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Research paper

Sleep and disruptive mood dysregulation disorder: A pilot actigraphy study

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ABSTRACT

Objective: To explore the clinical characteristics and motor activity profile during sleep periods of children and adolescents presenting with disruptive mood dysregulation disorder (DMDD).

Method: Twenty-one youths (mean age \pm standard deviation, 11.7 ± 3 years) wore a wrist actigraph for 9 consecutive days (including both school days and non-school days), to measure sleep parameters: sleep latency, sleep efficiency and the number and duration of periods of wakefulness after sleep onset (WASO). We divided the night-time actigraphy recording sessions into three sections and compared the first and last thirds of the night.

Results: All the study participants had a psychiatric comorbidity (primarily attention deficit hyperactivity disorder, depressive disorder or anxiety disorder). On non-school days, bedrest onset and activity onset were shifted later by about 1 h. There was no significant difference between school days and non-school days with regard to the total sleep time. Sleep efficiency was significantly greater on non-school days. Sleep was fragmented on both school days and non-school days. The mean number of episodes of WASO was 24.9 for school days and 30.9 for non-school days. Relative to the first third of the night, we observed a significantly greater number of episodes of WASO during the last third of the night, a period associated with a larger proportion of rapid eye movement (REM) sleep.

Discussion: Sleep appeared to be fragmented in the study population of youths with DMDD. The greater frequency of WASO in the last third of the night points to a possible impairment of the motor inhibition normally associated with REM sleep.

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1. Introduction

In pediatrics, emotional and behavioral disturbances are recurrent reasons for consultation. Interest in these disturbances has grown since they were recognized as a specific syndrome, namely disruptive mood dysregulation disorder (DMDD). This syndrome emerged from discussions about the prepubertal phenotype of bipolar disorder (BD) [1,2] and first appeared in the Fifth Edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) in 2013 [3]. According to the DSM-5, DMDD is marked by temper outbursts manifested verbally and/or behaviorally that are:

- grossly out of proportion in intensity or duration to the context;
- inconsistent with the child's developmental age and the context of the occurrence.

A further defining criterion is persistent dysphoric, angry or irritable mood between temper outbursts. The DSM-5 also lists a number of quantitative criteria required for a diagnosis of DMDD: at least three outbursts a week, the occurrence of outbursts in at least two settings (at school, at home, and/or with peers), the presence of outbursts for at least 12 months (and no periods without symptoms lasting 3 or more consecutive months) and an age of onset before 10 years. The condition can be diagnosed in children from the age of 6 and in adolescents. A diagnosis of DMDD cannot coexist with BD or oppositional defiant disorder (ODD). The clinical description given in the DSM-5 does not include excitability symptoms that are associated with attention deficit

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disorder with/without hyperactivity (ADHD) and listed among the diagnostic criteria initially suggested by Leibenluft [4]. DMDD tends to turn into a depressive disorder (with which it is classified in the DSM-5) rather than a BD [1,2].

In view of this very recent change in the diagnostic category, few epidemiological data on DMDD are available. A survey by Copeland et al. (based on the populations in the Duke Preschool Anxiety Study, the Great Smoky Mountains Study and the Caring for Children in the Community Study) estimated the prevalence of DMDD at between 0.8 and 3.3% [5]. This new syndrome is associated with a non-negligible comorbidity rate. The original study by Brotman et al. [4] showed that about a quarter of clinically severe mood dysregulation disorders were associated with:

- a disruptive disorder;
- a sevenfold increase in the risk of a depressive disorder.

With reference to the DSM-5 criteria for DMDD, the aforementioned survey by Copeland et al. [5] confirmed this association and estimated the odds ratio (OR) at between 9.9 and 23.5 (depending on the sample) for depressive disorders and between 52.9 and 103.0 for ODD. The latter association is no longer relevant because the two DSM diagnoses are mutually exclusive. The DMDD diagnostic category is expected to capture the clinical picture displayed by youths who express emotion dysregulation through behavioral disturbances. DMDD and ODD share common features, especially irritability and oppositional behaviors. Strong overlap between DMDD and ODD has been observed in cross-sectional studies. In a general population 6- to 12-year-old sample ($n = 665$), the presence of ODD and DMDD symptoms was investigated using the mother's ratings on a pediatric behavior scale [6]. Sixty-six percent of children with ODD had DMDD symptoms, supporting the claim that DMDD should be viewed as a disruptive disorder rather than depressive disorder subtype. However, longitudinal studies showed that youth irritability predicted adult depressive disorders instead of disruptive outcomes (see [1] for review).

