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SCIENTIFIC INVESTIGATIONS

A series of 8 cases of sleep-related psychogenic dissociative disorders and proposed updated diagnostic criteria

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Study Objectives: To identify the most relevant clinical and video-polysomnographic characteristics of patients with sleep-related dissociative disorders (SRDDs) and to propose a framework for new diagnostic criteria.

Methods: We searched potential SRDD cases from the scientific literature and from a database of patients referred for clinical and video-polysomnographic assessment in a single sleep disorders center for disruptive nocturnal behaviors (n = 731). The most relevant clinical and neurophysiological characteristics of the cases were extracted and a descriptive analysis was performed.

Results: Twenty-six SRDD cases (8 new and 18 previously published cases) were reviewed. Almost all cases of SRDDs occurred in a context of past traumatic events or abuse and were associated with at least 1 comorbid psychiatric disorder. We highlighted 4 relevant clinical characteristics of SRDD useful for the differential diagnosis with parasomnias: episodes of long duration of more than 1 hour (90.9%), self-inflicted injuries (83.3%), occurrence while awake close to bedtime (35.7%), and the presence of daytime dissociative symptoms (72.7%). The video-polysomnography documented typical episodes of SRDD with prolonged wakefulness before, during, and after the event in 11/26 cases. New diagnostic criteria for SRDD were proposed, with 3 levels of certainty for the diagnosis based on clinical, video-polysomnographic, and homemade video findings.

Conclusions: More than 30 years after its formal identification, SRDD is not currently recognized as an official diagnostic entity. We better delineate the clinical and neurophysiological features of SRDD and propose a framework for its reinstatement in the next revisions of the sleep and psychiatric disorders classifications.

Keywords: sleep-related dissociative disorders, dissociative disorders, parasomnias, non-REM sleep parasomnias, psychogenic nonepileptic seizures, diagnosis, psychiatric disorders

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BRIEF SUMMARY

Current Knowledge/Study Rationale: Although sleep-related dissociative disorders were formally described in 1989 and recognized as a diagnostic entity 15 years later in the second edition of the *International Classification of Sleep Disorders,* they are not included in the current psychiatric and sleep diagnostic classifications. Sleep-related dissociative disorders are often severe and debilitating conditions that require proper diagnosis for optimal management.

Study Impact: The clinical and video-polysomnographic findings from a new consecutive case series of 8 patients with sleep-related dissociative disorders gathered at a single center and from 18 published cases were systematically reviewed. We propose a framework for new diagnostic criteria of sleep-related dissociative disorders with three levels of diagnostic certainty, for inclusion in future editions of diagnostic classifications.

INTRODUCTION

An update on reported cases of sleep-related (psychogenic) dissociative disorders (SRDD) has recently been published in this journal,[1] with a call for additional cases of SRDD to be published on this uncommon condition that can be confused with parasomnias or sleep-related hypermotor epilepsy. Therefore, sleep clinicians should become better informed about SRDD as more cases are published, with important management implications. A defining objective hallmark of SRDD during video-polysomnography (vPSG) involves abnormal behaviors emerging during sustained electroencephalography wakefulness, either during wake–sleep transitions or after awakenings from light non-rapid eye movement (NREM) sleep, typically in patients with daytime dissociative disorders and other major psychopathology. SRDD had previously been included in the Parasomnias section of the *International Classification of Sleep Disorders*, second edition (ICSD-2).[2] Unfortunately, for inadvertent administrative reasons, this diagnostic entity was not included in the ICSD-3, even though it was included in the differential diagnosis of various NREM sleep and rapid eye movement (REM) sleep parasomnias in the ICSD-3 and in the

Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5).[3,4] SRDD was initially shifted from the Parasomnias section to the Psychiatric Disorders section of the ICSD-3. However, SRDD could not be included in the Psychiatric Disorders section of ICSD-3 because only sleep disorders contained in the psychiatric nosology (DSM-5) could be included in the Psychiatric Disorders section of ICSD-3. Although the DSM-5 contained dissociative disorders, it did not include SRDD in its sleep disorders section.

The clinical background for SRDD involves dissociative manifestations that can be defined as "unbidden intrusions into awareness and behavior, with accompanying losses of continuity in subjective experience and/or inability to access information or to control mental functions that normally are readily amenable to access or control."[1] Dissociative disorders are frequently found in the aftermath of trauma and encompass various clinical presentations such as dissociative identity disorder, dissociative amnesia, and depersonalization/derealization disorders.[4] SRDD were formally recognized in 1989 as a mimic of sleepwalking, in a case series of 8 patients reported from a single sleep disorders center.[5] The original diagnostic criterion for SRDD in the ICSD-2 was "a dissociative disorder, fulfilling *Diagnostic and Statistical Manual of Mental Disorders*, fourth edition diagnostic criteria is present and emerges in close association with the main sleep period," together with at least 1 of the following features: "the polysomnography demonstrates a dissociative episode or episodes that emerge during sustained wakefulness, either in the transition from wakefulness to sleep or after an awakening from Non Rapid Eye Movement (NREM) or Rapid Eye Movement (REM) sleep" and "in the absence of a polysomnographically recorded episode of dissociation, the history provided by observers is compelling for a sleep related dissociative disorder, particularly if the sleep related behaviors are similar to observed daytime dissociative behaviors."[2]

The diagnosis of SRDD in ICSD-2 called attention to an important intersection between the fields of sleep medicine and psychiatry, with the diagnostic criterion being a unique example in the modern history of the nosologies of sleep medicine and psychiatry that combined elements of both classifications in use at that time. If the publication of SRDD in the ICSD-2 allowed for increased awareness of this disorder among sleep experts, few new cases were reported afterward.[1] One explanation may lie in the sleep and psychiatric structure of these diagnostic criteria, making them difficult to apply, especially by the nonpsychiatrist sleep experts. Thus, these criteria would probably benefit from further refinement.

