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RBD: Historical Perspective

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Carlos H. Schenck

1.1 Introduction

The historical perspective on RBD encompasses (1) the formal discovery of RBD in 1986 and the early clinical RBD milestones, (2) the clinical historical background from 1966 to 1985, (3) the first experimental animal model of RBD from 1965, and (4) RBD described in classic literature and film.

1.2 Formal Discovery of RBD

The first report on RBD was published in 1986 in *Sleep*: "Chronic behavioral disorders of human REM sleep: a new category of parasomnia" [1]. The abstract read as follows:

> Four men, aged 67–72 years, had 4-month to 6-year histories of injuring themselves or their spouses with aggressive behaviors during sleep, often during attempted dream enactment. A 60-year-old woman had disruptive though nonviolent sleep and dream behaviors. Polysomnography did not detect seizures but did document REM sleep pathology with variable loss of chin atonia, extraordinarily increased limb-twitch activity, and increased REM ocular activity and density. A broad range of REM sleep behaviors was recorded on videotape, including stereotypical hand motions, reaching and searching gestures, punches, kicks, and verified dream movements. Stage 3–4 slow wave sleep was elevated for age in all patients. NREM sleep was devoid of harmful behaviors, although three men had periodic myoclonus. There was no associated psychiatric disorder, whereas serious neurologic disorder was closely associated in four cases: olivo-ponto-cerebellar degeneration, Guillain-Barré syndrome, subarachnoid hemorrhage, and an atypical dementia. Two patients had immediate and lasting sleep behavioral suppression induced by clonazepam, and another

C. H. Schenck
Minnesota Regional Sleep Disorders Center, and Departments of Psychiatry,
Hennepin County Medical Center and University of Minnesota Medical School,
Minneapolis, MN, USA
e-mail: schen010@umn.edu

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patient had the same response with desipramine. All instances of drug discontinuation prompted immediate relapse. In four cases there was associated dream hyperactivity, which resolved with behavioral control. These REM sleep neurobehavioral disorders constitute another category of parasomnia, replicate findings from 21 years ago in cats receiving pontine tegmental lesions, and offer additional perspectives on human behavior, neurophysiology, pharmacology, and dream phenomenology.

Despite the variable loss of the customary, generalized muscle paralysis of REM sleep ("REM atonia"), all other major features of REM sleep remain intact in RBD, such as REM sleep latency, REM sleep percent of total sleep time, number of REM sleep periods, and REM/NREM sleep cycling.

The discovery of RBD was described in the book *Paradox Lost: Midnight in the Battleground of Sleep and Dreams* [2]. I had just become a member of the Minnesota Regional Sleep Disorders Center. On my first day evaluating patients, September 11, 1982, the second patient on my schedule was a Mr. Donald Dorff, who complained of "physical moving dreams" and "violent moving nightmares." As described by Michael Long at the start of his story in the December 1987 issue of *National Geographic* magazine ("What Is This Thing Called Sleep?"), "The crowd roared as running back Donald Dorff, age 67, took the pitch from his quarterback and accelerated smoothly across the artificial turf. As Dorff braked and pivoted to cut back over tackle, a huge defensive lineman loomed in his path. One hundred twenty pounds of pluck, Dorff did not hesitate. But let the retired grocery merchandiser from Golden Valley, Minnesota, tell it: 'There was a 280-pound tackle waiting for me, so I decided to give him my shoulder. When I came to, I was on the floor in my bedroom. I had smashed into the dresser and knocked everything off it and broke the mirror and just made one heck of a mess. It was 1:30 a.m.'"

