

# Case Study: Sleep and Aggressive Behavior in a Blind, Retarded Adolescent. A Concomitant Schedule Disorder?

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## ABSTRACT

Blind people are prone to suffer from sleep-wake schedule disorders. This report describes 2 months of monitoring of sleep patterns and aggressive behaviors in a totally blind, severely retarded adolescent boy, hospitalized in a psychiatric hospital. The documented sleep-wake patterns seem to portray a sleep-wake schedule disorder with a monthly periodicity. Aggressive behaviors seem to echo the same periodicity, suggesting that a common or linked biobehavioral timing mechanism may underlie both sleep and episodic aggressive outbursts. The need to consider sleep schedule disorders as a primary process underlying some psychopathological disorders, and the related risks of misdiagnosis and mistreatment, are highlighted. *J. Am. Acad. Child Adolesc. Psychiatry*, 1995, 34, 6:820-824. **Key Words:** sleep, schedule disorder, aggressive behavior, blindness, retardation.

The light-dark cycle functions as a major regulator of the circadian sleep-wake rhythm. A growing body of evidence suggests that the neurohormone melatonin is directly controlled by the light-dark cycle. Melatonin is a pineal hormone released during hours of darkness and is considered to be a potent synchronizer (zeitgeber = timegiver) of circadian rhythms in animals and humans (for reviews on the role of melatonin in circadian regulation, see Armstrong, 1989; Reiter, 1986; Wurtman et al., 1991). It has been shown in humans that administration of melatonin results in sleepiness and that melatonin secretion is suppressed by light. In addition, disturbances in melatonin secretion have been associated with sleep disturbances, particularly in blind people (Arendt et al., 1988; Lewy and Newsome, 1983; Sack et al., 1987; Tzischinsky et al., 1991, 1992).

Research studies suggest that some blind people manifest a free-running sleep-wake rhythm in their natural environment (e.g., Miles et al., 1977; Sack et al., 1987). A free-running rhythm refers to a circadian periodicity that differs from the more usual 24-hour rhythm. It is usually longer and associated with delayed sleep onset and rise times from one day to next. Such patterns have been demonstrated in normal subjects in environments isolated from time cues (Moore-Ede et al., 1982).

The unique free-running rhythms seen in blind people are likely to influence their social adaptation and functioning, yet little is known about such interactions. The following case study demonstrates what appears to be a behavioral disorder associated with a sleep-wake schedule disorder.

## CASE PRESENTATION

P. was a 16-year-old, obese, large, Caucasian, blind male with profound mental retardation at the time he was referred to the Developmental Disabilities Unit of a children's psychiatric hospital. Chief complaints included aggressive outbursts characterized by self-injurious and other-injurious behaviors, generalized oppositional behaviors, and sleep problems.

## Developmental History

P. had a long history of medical problems starting during gestation. The pregnancy was planned. P.'s

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mother used a thyroid hormone and smoked cigarettes during her pregnancy. The length of the pregnancy was 30 weeks. Furosemide was administered during the week before delivery. Labor was complicated by abruptio placentae with emergency cesarean delivery. P.'s birth weight was 3 lb, 11 oz. Difficulty breathing and Rh incompatibility were noted and treated. An exchange transfusion was done, and P. used a respirator for 6 weeks and spent 2 months in the Neonatal Intensive Care Unit, where he suffered cardiac arrest twice. Subsequently, he developed retrolental fibroplasia with blindness secondary to positive pressure oxygen supplementation. Reportedly, P. disliked being touched and was unresponsive to common soothing practices. Speech and motor milestones appeared at appropriate ages.

P.'s early childhood was characterized by multiple family moves and changes in special education settings. P. was expelled from a number of schools because of aggressive and destructive behaviors. Since the age of 12 years, P. no longer was involved in formal special education because of the difficulties posed by his multiple handicaps and disordered behaviors.

During the final few years preceding the current hospitalization, P. had been cared for by his parents and older brother while living at home. His family remained devoted to his care but faced serious obstacles in managing his behavior. P. dictated a disordered daily routine for his family and responded with aggressive, destructive, and self-injurious behaviors to any of their limit-setting attempts. P.'s parents became unable to confront him and set limits. They also seemed anxiously attached to him and found it hard to accept prolonged separation. P. was socially isolated and had no opportunities to interact with other children or enhance his education in a full-time special education program.

