awakening, prolonged sleep duration, and non-restorative sleep. A familial predisposition for this chronic neurological disorder has been suggested (1-3). Enhanced activity at gamma-aminobutyric acid (GABA)-A receptors in cerebrospinal fluid of these patients and other central disorders with hypersomnolence has been detected (4).

Some of the differential diagnoses of hypersomlonence include narcolepsy type 1 and 2 and IH, but the most effective treatment for all of these conditions is modafinil. Multiple Sleep Latency Test (MSLT) following overnight polysomnography (PSG) needs to be done for definite diagnosis (1). According to International Classification of Sleep Disorders-Third Edition (ICSD-3), the diagnosis of IH is based on the presence of all these criteria: 1) daytime periods of irresistible sleep occurring for at least three months, 2) absence of cataplexy, 3) fewer than two sleep onset rapid eye movement (REM) periods or no sleep onset REM periods in MSLT, if REM latency, reported by overnight PSG, was ≤ 15 minutes, 4) mean sleep latency (MSL), reported by MSLT, ≤ 8 minutes or total sleep time (24-hour) ≥ 660 minutes (reported by PSG or combination of actigraphy and sleep log), 5) ruling out insufficient sleep

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syndrome (ISS), and 6) ruling out other sleep, psychiatric, or medical disorders and medication use, which manifest with hypersomnolence (5).

Here, we present a case with IH who was a shift worker in an automobile factory. Workplace implications and fitness for work in safety-sensitive jobs are especially important for patients with hypersomnolence, because rates of accidents increase in these situations (6). Thus, in this case report, we present a worker with IH and discuss about his workplace implications in the setting of occupational sleep medicine.

Case Report

The patient was a 34-year-old man referred to Baharloo Sleep Clinic, Tehran, Iran, because of EDS. He was a shift worker in an automobile factory and his employer reported few sleep attacks which had been led to some work-related accidents. His main task was working in robotic section and he worked three 8-hour shifts: morning, evening, and night shifts. He had two motor vehicle accidents outside of the factory. He was married but he had no children and his education level was associate degree. After taking history, the following data were obtained:

History of hypersensitive airway disease which caused some work limitation (no or limited exposure to air pollutants, allergens, and irritants). Drug history was negative. He reported occasional alcohol use.

In sleep history, he had no symptoms of snoring, apnea, insomnia, cataplexy, sleep paralysis, or hypnogogic and hypnopompic hallucination. The main complaints were hypersomnolence and sleep attacks with onset at 33.

After filling the questionnaires, Epworth Sleepiness Scale (ESS) score was 23 (normal < 10) (7). Beck Depression Inventory-II (BDI-II), Insomnia Severity Index (ISI), and STOP-BANG scores were 16 (normal range: < 14) (8), 14 (normal range: < 8) (9), and 3 (normal range: < 3) (10), respectively. Body mass index (BMI) was 23 and blood pressure (BP) and heart rate were normal.

The subject underwent MSLT after overnight PSG. PSG characteristics are shown in table 1.

He had no sleep onset REM periods in MSLT and the MSL was 5.75 minutes. After taking blood sample for human leukocyte antigen (HLA) typing, following HLAs were reported: DQB1*0602, DQA1*0102, DRB1*15, DQA1*0505, DRB1*11, and DQB1*03.

Table 1. Polysomnographic (PSG) characteristics

Characteristics	
Total sleep time (minute)	493.5
Time in bed (minute)	557.6
Sleep efficiency (%)	88.5
Sleep onset latency (minute)	27.5
REM latency (minute)	55.5
Wake after sleep onset (minute)	36.6
Stage N1 (%)	20.6
Stage N2 (%)	50.8
Stage N3 (%)	5.9
Stage REM (%)	22.7
RDI	5.7
Apnea (n)	5
Hypopnea (n)	42
Mean O2 saturation (%)	93.0
Minimum of O2 saturation (%)	88.0

REM: Rapid eye movement; RDI: Respiratory Disturbance Index

He filled out sleep log two weeks before MSLT which is shown in table 2.

Table 2. Sleep log of the subject

Day	AM												PM												AM						
	8	9	10	11	12	1	2	3	4	5	6	7	8	9	10	11	12	1	2	3	4	5	6	7							
1																															
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Dark blue out cells indicate patient being asleep

Discussion

The patient was referred with EDS occurring for years. He had severe EDS (ESS score: 23) (7). According to PSG and MSLT report, the diagnosis of narcolepsy was ruled out (no sleep onset REM periods in MSLT). Respiratory Disturbance Index (RDI) in PSG was 5.6/hour [mild obstructive sleep apnea (OSA)] (5). MSL in MSLT was 5.75 which is categorized as moderate sleepiness (11).

According to his sleep log, the patient slept more than 11 hours per 24 hours and had MSL lower than eight minutes. After ruling out ISS (according to mean total sleep time in his completed sleep log), other medical and psychiatric disorders (according to patient self-reports and examination of sleep medicine specialist), we diagnosed the disorder as IH (5).

After HLA typing, he had these HLAs: DQB1*0602, DQA1*0102, DRB1*15, DQA1*0505, DRB1*11, and DQB1*03. Some of these HLAs such as HLA DQB1*0602 are found more in patients with narcolepsy and IH compared to general population (12). This shows that sticking to ICSD-3 criteria for diagnosing causes of sleepiness is very important and HLA typing may mislead us in the diagnosis of IH.