Mr. Dorff had been acting out his dreams for several years, and after his doctor had found nothing medically wrong with him, nor had a psychiatrist found anything mentally wrong, he was referred to our sleep center. On September 16, 1982, 5 days after I had evaluated Mr. Dorff, he was studied in our sleep laboratory. During each of his apparent REM sleep periods, there were many jerks and twitches and sometimes more elaborate and violent behaviors that correlated with the dreams that he reported when he woke up. However, confirmation that these were truly REM sleep events came at daybreak. As I wrote, "The next morning, in reviewing Don Dorff's polygraphic sleep tracings and videotaped behaviors, Mark Mahowald, M.D. and Andrea Patterson, R.PSGT & R.EEGT, our sleep center director and our sleep laboratory manager and chief technologist, repeatedly challenged each other, going back and forth in playing 'Devil's advocate.' The question was whether Don's violent dream-enacting activity had occurred during REM sleep… So kudos to Mark and Andrea, who jointly discovered the polygraphic foundation of REM sleep behavior disorder-RBD" [2].

1.3 Early Clinical RBD Milestones

RBD was named in our second report published in *JAMA* in 1987 [3], and among the ten patients in the original series, five had diverse neurologic disorders etiologically linked with RBD, and five were idiopathic [1, 3]. As a larger group of

idiopathic RBD (iRBD) patients was gathered and followed longitudinally at our center, a surprisingly strong and specific association with eventual parkinsonism and dementia became apparent, with our first report published in 1996 (and extended to 2013) [4, 5]. Other early RBD clinical milestones from our center include RBD in the differential diagnosis of sleep-related injury [6]; forensic aspects of RBD [7], later updated to include "parasomnia pseudo-suicide" [8]); status dissociatus (with emergence of RBD behaviors during indeterminate EEG/ Polysomnographic (PSG) states) [9]; RBD affecting patients in intensive care units [10]; antidepressant medication-induced RBD [11]; RBD associated with narcolepsy-cataplexy [12]; association of RBD with specific HLA haplotypes [13]; and the parasomnia overlap disorder (RBD with NREM parasomnias) [14]. RBD has been included in each edition of the International Classification of Sleep Disorders, including the current 3rd edition [15]. A 16-year perspective on RBD was published in *Sleep* for its silver anniversary issue in 2002 [16]. Furthermore, the jerks, twitches, movements, and behaviors of RBD may represent the pathological reemergence of primordial ontogenetic and phylogenetic motor activity patterns [17].

The August 2013 issue of *Sleep Medicine* was devoted to RBD, with 18 peer-reviewed papers covering basic and clinical sciences and both original research and review articles. The Preface described how "RBD is situated at a strategic and busy crossroads of sleep medicine and the neurosciences. RBD offers great breadth and depth of research opportunities, including extensive inter-disciplinary and multinational research opportunities" [18]. The Preface to this book expands on these statements by listing and commenting on the large number of diverse research areas intersecting with RBD that provide many future interdisciplinary research opportunities. The "RBD odyssey" exemplifies the strong cross-linkage between the RBD basic and clinical sciences [19].

Finally, in 1987 a documentary film on RBD was produced at our sleep center, "Rapid Eye Movement Sleep Behavior Disorder" [20]. This film is contained in the archives at The National Library of Medicine, Department of Health and Human Services, Public Health Service, National Institute of Health (NIH), Bethesda, Maryland.

1.4 Clinical Historical Background of RBD: 1966–1985

Various PSG and clinical aspects of correlates of chronic and acute human RBD (as we now call it) were described since 1966 by investigators from Japan, Europe, and North America, almost exclusively in neurologic and drug intoxication/withdrawal settings, as reviewed [1, 16, 21, 22], and as discussed in Chap. 12. Two groups of pioneering investigators should especially be recognized, as reviewed [16]: (1) Passouant et al. from France in 1972 first identified a dissociated state of REM sleep with tonic muscle activity induced by tricyclic antidepressant medication. (2) Tachibana et al. from Japan in 1975 named "stage 1-REM sleep" as a peculiar sleep stage characterized by muscle tone during an REM sleep-like state that emerged during acute psychoses related to alcohol and meprobamate abuse [23]. Also, clomipramine therapy of cataplexy in a group of patients with narcolepsy commonly