At the time of P.'s admission to the Developmental Disabilities Unit, his sleep difficulties were seen as a major problem. His sleep-wake schedule was disorganized and there were severe difficulties in sleep initiation, often coupled with aggressive behaviors at bedtime until he fell asleep.

A number of drug trials had been conducted in the course of P.'s treatment prior to this admission. These trials involved, separately and in different combinations, haloperidol, thioridazine, chlorpromazine, phenytoin, fluphenazine, triazolam, and chloral hydrate; however,

these drugs had no positive effects on P.'s sleep-wake patterns or disordered behaviors.

After P. was admitted to the unit, the initial *DSM-III* diagnosis included stereotypy/habit disorder and parent/child problem on Axis I; profound mental retardation on Axis II; morbid obesity, blindness, frontal/temporal lobe atrophy, and sleep disorder on Axis III; chronic behavioral disturbances on Axis IV; and a Global Assessment of Functioning score of 10 on Axis V. Standard tests of cognitive functioning could not be completed because of P.'s multiple handicaps.

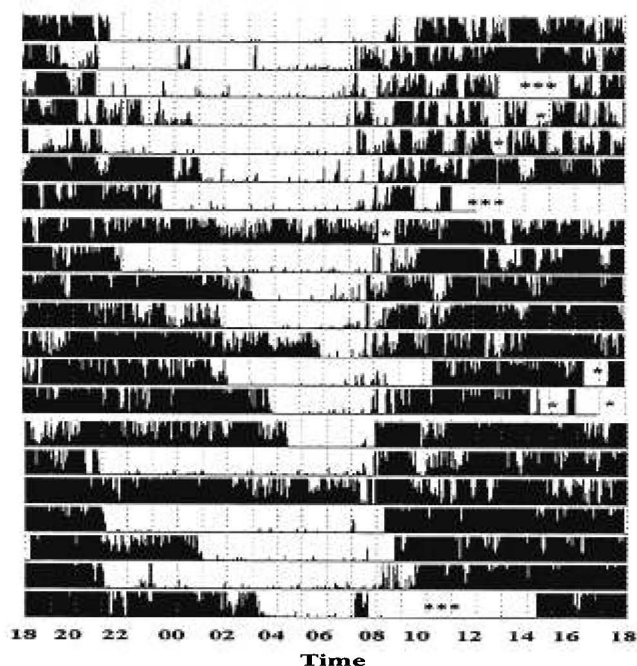
#### Assessment

To evaluate P.'s disorganized behavior including his sleep-wake schedule disorder, P. was gradually weaned from all medication. This was done in an attempt to sort out P.'s primary disorder from possible drug effects and side effects. After a washout period, a 58-day assessment period of P.'s sleep-wake patterns and aggressive behavior was completed.

During the 2 months of the study, P.'s aggressive behaviors were charted in detail by unit's nursing and milieu clinicians. An aggressive act was defined as a completed or attempted self-injurious or other-injurious behavior. P.'s aggressive acts included self-mutilation, pushing, hitting, kicking, biting, throwing objects, and destroying objects. Each event was documented as one episode regardless of its severity or consequences. Data were analyzed separately for morning hours (from 7:00 A.M. to 3:00 P.M.) and evening hours (from 3:00 P.M. to 11:00 P.M.).

P.'s sleep-wake patterns were monitored by direct observations. These observations resulted in daily records of P.'s daily bedtime, sleep onset time, rise time, night awakenings, and daytime naps. In addition, actigraphic monitoring objectively and independently assessed P.'s sleep-wake patterns to validate the reports of the nursing staff. The actigraph is a computerized activity monitor, worn on the wrist. It records limb activity continuously. Actigraphic recordings provide meaningful body activity measures over time that can be translated validly by sleep-scoring algorithms into sleep-wake measures (Cole et al., 1992; Sadeh et al., 1989, 1991, 1994).