The lack of recognition of SRDD as an officially recognized disorder with established diagnostic criteria remains a problematic issue. It will hinder the proper diagnostic process and therapeutic intervention to the detriment of patient care. Case series need to be reported to further call attention to this diagnostic entity that requires specialized therapy, to better delineate its clinical and neurophysiological features and to propose a framework for a potential reinstatement of SRDD as an official diagnosis of the next revisions of the sleep and psychiatric disorders classifications.

Our first objective was to systematically extract and describe the clinical and vPSG findings from previously published cases of SRDD and then to report on a consecutive case series of patients with suspected SRDD from our own single center. Our second objective was to propose a framework for new diagnostic criteria of SRDD that may be used for further classifications in sleep medicine and psychiatry fields.

METHODS

Review of the published cases

The PubMed, PsycINFO, and Embase electronic databases were searched for literature published on SRDDs. Inclusion criteria were (1) full text available in English or French, (2) performed up to June 2021, (3) individual clinical data provided (demographic, description of the episodes, comorbidities), and (4) available vPSG findings.

Case series

We searched in our database of adult and child patients referred for clinical and vPSG assessment in a single sleep disorders center (Montpellier, France) between January 2012 and January 2021 for disruptive nocturnal behaviors (n = 731). The medical charts and the vPSG recordings of patients with suspected SRDD were carefully reviewed.

All patients with suspected SRDD and their relatives, when present, had a comprehensive medical interview by a sleep expert and a psychiatrist. A history of psychological stressors (ie, stressful event before the onset of episodes and past history of sexual, physical, or psychological abuse) was systematically assessed, as well as past or current comorbid mental disorders (posttraumatic stress disorder, borderline personality disorder, anxiety disorder, major depressive episode, suicidal behaviors, psychotic symptoms or disorder, substance use disorder, eating disorder, conversion disorder, and neurodevelopmental disorder). The presence of sleep complaints (insomnia, daytime sleepiness, nightmare and snoring, and apneas) was systematically assessed, as was the occurrence of daytime dissociative manifestations (derealization, depersonalization symptoms or disorders, dissociative amnesia, dissociative fugues, or dissociative identity disorder).[4]

Information on the frequency of the behavioral manifestations, the maximum duration, and the usual time of occurrence were noted. The degree of awareness and amnesia associated with the episodes was assessed. We collected the types of behaviors reported by the patients and potential witnesses (violent behaviors toward others, self-inflicted injuries, inappropriate sexual and eating behaviors, urination in inappropriate places, manipulation of potentially dangerous objects such as sharp objects or objects that may set a fire, going outside the home, driving or using public transport).

All patients had 1 night of vPSG assessment. Sleep stages, microarousals, periodic limb movements, and respiratory events were scored manually according to standard criteria by a single scorer (R.L.). Particular attention was paid to motor behaviors, tonic and phasic chin muscle activities during REM sleep,[6] and N3 sleep fragmentation parameters (slow-wave sleep fragmentation and slow/mixed arousal indices).[7,8]

Statistical analyses

The available demographic, clinical, and polysomnographic characteristics of the reviewed cases have been extracted and treated in a Microsoft Excel Spreadsheet (Mac OS). The SRDD cases were described using percentages for categorical variables and continuous variables grouped into categories. Statistical comparisons between the published and newly reported cases were performed by using Fisher's exact test (jamovi software version 1.6.23, The jamovi project, Sydney, Australia).

RESULTS

Literature search and summary of the cases

Eleven published articles met our inclusion criteria, including 10 single case reports and 1 case series of 8 patients, for a total of 18 cases.[5,9–18] Most of these cases were already discussed in a literature review recently published in this journal.[1] Twelve other published cases were not included due to the absence of polysomnographic findings,[19–22] as well as another single case report with documented PSG findings but with insufficient clinical information available.[23] **Table 1** presents the main clinical and polysomnographic findings of the 18 published cases.

Patients were generally young, and a large preponderance of them were female. A triggering stressful event was found in 80% of the cases and a majority of patients reported a history of sexual, physical, or emotional abuse. All patients had at least 1 psychiatric disorder, with depression and posttraumatic stress disorder being the most frequent comorbidities.

Most patients had at least weekly episodes. According to the clinical interviews, the episodes usually lasted more than 1 hour and up to 6 hours, and mostly occurred at least 1 hour after going to bed. Amnesia was always present, with a few patients reporting partial awareness or associated dream-like mentations. During the episodes, most of the cases were violent with self-inflicted injuries (frequently with knives or razors) and also with frequent violent behaviors toward others. Inappropriate sexual and eating behaviors were rarely reported. Daytime dissociative episodes or disorders were observed in 85.7% of the cases.

Behavioral episodes occurring while awake were objectively confirmed by the vPSG assessment in 11 cases (61.1%). Eight patients had episodes that fully corresponded to the clinical description and could be diagnosed with definite SRDD. Three other cases could be considered as probable SRDD as they exhibited less-typical or shorter related episodes while awake. The remaining 7 cases without documented vPSG episodes should be classified as possible SRDD.

Clonazepam treatment was prescribed to 6 patients and was found to be ineffective and even to worsen the severity and the frequency of the episodes in 2 cases.

Case series

Among 731 patients referred with disruptive nocturnal behaviors for clinical and vPSG evaluations a diagnosis of parasomnia was made in 706 (96.6%) patients, including 452 (61.8%) with disorders of arousal (DOA), 229 (31.3%) with REM sleep behavior disorder (RBD), 9 (1.2%) with parasomnia overlap disorder, 7 (1.0%) with sleep-related eating disorder, and 9 (1.2%) with sexsomnia. Also, sleep-related hypermotor epilepsy was diagnosed in 9 (1.2%) cases and nocturnal panic attacks in 8 (1.1%) patients. Finally, the diagnosis of SRDD was suspected in 8 (1.1%) patients (6 females, 4 children), with a median age of 17 years (range 8–67 years).