induced REM without atonia (RWA) in a 1976 study [24]. Elements of both acute and chronic RBD manifesting with "oneirism" were represented in the early literature, along with isolated RWA: delirium tremens (DTs) and other sedative and narcotic withdrawal states, anticholinergic use, spinocerebellar and other brainstem neurodegenerative disorders, and brainstem tumor [25] . The "REM rebound and REM intrusion" theories were proposed and discussed in many of these early reports. Finally, the 1986 report in *Sleep* firmly established that RBD is a distinct parasomnia that occurs during unequivocal REM sleep and which can be either idiopathic or symptomatic of a neurologic disorder [1]. PSG monitoring of these patients established that RBD did not emerge from a "stage-1 REM sleep" that was distinct from REM sleep, nor did RBD emerge from a poorly defined variant of REM sleep, nor from an unknown or "peculiar" stage of sleep, nor during "delirious" awakenings from sleep—all of which had been mentioned in the prior literature.

1.5 Experimental Animal Model of RBD

An experimental animal model of RBD was first reported in 1965 by Jouvet and Delorme from Lyons [16], with subsequent work on the model by Morrison and colleagues at the University of Pennsylvania beginning in the early 1970s [16]. Lesions in the peri-locus ceruleus area released a spectrum of "oneiric" behaviors during REM sleep (also called paradoxical sleep). These oneiric behaviors in cats closely match the repertoire of RBD behaviors in humans. Chapters 42 and 43 discuss the animal models of RBD in cats, rats, and mice. A therapeutic animal-human circle is completed with RBD. There is the historical progression from an experimental animal model of RBD shedding light on human RBD, which in turn has encouraged better recognition and management of RBD affecting cats and dogs presenting to veterinary clinics with violent sleep behaviors [16, 26].

1.6 RBD Described in Classic Literature and Film

Miguel de Cervantes described RBD in *Don Quixote* more than 400 years ago, in 1605: "He was thrusting his sword in all directions, speaking out loud as if he were actually fighting a giant. And the strange thing was that he did not have his eyes open, because he was asleep and dreaming that he was battling the giant… He had stabbed the wine skins so many times, believing that he was stabbing the giant, that the entire room was filled with wine" (Cap. XXXV *Aventura De Los Cueros De Vino*). Furthermore, there is strong suggestive evidence from a careful reading of *Don Quixote* that he also suffered from dementia with Lewy bodies (DLB) with fluctuating cognitive decline, complex visual and auditory hallucinations, and paranoid delusions [27]. (Chap. 6 discusses the strong link of RBD with DLB). Finally, the eighteenth-century philosopher Immanuel Kant may have suffered from combined DLB-RBD as manifestations of his 8-year terminal neurological illness [28].

RBD was depicted in Disney animated films long before the formal identification of RBD in humans in 1986 [29]. In *Cinderella* (1950), a dog had nightmares with dream enactment, and three additional dogs with presumed RBD appeared in *Lady and the Tramp* (1955), *The Fox and the Hound* (1981), and in the short film *Pluto's Judgment Day* (1935). These dogs were elderly males who would pant, whine, snuffle, howl, laugh, paddle, kick, and propel themselves while dreaming that they were chasing someone or running away. Moreover, in *Lady and the Tramp*, the dog was also losing his sense of smell and his memory, two prominent associated features of human RBD as an evolving neurodegenerative disorder. The Disney screenwriters were astute observers of sleep and its disorders, including RBD.

Conclusion

RBD is an "experiment of nature" in which knowledge from the study of motor-behavioral dyscontrol during REM sleep, with dream enactment, has cast a broad and powerful light on a multitude of CNS disturbances, their evolution, and their comorbidities. RBD has also cast light on the pervasive phenomenon of state dissociation [9, 30–33], as discussed in Chap. 28. The expanding and deepening knowledge on RBD is well reflected in the 45 chapters contained in this book.

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