P.'s daily sleep-wake patterns are illustrated in Figures 1 and 2A. Figure 1 portrays P.'s actigraphic records reflecting sleep-wake patterns over 3 weeks of monitoring. The staff observations of sleep duration were highly



**Fig. 1** Three nonconsecutive weeks of actigraphic recordings: black bars indicate each epoch's activity level. Dark areas represent waking periods with increased activity. White areas represent sleep periods with decreased activity. Periods of "actigraph off" are marked with stars.

correlated with actigraphic measures ( $r = .90$ ,  $p < .0001$ ). It appears that when P. was asleep, his sleep was sound and quiet with no interruptions by night awakenings. His mean actigraphic sleep efficiency (percent of actual sleep of total sleep period) was 97.1% and his average sleep duration was 5.94 hours. However, the circadian regulation was significantly disorganized. Nights characterized by a relatively reasonable amount of sleep alternated with sleepless nights or nights characterized by very short sleep times.

Figure 2A illustrates P.'s sleep duration during the 58 days of monitoring. Since P. was awakened each morning by staff at approximately the same time (7:00 A.M. to 9:00 A.M.), sleep duration was mainly determined by P.'s sleep onset time. The pattern seems to reflect periods of alternation between long sleep periods (e.g., 10 hours of sleep) and sleepless nights (days 13 to 20 and days 41 to 50). The periods are preceded by periods in which sleep becomes gradually shorter (days 1 to 8 and days 31 to 41), and followed by periods of relative stability (days 21 to 34 and days 53 to 58).

On average, P. engaged in 5.3 acts of aggression or attempted aggression per day during the 58 days of

monitoring, divided into 3.2 aggressive acts and 2.1 attempted acts on average. More aggressive acts were noted during the evening hours (mean = 3.36, SD = 0.95) compared to morning hours (mean = 1.93, SD = 0.70). Evening and morning levels of aggression were highly correlated on a day-by-day basis ( $r = .57$ ,  $p < .0001$ ).

The overall pattern of P.'s daily aggressive activity is illustrated in Figure 2B. There are two distinct periodic peaks separated by a 28-day interval.

Sleep and aggressive behavior data were correlated across days. However, there were only nonsignificant or modest correlations between specific daily aggressive activity and nocturnal sleep duration. For example, sleep duration was negatively correlated with the number of aggressive episodes in the evening hours ( $r = -.32$ ,  $p < .05$ ). Evenings with increased violence were followed by nights with shorter sleep. No significant correlations were found between sleep duration of the preceding night and aggressive behavior on the following day.

To assess possible cyclic relationships between sleep and aggressive behavior, we computed the nightly differences in sleep duration from the previous night and smoothed this measure and the measures of aggressive episodes with a 3-day moving average window. The smoothed daily number of aggressive episodes was highly correlated with the smoothed day-to-day variation in sleep duration ( $r = .63$ ,  $p < .0001$ ).

To examine the rhythm underlying both phenomena, an autocorrelation analysis was performed with the smoothed measures. Autocorrelation analysis peaked on the 28th day for both aggressive behavior ( $r = .95$ ,  $p < .0001$ ) and average sleep duration variability ( $r = .64$ ,  $p < .0005$ ), suggesting that a 28-day rhythm underlies these two phenomena.

The results of these analyses are visually obvious (Fig. 2). P.'s aggressive behavior peaked at the time that his sleep-wake system was in its most disorganized state, alternating between nights with extended sleep and sleepless nights. As soon as P.'s sleep-wake system stabilized, his aggressive behavior subsided.

## DISCUSSION

The tendency of normal individuals to develop free-running or otherwise disordered sleep-wake schedules under artificial conditions has been demonstrated in a