Case 1

A 67-year-old woman was admitted to the sleep unit for a 4-year history of nocturnal complex behavioral episodes with partial to complete amnesia. She reported very long episodes, usually lasting several hours, during which she performed complex tasks, such as going out of the house, talking to strangers, and offering sexual favors. On several occasions she drove her vehicle or took a night train. She regularly called and insulted people on the telephone, often her daughter. She also had sent long and incoherent emails to public services. The episodes usually occurred 20 minutes after bedtime. The first episode occurred after she had been visited by a judicial officer because of financial problems; she reported having burned her money during sleep the following night. The frequency gradually increased from monthly to almost daily episodes at the time of the medical consultation. The psychiatric examination revealed a borderline personality disorder, with a history of recurrent depressive episodes and suicide attempts. Her personal history was marked by several psychological traumas; however, she did not meet diagnostic criteria for posttraumatic stress disorder. The patient had no personal or family history of parasomnia. Before the sleep evaluation, several antidepressants and benzodiazepine treatments had been tried, without efficacy. The vPSG revealed a normal sleep architecture, except for long periods of quiet wakefulness without behavioral manifestations suggestive of SRDD. No behavioral or polysomnographic markers of NREM or REM sleep parasomnias were evidenced during the recording.

Case 2

A 20-year-old man had been hospitalized in a secure psychiatric unit because of severe, high-risk violent behaviors occurring during sleep. He reported no personal or family history of parasomnia. The first episodes started 4 years earlier, without a triggering stressful event. They were initially infrequent, of short duration, and nonproblematic. The first violent sleep-related behavior occurred 1 year later, when he verbally threatened his girlfriend. Several similar episodes occurred with increasing frequency, complexity, and violence. During the episodes the patient could manipulate knives and made physical and sexual threats. He never hurt anyone during these episodes, but he did injure himself. Psychiatric manifestations of dissociative behavior were also reported during the day. The initial psychiatric examination found no diagnosis of mood, personality, or posttraumatic stress disorder. He only had a mild generalized anxiety disorder. The episodes increased in frequency and intensity during a stressful stay in England. He was then repatriated to France and was admitted to a psychiatric department. Despite the prescription of psychotropic treatments (olanzapine, alimemazine, cyamemazine, and bromazepam) the

Table 1—Demographic, clinical, and polysomnographic findings in patients with SRDD.

	Published Cases (n = 18)		Case Series (n = 8)		Pooled Cases (n = 26)	
	n	%	n	%	n	%
Demographic characteristics						
Sex (female)	14/18	77.8	6/8	75.0	20/26	76.9
Age ≤ 30 y	7/18	38.9	6/8	75.0	13/26	50.0
Psychological stressors						
Stressful event(s) before the onset of episodes	8/10	80.0	4/8	50.0	12/18	66.7
History of sexual abuse	7/10	70.0	1/8	14.3	7/18	38.9
History of physical abuse	8/11	72.7	2/8	25.0	10/19	52.6
History of psychological abuse	10/10	100.0	5/8	62.5	14/18	77.8
Comorbid mental disorders						
At least 1 comorbid mental disorder	16/16	100.0	6/8	87.5	22/24	91.7
Posttraumatic stress disorder	8/12	66.7	3/8	3/8	11/20	59.7
Borderline personality disorder	4/6	66.7	3/8	37.5	7/14	50.0
Past or current anxiety disorder	5/9	55.6	6/8	75.0	11/17	64.7
Past or current major depressive episode	11/13	84.6	5/8	62.5	16/21	76.2
History of suicidal behaviors	1/4	25.0	2/8	28.6	3/10	30.0
Past or current psychotic symptoms	0/5	0.0	2/8	25.0	2/13	15.4
Past or current substance use disorder	4/8	50.0	0/8	0.0	4/16	25.0
Past or current eating disorder	2/5	40.0	1/8	12.5	3/13	23.1
Past or current conversion disorder	5/5	100.0	0/8	0.0	5/13	38.5
Neurodevelopmental disorder	2/5	40.0	0/8	0.0	2/13	15.4
Comorbid sleep disorders						
Personal history of NREM sleep parasomnia	2/3	66.7	2/8	25.0	4/11	36.4
Family history of NREM sleep parasomnia	1/2	50.0	2/7	28.6	3/9	33.3
Insomnia	2/3	66.7	4/8	50.0	6/11	54.5
Sleepiness	1/1	100.0	2/7	28.6	3/8	37.5
Sleep apnea syndrome	1/3	33.3	1/8	12.5	2/11	18.2
Nightmares	2/3	66.7	2/6	33.3	4/9	44.4
Semiological characteristics						
Daytime dissociative episodes or disorder	12/14	85.7	4/8	50.0	16/22	72.7
Frequency of episodes (at least weekly)	7/10	70.0	7/8	87.5	14/18	77.8
Maximum duration of episodes (≥ 60 min)	6/6	100.0	5/6	83.3	10/11	90.9
Time of occurrence (≤ 30 min)	1/8	12.5	4/6	66.7	5/14	35.7
Time of occurrence (60–120 min)	4/8	50.0	2/6	33.3	6/14	42.9
Time of occurrence (> 120 min)	3/8	37.5	0/6	0.0	3/14	21.4
Episodes during daytime sleep periods	1/1	100.0	4/8	50.0	5/9	55.6
Total awareness	0/17	0.0	0/8	0.0	0/25	0.0
Partial amnesia (dreamlike reports)	4/17	23.5	2/8	25.0	6/25	24.0
Complete amnesia	13/17	76.5	6/8	75.0	19/25	76.0
Violent behaviors	11/12	91.7	7/8	87.5	18/20	90.0
Interaction with other people	3/5	60.0	7/8	87.5	10/13	76.9
Manipulating sharp objects (knife, tools)	6/9	66.7	6/8	75.0	12/17	70.6
Manipulating objects that may set a fire	2/5	40.0	2/8	25.0	4/13	30.8
Going out of the home	6/9	66.7	4/8	50.0	10/17	58.8
Driving or using public transportation (train, plane)	1/4	25.0	1/8	12.5	2/12	16.7
Self-inflicted injuries	9/10	90.0	6/8	75.0	15/18	83.3