Patient's treatment was performed with non-pharmaceutical interventions and concomitant pharmaceutical therapy with a wake-stimulating agent (100 mg modafinil per day). The recommended non-pharmaceutical interventions for the patient included: education of sleep hygiene (stimulus control and sleep restriction), napping during work (during noon for 20 minutes), exercise before evening (13), and also a letter towards patient's employer for implementation of several workplace limitations. Following work limitations were recommended to his employer: not being assigned for safety-sensitive tasks requiring a high level of alertness, such as driving and limiting night shift. We recommended him to have regular follow-ups for tracking symptoms and also extending occupational limitations.

Shift work restriction was also recommended and he was said to work only in the morning or evening shifts. The important point about this patient is that he was referred from his company because of sleepiness. However, a lot of workers and employees do not refer or be referred from their organization, because of this problem as many occupational accidents occur and accident analysis is not performed for them (14). Accordingly, further evaluation of the workers with sleepiness and accident analysis by organizations are very important issues for increasing performance and quality of life (QOL) in the workers.

Workplace limitations of sleepiness are little discussed in sleep medicine-related texts and most of the recommendations (bright light, sleep education, napping during shift, and other interventions such as exercise) focus on sleepiness because of shift work and sleep loss (15). This may be due to different tasks of workers in various workplaces; thus, a simple rule cannot be implemented for all the workers with sleepiness especially the ones who suffer from sleep disorders such as IH or narcolepsy. As mentioned above, we recommended our patient several limitations at the workplace which the main one was limiting night shifts. However, as the recommended non-pharmaceutical interventions were not efficient despite of concomitant consumption of wake-stimulating medication, i.e., modafinil, the patient had to change his job and even he moved to another city for changing his job. Evidence for effectiveness of non-pharmaceutical interventions in the management of IH is lacking and further research is required for improvement of symptoms in patients with IH (16).

Furthermore, in the last sleep clinic visit, the patient still was complaining of EDS. Our patient used modafinil for improvement of his symptoms. Emerging evidence and ongoing clinical trials may authorize new wake-stimulating agents such as sodium oxybate, pitolisant and solriamfetol in patients with IH (these drugs are not available for our patients at the current time in Iran) (16). Insufficient social support of these patients also can aggravate their symptoms. Developing non-governmental networks for these patients with more introduction of social and workplace problems of these patients may be a helpful strategy in this regard. Furthermore, we know that motor vehicle accidents are higher in these patients. Uncontrolled sleepiness limits social, driving, and workplace activities of such patients. So, developed public transportation services would be very helpful for such people for promotion of QOL.

Alert and collaborative workplace and organization is very helpful for management of a worker with excessive sleepiness especially patients with IH. A collaborative workplace would be the one which screens and tracks its employees for sleepiness and related accidents and enforces medically-prescribed limitations for these patients in its organization (14).

We think that according to the chronicity of sleepiness in the diseases such as IH or narcolepsy, pre-employment evaluation of these patients and tracking workplace accidents of the new employed workers regarding sleep disorders would be very crucial. Accordingly, safety-sensitive jobs will not be performed by high-risk workers and they will be managed appropriately. Monitoring sleepiness-related accidents and screening of symptomatic workers in an integrative sleepiness or fatigue risk management system (FRMS) would be another appropriate strategy for prevention of occupational accidents. At the current time, there is no FRMS specific for such patients (17); most of them are focusing on sleepiness that is because of sleep loss not sleep disorders such as IH. Patients

with IH or narcolepsy suffer from more sleepiness or different types of sleepiness according to pathophysiology of the disease. Moreover, shift scheduling programs are also focusing on decreasing shift effects of normal people. As we know, if we consider early definition of early morning shifts, every worker may be a kind of shift worker (14), so limiting shifts for these people may impact their occupational careers and QOL. Therefore, disease-specific shift scheduling and more investigation in this regard would be beneficial. This field in the occupational sleep medicine setting needs more exploration and clinical trials (18).

Another gap of knowledge is that occupational restrictions for these patients, as the available ones, are very general and are recommended for every type of sleepiness. Randomized controlled trial (RCT) in this regard also would be very helpful. Disease-specific organizational strategies would promote job performance and QOL of patients with IH. Despite all available evidence regarding management of IH, controversies about these patients remain which are complex, poorly understood, and need further elucidation. These controversies include the component of circadian system disturbance and mild sleep apnea of this patient (our patient also had mild sleep apnea and he refused treatment of this issue) (19, 20).

Another important raising issue regarding occupational implications of patients with IH is that as sleep medicine specialists, we do not pay attention to patients' jobs and their tasks appropriately during routine visits. Thus, some of patients with reduced job performance or decreased QOL are being overlooked. Patients with sleepiness may be drivers or may perform safety-sensitive tasks which will not be explored if we do not take thorough medical and occupational history. Accordingly, occupational implications of such patients are overlooked during sleep clinic visits, leading to more occupational accidents and/or reduced job satisfaction and performance (21). Thus, special attention of sleep specialists to this issue is very important.

Conclusion

Here, we reported a case of IH. Attention to past medical history, occupational history, sleep log or actigraphy, PSG, and MSLT findings is very helpful for diagnosis and confirmation of IH. However, HLA typing results could be misleading, as some of HLAs are common with narcolepsy. Pre-employment and periodic evaluation of sleepiness, accident analysis, and tracking in an integrative sleepiness or fatigue management system would be very useful. Moreover, sleep medicine specialists must pay attention to occupational tasks of their patients for further management of the sleep disorder and promotion of QOL.

Conflict of Interests

Authors have no conflict of interests.

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