No longitudinal studies of DMDD per se have been conducted, other than those dealing with its semiological components (e.g., irritability and mood dysregulation). The latter appears to be present from early childhood onwards and its progression is associated with a greater risk of suicidal behavior [7,8]. Hence, it is too early to know whether this new syndrome is related to anxiety disorders (and notably separation anxiety disorders) or whether it subsequently progresses to a personality disorder. Clinically, DMDD is a source of great suffering for the child, who is desperate to stop the outbursts and build a more placid relationship with his/her family and friends. The child's parents tend to be particularly disoriented by the intensity of the temper outbursts, since they cannot control the outbursts' determinants or intensity and are thus unable to relieve the child's suffering [9]. The emergence of the child's emotional control develops through constant interactions with caregivers. Indeed, the intersubjective component has been confirmed by developmental studies showing that the progression of mood dysregulation in childhood is mediated by the quality of parenting and peer relations [8]. In this context, the first step in management is to restore the child's and parents' self-esteem, and explore a number of treatment options that, for the moment, are essentially symptomatic and nonspecific [10,11]. Cognitive remediation may be an avenue worth exploring [12].

In the absence of an animal model of DMDD, current research is focusing on the disorder's neuronal and cognitive correlates in general and the cognitive aspects of facial expression processing in particular. A now dated article by Leibenluft et al. [13] set out several avenues for psychopathological and physiological research, with regard to the various potentially determinant components of DMDD. Considering the pre-eminent symptom (temper outbursts),

the first component to be described is irritable mood. The threshold for emotional release is lower than for most individuals of the same age, which therefore induces intense, long-lasting outbursts with a longer recovery period. The second component to be described is a possible impairment in motor inhibition; the individual is not able to anticipate or prevent temper outbursts. The question then arises as to whether the patient's levels of basal disinhibition or basal motor activity are greater than for other children of the same developmental age, while taking account of the circadian distribution of the individual's basal motor activity. A number of noninvasive measurement tools are now available; these include actigraphy, the use of which is widespread in the field of ADHD and which has been applied to studies of affective disorders in adolescents [14,15]. One advantage of actigraphy is its ability to record wake-sleep cycles over several days; this enables the recording of both periods with zeitgebers (i.e., constraints that influence the child's schedule, such as a school schedule) and periods without. Recording both types of period provides greater insight into circadian features (and notably successive periods of rest and activity), while taking into account external constraints on the child's schedule.

Actigraphy can provide a better understanding of sleep patterns in youth with DMDD. Although insomnia was included in Leibenluft's initial criteria [4] but not included in the DSM-5 criteria, subjectively assessed sleep problems are often reported by children and adolescents with DMDD and/or by their parents. The link between sleep alterations and psychiatric disorders has been clearly established in several domains (such as suicidal behavior, depression and oppositional defiant disorder) [16]. Given that DMDD combines disturbances in two domains (mood and relationships), we hypothesized that it might be associated with disturbed sleep.

Hence, the objective of the present pilot study was to perform an actigraphic investigation of sleep/wakefulness patterns in youths with DMDD. With respect to recent epidemiological studies [17], it seems uneasy to discriminate between DMDD and other mental disorders in youth based on symptomatology only. Consequently, it is suggested to explore other sources of clinical information, e.g., markers of the reactivity of the autonomic nervous system [17]. In the same vein, this study will contribute to exploring the usefulness of actigraphy in examining DMDD in youth.

2. Methods

2.1. Procedure

The study's procedures were approved by the local independent ethics committee (CPP Nord Ouest 2, Amiens, France). The study participants meet the following inclusion criteria:

- male or female gender;
- age between 6 and 16 years of age;
- referral to our child and adolescent psychiatry out-patient clinic for an indication of suicidal ideation or behavior;
- a clinical diagnosis of DMDD.

Youth presenting with intellectual impairment, a major developmental disorder, a psychotic disorder, drug/alcohol abuse at the time of admission or sleep/vigilance disorders with an organic etiology (respiratory and motor disorders of sleep, narcolepsy, etc.) were not included.

If a youth met all of the inclusion criteria and none of the exclusion criteria, his/her legal representative was given initial verbal or written information on the study's objectives and procedures. The legal representative was asked to then give the

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