(continued on following page)

Table 1—Demographic, clinical, and polysomnographic findings in patients with SRDD. (*Continued*)

	Published Cases (n = 18)		Case Series (n = 8)		Pooled Cases (n = 26)	
	n	%	n	%	n	%
Violent behaviors toward other	3/5	60.0	6/8	75.0	9/13	69.2
Inappropriate urination	1/2	50.0	1/8	12.5	2/10	20.0
Inappropriate sexual behaviors	1/3	33.3	3/8	37.5	4/11	36.4
Inappropriate eating behaviors	1/3	33.3	0/8	0.0	1/11	9.1
Video-polysomnographic findings						
Disrupted behaviors occurring while awake	11/18	61.1	3/8	37.5	14/26	53.8
Before sleep onset	5/11	45.5	2/3	66.7	7/14	50.0
After an awakening	7/11	63.6	2/3	63.6	9/14	64.3
NREM sleep behavioral episode/N3 sleep fragmentation	1/12	8.3	3/8	37.5	4/20	20.0
REM sleep behavioral episode/REM sleep without atonia	1/9	11.1	1/8	12.5	2/17	11.8
Epileptic abnormalities	0/12	0.0	0/8	0.0	0/20	0.0
Treatment						
Positive response to clonazepam treatment	0/6	0.0	2/4	50.0	2/9	22.2
Worsening with clonazepam treatment	2/6	33.3	0/4	0.0	2/9	22.2

NREM = non-rapid eye movement, REM = rapid eye movement, SRDD = sleep-related dissociative disorders.

nocturnal episodes persisted during the hospitalization. Most often they occurred 30 minutes after bedtime, and also during naps. A withdrawal from the pharmacologic treatments had been carried out before the PSG examination, without aggravation of the nocturnal manifestations. The PSG recording revealed a nonviolent, 3-minute episode of confusion, 45 minutes after falling asleep. The episode started after 5 minutes of stable wakefulness after an awakening from N2 sleep. The sleep structure was normal, and there was no argument for REM or NREM sleep parasomnias.

