Sleep and Vigilance
https://doi.org/10.1007/s41782-019-00071-7

ORIGINAL ARTICLE

A Case of Severe Delayed Sleep–Wake Phase Disorder and Simultaneous Restless Legs Syndrome

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Received: 22 May 2019 / Revised: 18 July 2019 / Accepted: 31 July 2019

Abstract
Patients with delayed sleep–wake phase disorder (DSWPD) have an extremely delayed endogenous circadian rhythm. They are unable to sleep until late at night and therefore need to sleep for at least the first half of the day. This in turn often results in major problems with school and work. The symptoms of restless legs syndrome (RLS) follow the circadian rhythm. According to the definition, patients with RLS have worse symptoms in the evening or night than during the day. We present a case of DSWPD where sleep begins at 8 a.m. and the symptoms of RLS begin one or more hours beforehand, which could support the theory that RLS symptomatology is under circadian control.

Keywords Restless legs · Delayed sleep · Circadian rhythm

1 Introduction

The term «delayed sleep phase» was described as late as 1981 by Weitzman et al. [1], who also minted the clinical and scientific term: Delayed Sleep Phase Syndrome (DSPS). However, according to International classification of sleep disorders 3 (ICSD 3) the new name is now delayed sleep–wake phase disorder (DSWPD). At least 0.17% of adult Norwegian citizens are likely to suffer from DSWPD [2]. Despite this, there is still very little research in the area, and the syndrome is considered to be greatly underdiagnosed by doctors and quite unknown to the public.

Difficulties at school in the form of learning disorders and adaptation to the requirements of working life are common among those suffering from DSWPD. As therapeutic options are available for DSWPD, clinical awareness and early diagnosis are important for this type of circadian rhythm sleep disorder [3]. Early diagnosis and effective treatment can contribute to preventing great suffering and undesirable consequences in the form of negative impact on social life, such as inadequate grades in school or difficulties in connection with choosing a profession.

Sleep is controlled by both circadian and homeostatic mechanisms. The light–dark cycle is the backbone of the circadian clock. The circadian clock has been localised to the suprachiasmatic nucleus in the hypothalamus. This endogenous clock is normally set to a period of longer than 24 h and is normally trained to adapt to various rhythms in our external environment (light/dark, stimulus in the form of work, sound, etc.). Poor adaptation of the circadian clock can lead to circadian rhythm sleep disorders. In patients with DSWPD, this endogenous clock has likely become «stuck» in a delay of the circadian rhythm when compared with the desired sleep-awake schedule [3].

The pathophysiology of DSWPD patients is far from established. However, one essential pathologic mechanism has been established: the homeostatic and circadian mechanisms, which govern sleep, are disrupted in DSWPD [3].

Restless legs syndrome (RLS), also known as Willis–Ekbom disease (WED), is considerably more common than DSWPD, and is likely more well-known both within healthcare and among the public. In Sweden, RLS is reported among 11.4% of women in the general population [4]. Based on criterion 4 in the diagnostic criteria for RLS established by the International RLS Study Group (IRLSSG) in 1995, the urge to move the legs and any accompanying unpleasant sensations during rest or inactivity only occur or are worse in the evening or night than during the day [5, 6].

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We present the case of a patient suffering from both DSWPD and RLS.

2 Case

A 25 year-old woman referred herself to our sleep center due to a delayed circadian rhythm which she experienced during the last 10 years. From having had a time of sleep onset around 10–11 p.m., the sleep onset shifted, over a short period, to 8 AM. She had only made medical appointments on a few occasions and been offered hypnotics such as Zolpideme and Zopiclone, but these attempted therapies have had no effect. No understanding could be reached with regard to the underlying sleep disorder. High school was problematic; she was absent much of the time, but she managed to complete her studies. She discovered that if she were to sleep for a number of hours during the day, she was able to function well in the evening and during the night, which is why she worked nights in the hotel sector. She had problems initiating and maintaining social contact due to her great need of sleep during the daytime. Eventually, the patient found information on the internet regarding Delayed Sleep Phase Syndrome.

The woman also reported that 2–3 h before sleep onset she experienced a tingling sensation in both lower legs while resting, where physical activity immediately resulted in total relief of the problem. During the visit to our clinic, she described typical symptoms for RLS and the sleep expert diagnosed mild to moderate RLS. Despite DSWPD and RLS, she was able to fall asleep and experienced undisturbed sleep until she awoke thoroughly rested 8–9 h later.

The patient was mainly interested into get the diagnosis of DSWPD confirmed by a sleep expert. However, she was not interested in further examinations like actigraphy and/or polysomnography, or in the treatment of her delayed sleep phase. She had accepted her current life style with night shifts and following sleep during the daytime. She however decided to intermittently treat her RLS problems with short-acting levodopa on demand, with great success.

3 Discussion

To our knowledge, this case is the first in literature with a typical clinical picture of RLS in a patient with an extremely delayed sleep phase. The RLS symptoms begin hours before sleep onset, early in the morning and not in the evening or night as stated in the internationally accepted definition. The validity of a RLS diagnosis can be verified by a strong and repetitive treatment response of levodopa.

The background of the symptom development of RLS as per criterion 4 of IRLSSG's criteria being most prominent hours before and putative sleep onset is still unclear, according to a review of the subject by Baier and Trenkwalder [7]. Our case study supports the theory that the symptoms of RLS are dependent on mechanisms of the circadian rhythm.

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