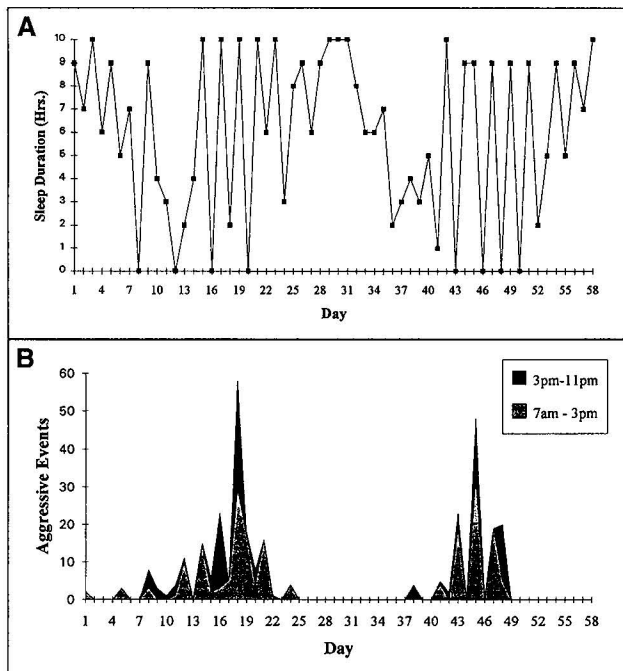


Fig. 2 Sleep duration (A) and number of aggressive episodes during morning and evening hours (B) as documented during a 58-day period.

number of studies (see Moore-Ede et al., 1982). To develop a free-running schedule, sighted people require total isolation from the light-dark cycle and other social time cues. Blind individuals are deprived of the major light-dark zeitgeber, and therefore they are more prone to develop schedule disorders (Arendt et al., 1988; Lewy and Newsome, 1983; Miles et al., 1977; Sack et al., 1987; Tzischinsky et al., 1991). In an uncontrolled environment, in which medications are used to control behavior and in which social expectations and demands are ineffective, the tendency for a regulated sleep-wake schedule to be replaced by a disorganized pattern such as the one manifested by P. seems obvious.

The reduction in sleep duration, seen in P.'s data prior to his most violent and disorganized periods, most likely reflects a tendency for later sleep onset times, possibly secondary to an underlying free-running sleep-wake rhythm. His free-running pattern is masked by the environmentally induced morning awakenings which, in turn, lead to a sleep debt and set up regulatory mechanisms that result in extreme alternation between nights with extended sleep and sleepless nights.

Although a monitoring period of 58 days is relatively short for assessing monthly periodicity, the recurrent patterns of both sleep-wake and aggressive behaviors

suggest that such periodicity exists. P.'s aggressive episodes were most likely influenced by other factors as well. It was observed that P.'s contacts with his family often resulted in an increased level of agitation, but since those visits were on a regular basis they could not account for the cyclic pattern seen in his behavior.

Sleep disorders are often a major component of psychiatric disorders (Dahl and Puig-Antich, 1990). The sleep disruptions may be considered as a symptom (or even one of the diagnostic criteria) of a psychiatric syndrome (Dahl and Puig-Antich, 1990). Such reasoning might potentially mislead professionals from considering the possibility that in some behavior disorders, symptoms may result from or be exacerbated by a major sleep disorder (Bergman, 1976; Dahl et al., 1991; Ford and Kamerow, 1989). P.'s behavior problem seems to reflect a major sleep-wake schedule disorder which had not been diagnosed and treated. Psychiatric and sleep-inducing medications have little effect on sleep-wake schedule disorders.

Advances in our understanding of schedule disorders and the role played by the neurohormone melatonin in the regulation of sleep-wake cycles have resulted in a new treatment for these disorders with demonstrated efficacy in blind people (Arendt et al., 1988; Folkard et al., 1990; Jan et al., 1994; Palm et al., 1991; Sack et al., 1990, 1991; Tzischinsky et al., 1992). Unfortunately, melatonin therapy, as an experimental treatment, was not permitted by P.'s parents, who were in continuous conflict with the staff about the need for P.'s prolonged hospitalization far away from home.

It is also important to emphasize that P., in addition to being blind, was severely retarded. Mental retardation and brain impairments have been linked to sleep-wake disorders (Jan et al., 1994; Okawa and Sasaki, 1987; Stores, 1992). Okawa et al. (1987), who studied blind children with moderate to severe mental retardation, concluded that mental retardation played an important role in these children's inability to perceive social cues related to normal sleep-wake rhythms. Similarly, our study suggests that sleep-wake schedule disorders in blind, retarded children should be considered in evaluations of their disordered behaviors while awake.