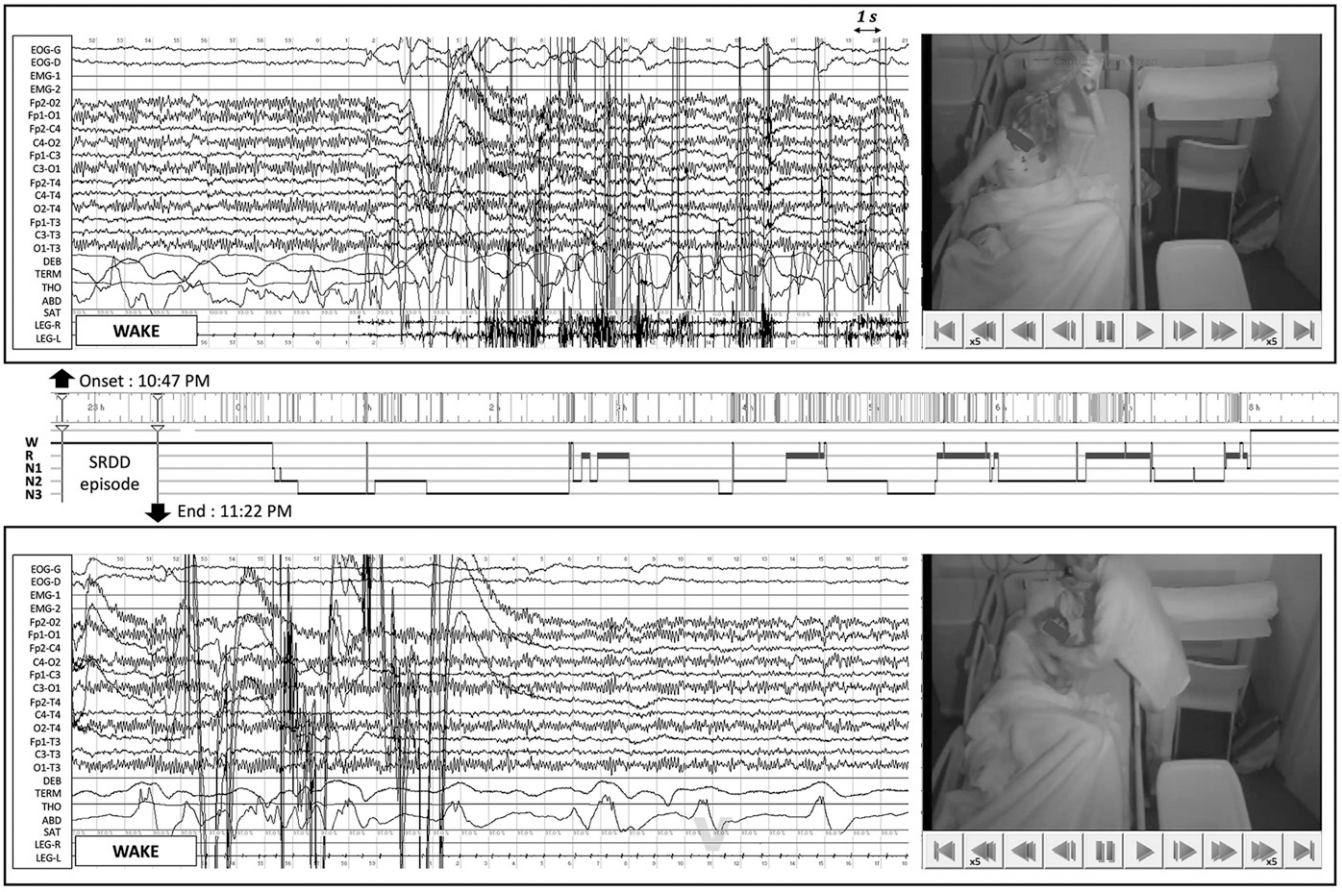
Case 3

A 16-year-old girl was referred in emergency to the sleep unit for episodes of nocturnal violent behaviors. Since the age of 5 she had infrequent but typical sleepwalking episodes. She also had a family history of sleepwalking. The episodes progressively disappeared, with the persistence of occasional confusional arousals. At the age of 12, prolonged and complex nocturnal behaviors suddenly started several months after she saw her father die at home from a myocardial infarct. She also developed at the same time typical symptoms of posttraumatic stress disorder. The episodes occurred on a weekly basis; they usually started 60–90 minutes after sleep onset and lasted 30–90 minutes. They were characterized by ambulation and violent behaviors toward the family members or her dog, but also with frequent self-inflicted injuries (biting herself, head punching, scarifications, and so on), and sometimes with urination or sexual vocalizations. Two episodes also occurred after an awakening from a nap, including one in a car with visual hallucinations (she saw her father walking along the road). Several episodes were videorecorded by smartphone and showed nonstereotyped, violent episodes accompanied with screams and unintelligible speech, confirming the clinical description retrieved from the family interviews. Her eyes were most often open, and none of the behaviors were suggestive of epileptic or psychogenic nonepileptic seizures. The vPSG recording did not highlight disrupted behaviors during sleep or wakefulness. The microstructure of the NREM sleep revealed a discrete N3 sleep fragmentation and persistent slow waves during N3 sleep interruptions, a pattern commonly found in patients with NREM sleep parasomnia.

Case 4

An 8-year-old girl was admitted for suspected sleepwalking episodes. The nocturnal behaviors systematically occurred 5 to 10 minutes after bedtime and were associated with complete amnesia. The episodes began a few months earlier, shortly after her sister returned to live in the family home. The frequency of the episodes gradually increased from a weekly to daily basis at the time of medical evaluation. The family reported dangerous, sometimes violent, behaviors. She frequently put herself in danger by climbing on household furniture or taking kitchen knives. Several videos captured by smartphones were examined, showing the patient walking with her eyes closed, hitting her brother with toys or behaving like a dog. The behaviors were nonstereotyped without abnormal movements or large hip movements as during nonepileptic seizures. Initially the episodes could be rapidly interrupted by her parents, but at the time of the medical evaluation episodes could last more than 1 hour. Once the episode was over, she would lie down in a bed in her parents' room. Sleep at night thereafter was continuous, with no reported nighttime awakenings. The vPSG study documented a long episode of 35 minutes which arose from electroencephalography-defined wakefulness a few minutes

Figure 1—Electroencephalogram results for case 4. SRDD = sleep-related dissociative disorders.



after she lay quietly in bed with eyes closed, prior to sleep onset. During this episode, she danced, tried to get over the bed rails, and did not respond to the nurses or her mother who tried to interact with her. Her eyes were mainly closed during the episode. The electroencephalogram showed no epileptic-type activity, and an alpha wakefulness rhythm was observed during most of the episode (**Figure 1**). Subsequent sleep was normal.

Case 5

A 44-year-old female presented with a long history of disturbing nighttime behaviors with shouting and insults. Episodes occurred several times a week, 60 to 90 minutes after bedtime. She did not recall any of the episodes. The police had been called by neighbors on several occasions, and she had to move three times. The characterization of the episodes was made difficult by her amnesia, as she also slept alone. The episodes began around the age of 20; she had no family or personal history of sleepwalking or night terrors as a child. The psychiatric examination found chronic depression and the presence of paranoid psychotic symptoms. She also reported several daytime manifestations that corresponded to dissociative amnesia disorder. She reported significant difficulty falling asleep with an anticipatory anxiety of the episodes. She also reported loud snoring and a complaint of attention problems, without excessive daytime sleepiness. During the vPSG the sleep latency was of 120 minutes, with a long awakening between 3:30 and 4:30 AM. No behavioral manifestations were observed during wakefulness.

Case 6

A 16-year-old adolescent, a migrant of Malian origin, had severe posttraumatic stress disorder and major depressive and anxiety disorders related to multiple psychological, physical, and sexual traumas that had occurred over the past year. He was referred for suspected atypical sleepwalking episodes that had been evolving since his arrival in France. During the nocturnal episodes, he often left the house and woke up sometimes several kilometers away. Several episodes were observed by the health care staff, with self-inflicted injuries and violent reactions toward others. On several occasions he grabbed knives or used a lighter. The episodes could last up to 2 hours, with partial to complete amnesia. He underwent a vPSG assessment under mirtazapine treatment. The general sleep architecture was normal, with a discrete fragmentation of N3 sleep without parasomniac episodes. A sudden awakening was recorded during REM sleep due to a violent nightmare. There were no

pathological levels of phasic and tonic electromyography activities during REM sleep. No behavioral episodes suggestive of SRDD were recorded.

Case 7

An 18-year-old woman was referred for amnestic nocturnal behaviors with self-inflicted lesions. She had no particular somatic history. She had a history of multiple traumas: She was bullied at school at the age of 9, then at the age of 12 she was physically and verbally abused by her father. She was followed up in child psychiatry from the age of 8 for emotional dysregulation, and a depressive episode was diagnosed at the age of 10. Since the age of 8 years she made several suicidal attempts, by hanging, then by phlebotomy, then by interrupted or aborted attempts at defenestration, self-immolation, and electrocution. She was recently hospitalized for 2 months in a psychiatric department for a depressive episode with psychotic symptoms. For the past 4 months she regularly woke up in the morning with burns and bumps on her face supposed to be self-inflicted during the night, without any recollection of it. While she was in the hospital with diazepam and cyamemazine at bedtime 10 episodes were noted by the medical team. The patient did not leave her room and did not make any noise; the episodes themselves were not observed, but lesions were noticed in the morning by the medical team. The psychiatric examination also revealed daily dissociative symptoms of depersonalization and derealization. She had no personal or family history of parasomnia. No behavioral manifestations were observed during the vPSG assessment. The sleep structure was normal, and there was no argument for REM or NREM sleep parasomnia.

Case 8

A 12-year-old girl had daily episodes of abnormal and amnestic behaviors that suddenly started a month before, with no potential triggering factor identified. The clinical manifestations were nonstereotyped, and they systematically occurred in relation with sleep periods. Several episodes were captured by smartphone video and showed violent behaviors (slapping, rocking movements, smashing toys) associated with vocalizations (moaning, incoherent speech), with her eyes closed. She would become violent toward her parents when they tried to interrupt the episode. These episodes arose within the first minutes after bedtime, with rare episodes reported in the early morning. Several episodes might occur within a single night and lasted from 10–30 minutes. No dissociative symptoms were reported during the daytime, except during naps. The psychiatric examination was normal. She had a prior history of infrequent but typical sleepwalking and sleep terrors. The vPSG demonstrated a long sleep latency and wake after sleep onset duration. A typical and brief confusional arousal was observed out of N3 sleep. Five nonstereotyped behavioral episodes of durations of 17, 16, 8, 6, and 4 minutes, respectively, were recorded during electroencephalography-defined wakefulness without epileptic-type activity. The first episode occurred 3 minutes after lights-off. Four episodes were recorded before sleep onset and the last one was at 6 AM, 70 minutes after the final awakening. Episodes were violent and consisted of incoherent speech, shaking the bedrails, rocking movements, or pulling out the alarm bell. Her eyes remained closed during the entire episodes.

Low doses of clonazepam were prescribed (cases 1, 2, 3, and 5) and were effective in 2 cases. All adolescent and adult patients were referred for psychiatric evaluation and management. Reassurance and behavioral advice were given to the family of the 2 children (case 4 and case 8).

The main polysomnographic characteristics of the 8 cases were unremarkable, except for a low sleep efficiency in case 1 and case 8 and a mild obstructive sleep apnea syndrome in case 5. One patient had both NREM sleep parasomnia and SRDD episodes during the vPSG. Fragmentation in N3 sleep was in the normal range, except for 2 patients (including 1 with a past history of DOA). None of the patients had pathological levels of tonic or phasic electromyography activity during REM sleep, and one had a sudden arousal from REM sleep associated with a nightmare episode. Three patients exhibited abnormal behaviors during sustained wakefulness (case 4 and case 8 with typical SRDD episodes and case 2 with a mild episode). Finally, the diagnosis of SRDD was definite for case 4 and case 8, probable for case 2, and possible for the remaining 5 cases.

The main clinical and polysomnographic characteristics of the 8 patients are reported in **Table 1** (individual clinical and neurophysiological data are also provided in **Table S1** and **Table S2** in the supplemental material). Comparisons between the 8 new cases and the 18 published cases revealed no difference for clinical or neurophysiological characteristics and were then pooled for descriptive analysis (**Table 1**).

DISCUSSION

Prevalence and frequency

The prevalence of SRDDs is supposed to be low but is still unknown. Our series of 8 cases, matching the largest series reported to date, confirms that SRDD is a condition rarely seen in tertiary sleep disorders units, with around 1% of patients referred for disrupted nocturnal behaviors diagnosed with SRDDs. This frequency is lower than previously reported in a series of 100 consecutive patients presenting with histories of sleep-related injury to self and/or others, with 7 patients diagnosed with SRDD.[24] These values should be interpreted with caution and could be underestimated because of a possible recruitment bias in sleep disorders units. Indeed, the frequency of SRDD in psychiatric settings, especially in patients with dissociative disorders, could be higher. In a large multicentric clinical sample of 303 patients with dissociative identity disorder, frequent "sleepwalking experiences" were reported by 38.6%.[25] In another smaller but well-defined series of 29 consecutive patients with a diagnosis of dissociative disorder, 8 (27.5%) also presented with SRDD episodes.[19]

Clinical manifestations

All 26 patients had complex behaviors that emerged throughout the nighttime period, and more than half also had episodes during the daytime, with a partial or complete amnesia. Despite some behavioral and cognitive similarities between SRDDs and

parasomnias, we highlighted 4 clinical characteristics potentially useful for the differential diagnosis. First, in most cases (90.9%), SRDD episodes could last up to hours. In contrast, RBD episodes usually last a few seconds or minutes, and long NREM sleep parasomniac episodes of over an hour have only exceptionally been described.[26] Furthermore, prolonged episodes have also been rarely reported in the automatism-amnesia syndrome associated with GABAergic medications (Z-drugs or sodium oxybate).[27,28] Malingering and Munchausen syndrome by proxy are rare conditions that may mimic SRDD. These differential diagnoses need to be considered in cases of apparent sleep-related violent and prolonged episodes.[29] Similar findings were reported in psychogenic nonepileptic seizures (PNES), which are usually longer than epileptic seizures.[30,31] Second, the timing of occurrence of the nocturnal behaviors may also help the clinician in the diagnostic process. SRDD episodes occurred within minutes after bedtime in more than one-third of the cases. In contrast, NREM sleep parasomnia episodes occur usually after at least 30 minutes of sleep and RBD episodes are mainly observed in the second half of the night. Third, if injuries during DOA and RBD are frequent,[32,33] they are often accidental in contrast with SRDD cases, which are mainly self-inflicted, and manifestations of psychopathology. Such self-harm or pseudo-suicidal behaviors were exceptionally reported in NREM sleep parasomnia and never in RBD.[34] Fourth, we confirmed that daytime dissociative disorders are frequent in patients with SRDD. Although high levels of dissociative symptoms have been reported in patients with sleep disorders,[35] especially in patients with parasomnias[36,37] including DOA,[21,38] the presence of a daytime dissociative disorder would support the positive diagnosis of SRDD. All cases had episodes with at least 1 of the 4 characteristics (ie, long duration, occurrence within minutes around the bedtime period, self-inflicted injuries, and presence of dissociative symptoms during daytime).