## REFERENCES

- Arendt J, Aldhous M, Wright J (1988), Synchronization of a disturbed sleep-wake cycle in a blind man by melatonin treatment. *Lancet* 1:77-773

- Armstrong SM (1989), Melatonin and circadian control in mammals. *Experientia* 45:932-938
- Bergman RL (1976), Treatment of childhood insomnia diagnosed as "hyperactivity." *J Behav Ther Exp Psychiatry* 7:199
- Cole RJ, Kripke DF, Gruen W, Mullaney DJ, Gillin JC (1992), Automatic sleep/wake identification from wrist actigraphy. *Sleep* 15:461-469
- Dahl RE, Pelham WE, Wison M (1991), The role of sleep disturbances in attention deficit disorder symptoms: a case study. *J Pediatr Psychol* 16:229-239
- Dahl RE, Puig-Antich J (1990), Sleep disturbances in child and adolescent psychiatric disorders. *Pediatrician* 17:32-37
- Folkard S, Arendt J, Aldhous M, Kennett H (1990), Melatonin stabilizes sleep onset time in a blind man without entrainment of cortisol or temperature rhythms. *Neurosci Lett* 113:193-198
- Ford DE, Kamerow DB (1989), Epidemiologic study of sleep disturbances and psychiatric disorders. *JAMA* 262:1479-1484
- Jan JE, Abrams IF, Freeman RD, Brown GM, Espezel H, Connolly MB (1994), Rapid cycling in severely multidisabled children: a form of bipolar affective disorder? *Pediatr Neurol* 10:34-39
- Lewy AJ, Newsome DA (1983), Different types of melatonin circadian secretory rhythms in some blind subjects. *J Clin Endocrinol Metab* 56:1103-1107
- Miles LEM, Raynal DM, Wilson MA (1977), Blind man living in normal society has circadian rhythm of 24.9 hours. *Science* 198:421-423
- Moore-Ede MC, Sulzman FM, Fuller CA (1982), *The Clocks That Time Us*. Cambridge, MA: Harvard University Press
- Okawa M, Nanami T, Wada S et al. (1987), Four congenitally blind children with circadian sleep-wake rhythm disorder. *Sleep* 10:101-110
- Okawa M, Sasaki H (1987), Sleep disorders in mentally retarded and brain-impaired children. In: *Sleep and Its Disorders in Children*, Guilleminault C, ed. New York: Raven Press
- Palm L, Blennow G, Wetterberg L (1991), Correction on non-24-hour sleep-wake cycle by melatonin in a blind retarded boy. *Ann Neurol* 29:336-339
- Reiter RJ (1986), *Pineal Melatonin Production: Photoperiodic and Hormonal Influences*. London: John Libby, pp 77-78
- Sack RL, Lewy AJ, Blood ML, Stevenson J, Keith D (1991), Melatonin administration to blind people: phase advances and entrainment. *J Biol Rhythms* 6:249-261
- Sack RL, Lewy AJ, Hoban TM (1987), Free-running melatonin rhythms in blind people: phase shifts with melatonin and triazolam administration. In: *Temporal Disorders in Human Oscillatory Systems*, Rensing L, van der Heiden U, Mackey MC, eds. Heidelberg: Springer-Verlag
- Sack RL, Stevenson J, Lewy AJ (1990), Entrainment of a previously free-running blind human with melatonin administration. *Sleep Res* 19:404
- Sadeh A, Alster J, Urbach D, Lavie P (1989), Actigraphically based automatic bedtime sleep-wake scoring: validity and clinical applications. *J Ambulatory Monitor* 2:209-216
- Sadeh A, Lavie P, Scher A, Tirosh E, Epstein R (1991), Actigraphic home-monitoring of sleep-disturbed and control infants and young children: a new method for pediatric assessment of sleep-wake patterns. *Pediatrics* 87:494-499
- Sadeh A, Sharkey K, Carskadon MA (1994), Activity-based sleep-wake identification: an empirical study of methodological issues. *Sleep* 17:201-207
- Stores G (1992), Annotation: sleep studies in children with a mental handicap. *J Child Psychol Psychiatry* 33:1303-1317
- Tzischinsky O, Pal I, Epstein R, Dagan Y, Lavie P (1992), The importance of timing in melatonin administration in a blind man. *J Pineal Res* 12:105-108
- Tzischinsky O, Skene D, Epstein R, Lavie P (1991), Circadian rhythms in 6-sulfatoxymelatonin and nocturnal sleep in blind children. *Chronobiol Int* 8:168-175
- Wurtman RJ, Lynch HJ, Sturner WQ (1991), Melatonin in humans: possible involvement in SIDS, and use in contraceptives. *Adv Pineal Res* 5:319-327