Psychiatric examination

Our findings point to the importance of performing a detailed psychiatric interview in patients with suspected SRDD and searching for comorbid mental disorders and traumatic events and a past history of abuse as potential triggers of the episodes. At least 1 comorbid psychiatric disorder was diagnosed in 91.7% of the patients. These findings are in line with those obtained in dissociative disorders,[19,39,40] but contrast with the lower rates of psychiatric disorders found in DOA or RBD.[33,41–43] Of interest, similar discrepancies in the frequency of psychiatric comorbidities were found in epilepsy and PNES.[44] In regard to case 8, the convincing diagnosis of SRDD in this 12-year-old girl, without identified psychopathology or trauma, would serve as a "red flag" warranting in-depth psychiatric evaluation to pursue suspected trauma and abuse as the basis for her SRDD.

As previously reported,[4] posttraumatic stress disorder was diagnosed in more than half of patients. We also found a high frequency of borderline personality disorder, another condition that often occurs in the context of past history of physical, psychological, or sexual abuse, and with daytime manifestation of self-injurious behaviors. We confirmed that almost all reported cases of SRDD occurred in a context of past traumatic events or abuse, as reported in samples of patients with dissociative disorders[19,45,46] and PNES,[47] but not in typical NREM or REM sleep parasomnias.[21,33,41] However, authentic parasomnia behaviors may develop after traumatic experiences.[48,49]

Objective findings

Our PSG findings showed that SRDDs are rarely associated with disrupted sleep. An increase in waking dissociative symptoms had been reported in relation with sleep disruption and deprivation.[35] However, the potential role of sleep disruption or deprivation in triggering SRDD episodes could not be assessed.

The absence of spontaneous or provoked[9,15,16] parasomnia episodes and the lack of pathological PSG markers of parasomnia would bring evidence against parasomnias, which are the principal differential diagnoses. In clinical samples of patients with NREM sleep parasomnias, 50%–60% of patients displayed at least 1 parasomnia episode during a single night of vPSG assessment.[7,8] Assessment of N3 sleep fragmentation or REM sleep without atonia is a useful objective biomarker of NREM sleep parasomnia and RBD, respectively. We provided such quantitative measures in patients with SRDD with the slow-wave sleep fragmentation index and the slow mixed arousal index, 2 recently described diagnostic markers of DOA[7,8] in the normal range, except for 2 patients (including 1 with a past history of DOA). Also, none of the patients had pathological levels of tonic or phasic electromyography activity during REM sleep.[6] However, we highlighted that SRDD behaviors may co-occur with typical NREM sleep parasomnia episodes (case 8). Thus, the absence of current or past parasomnia should not be a requirement for the diagnosis of SRDD.

The diagnosis of SRDD may be best confirmed by recording clinically compatible SRDD episodes simultaneously on video and polysomnography and by finding the presence of episodes while awake before, during, and after the event. In such a situation, a positive diagnosis can be made with a high degree of certainty, as illustrated by cases 4 and 8. However, in some cases, less-severe or less-complex episodes that do not fully correspond to the clinically described symptoms may be recorded (see case 2). Accordingly, the diagnosis of SRDD could be provided, but with a lower level of certainty. As typical or mild dissociative episodes were observed during vPSG in only 3 among the 8 cases, homemade videos using smartphones or infrared cameras applied in the patient's bedroom could be a good option to record SRDD episodes in the sleeper's usual environment. This alternative of video recording may be useful to confirm the clinical features provided by the medical interview, as already suggested in the context of epilepsy or DOA.[50] Cases 3, 4, and 8 illustrated the potential usefulness of homemade video in the diagnostic process of SRDD, especially for case 3, who did not exhibit dissociative episodes during the vPSG assessment.

Nosological and diagnostic considerations for sleep medicine

All patients from our case series, together with the only published case series that also involved 8 cases of SRDD,[5] had been referred to sleep disorders centers on account of disruptive

nocturnal behaviors. Although considered a psychiatric disorder, SRDD are a diagnosis that also falls within the nosology of sleep medicine, and sleep clinicians who see these cases need diagnostic information for their proper management. In sleep medicine, the diagnosis of SRDD requires a careful clinical interview of the patient and witnesses by an experienced sleep expert and a systematic psychiatric examination. Accordingly, we propose new diagnostic criteria for SRDD (**Table 2**) following the recent conceptual framework developed to improve the homogeneity of future revisions of the ICSD.[51]

We propose standard criteria for the diagnosis of SRDD: "episode(s) of complex dissociative behaviors that emerge throughout a rest period" (criterion A) and with additional supportive features: episodes of long duration (typically more than 1 hour, although shorter episodes can occur, eg 15–30 minutes), occurrence while awake close to bedtime, self-inflicted injuries, or the presence of dissociative symptoms during daytime (criterion B). The episodes should be associated with a partial or complete loss of conscious awareness and with subsequent impaired recall (criterion C). A significant distress or functional impairment should be evidenced (criterion D). The behaviors should not be better explained by another sleep, medical, or psychiatric disorder; medication; or substance abuse. However, the presence of another sleep disorder should not automatically exclude the diagnosis, as SRDDs may co-occur with other parasomnias, as can typical PNES co-occur in patients with epileptic seizures.[52–54] Other differential diagnoses include other nocturnal manifestations of psychiatric origin (ie, nocturnal panic attacks, nocturnal eating syndrome), another medical disorder such as sleep-related hypermotor epilepsy, or medication- or substance-induced nocturnal confusion (criterion E).

Based on the information retrieved from the medical interview and those obtained from vPSG or homemade videos, we propose, as an option, to define three levels of diagnostic certainty, in line with the approach of the International League Against Epilepsy for the diagnosis of PNES.[55] The diagnosis of SRDD is *possible* when only based on the clinical evaluation and *probable* when typical SRDD episodes are documented by homemade videos and/or when the vPSG shows episodes that do not fully correspond with the clinically reported manifestations. Finally, the diagnosis can be considered as *definite* when the vPSG documents typical episodes of SRDD with sustained wakefulness before, during, and after the event (see footnotes to **Table 2**).

The 26 detailed cases met the proposed diagnostic criteria for SRDD. Among the 8 newly described cases, 2 had a diagnosis of definite SRDD (case 4 and case 8) and 2 had a diagnosis of probable SRDD (case 2 with a mild vPSG-documented episode and case 3 with several typical video-documented episodes); the 4 remaining cases with negative vPSG findings had a diagnosis of possible SRDD. Among the 18 published cases, 8 could be considered with a diagnosis of definite SRDD, 3 of probable SRDD, and 7 of possible SRDD.

Nosological and diagnostic considerations for psychiatry

Preliminary findings suggested that SRDDs may be not as rare as initially thought, especially in patients with dissociative disorders,[19,25] and probably go largely undiagnosed. We also

Table 2—Suggestions for new SRDD diagnostic criteria for the *International Classification of Sleep Disorders*.

Criteria	Definition
Clinical manifestation	A. Episode(s) of complex dissociative behaviors that emerge throughout a rest period. B. The presence of at least 1 of the following in association with the dissociative episodes: • Some episodes last more than 1 hour • Some episodes occur around bedtime while awake • Self-inflicted injuries during the episodes • The patient presents daytime dissociative symptoms C. There is a partial or complete loss of conscious awareness during the episode, with subsequent impaired recall
Distress/disability	D. The disturbances cause clinically significant distress or impairment in mental, physical, social, occupational, educational, or other important areas of functioning, as indicated by the report of at least 1 of the following: • Sleep resistance (eg, bedtime anxiety, fear of sleep, or subsequent episodes) • Daytime sleepiness, insomnia symptoms, and/or fatigue • Impaired occupational or educational function • Impaired interpersonal/social function
Exclusion	E. The behaviors are not better explained by another sleep disorder, mental disorder, other medical disorder, medication, or substance use.
Levels of certainty	
Possible: The diagnosis is based only on clinical interview (witness or self-report description) Probable: The diagnosis is based on clinical interview and: • Home video recordings demonstrate typical behavioral episode(s) consistent with the clinical characteristics retrieved from the patient's or witnesses' interview. • The video-polysomnography documents episode(s) that does not fully correspond to the clinical description retrieved from the clinical interview, with sustained wakefulness before, during, and after the event. Definite: The diagnosis is based on clinical interview and the video-polysomnography documents typical behavioral episode(s) with sustained wakefulness before, during, and after the event.	

SRDD = sleep-related dissociative disorders.

Table 3—Suggestions for the "other specified dissociative disorder" section of the *Diagnostic and Statistical Manual of Mental Disorders.*

Sleep-related dissociative disorder	This condition is characterized by dissociative behaviors that emerge throughout a sleep period during wakefulness, either at the transition from wakefulness to sleep or within several minutes after an awakening from sleep. The nocturnal dissociative symptoms often occur with corresponding daytime episodes of disturbed behavior. In patients with nocturnal episodes only, a transdisciplinary diagnostic process is suitable with experienced sleep specialists to differentiate SRDD from non-rapid eye movement sleep arousal disorders, rapid eye movement sleep behavior disorder, and nightmare disorder.

SRDD = sleep-related dissociative disorders.

highlighted that 27.3% of patients with SRDD did not present with diurnal dissociative manifestations, a finding that favors recognizing SRDD as a variant of dissociative disorder and not just a nocturnal expression of diurnal dissociative manifestations. In the early 2000s, the same type of considerations led to the recognition of the night eating syndrome as a variant of eating disorders. This condition was included as an example of presentations of the "Other Specified Feeding or Eating Disorders" in the DSM.[4] To facilitate the identification of SRDD by psychiatrists and to ensure a better internal coherence for the DSM, we suggest including SRDD as one of the examples of presentations of the "Other Specified Dissociative Disorders." A short paragraph describing the essential features of SRDD is proposed (**Table 3**).

Limitations

Our work highlighted a certain amount of heterogeneity in the clinical and neurophysiological characteristics of SRDD. Although the 8 newly reported cases appeared similar to those already reported in the literature, the small sample size lacked sufficient power to allow for valid statistical comparisons. More cases need to be reported to confirm the validity of the proposed diagnostic criteria.

CONCLUSIONS

We reported here 8 new cases of SRDD with a full comprehensive sleep, psychiatric, and vPSG assessment and systematically extracted the main clinical and neurophysiological characteristics of 18 other cases reported in the literature. As recently emphasized in a systematic analysis of ICSD-3 diagnostic criteria,[51] an aim of the ICSD-3 is "to define the domain of a given discipline, a factor of particular importance for fields such as sleep medicine which cut across many related specialities."[3] SRDDs should ideally be included in the nosologies of both sleep medicine and psychiatry. We propose updated diagnostic criteria for the fields of sleep medicine and psychiatry, to be eventually included in the next revisions of the ICSD and the DSM to better diagnose these patients, and to further understand the pathophysiology and open new avenues for treatment.

ABBREVIATIONS

DOA, disorders of arousal
DSM, *Diagnostic and Statistical Manual of Mental Disorders*
ICSD, *International Classification of Sleep Disorders*
NREM, non-rapid eye movement
PNES, psychogenic nonepileptic seizures
RBD, REM sleep behavior disorder
REM, rapid eye movement
SRDD, sleep-related dissociative disorders
vPSG, video-polysomnography

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