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Polysomnographic study of sleep in children and adolescents with developmental disabilities: A review

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Abstract

Aim: With this review, I aim to provide PSG study of sleep in people with developmental disabilities. **Method:** A literature was conducted and subject's characteristics, study design (EEG pattern in sleep cycle, integrated EEG, compressed spectral array, clinical EEG, ontogeny of sleep patterns, REM sleep latency, sleep spindles, REM activity, skin potential responses, skin temperature, heart rate, and respiration), and developmental disabilities were collected. The PSG recording was carried out during nocturnal sleep. **Results:** I identified 27 studies of 121 children with developmental disabilities (4 mo. to 6 yr.). They, moreover, included 79 retarded people (6 mo. to 20 yr.). They showed some abnormal sleep EEG patterns, using by the integrated EEG and the compressed spectral array. Their ontogeny of sleep patterns was similar to normal peoples. The distribution of their REM sleep latency was similar to that obtained by other authors from normal infants under 3 months of age. In their sleep spindles, the number of spindles longer than 0.4 sec may serve as a useful indicator of abnormality. In their REM activity, developmental psychomotor scores were correlated with % of REM20. In their skin potential response, type B and C were observed more frequently the subjects with low DQ and abnormal clinical EEG. Moreover, their skin temperature, heart rate and respiration were reported. **Interpretation:** Notwithstanding varying etiologies and varying degrees of retardation of these studies, many findings were presented. The significance of PSG recorded all night should be noticed, at least, in the people with developmental disabilities.

Key word : polysomnographic study, sleep, children and adolescents,
developmental disabilities, a review

I Introduction:

Little is known of the characteristic of sleep measurements in people with developmental disabilities. Polysomnography (PSG) in healthy children has been studied¹²⁾, but there are few studies on PSG in children with developmental disability. In most studies, the subjects

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were adults with developmental disabilities. In these adults several researchers have found an association between various sleep parameters and developmental level. For example, mentally retarded Down syndrome subjects presented a reduction of rapid eye movement (REM) sleep percentage and R index (number of high frequency REMs against number of low frequency REMs) and this was positively correlated to a low IQ⁶⁾. REM rates and REM percentage were reduced in people with mental retardation in proportion to the magnitude of their mental retardation⁸⁾.

Young people with developmental disabilities are recently reported to have frequent and persistent poor sleep than their healthy peers^{3), 5), 10), 18), 51), 51)}. The prevalence of poor sleep among individuals with intellectual disabilities has been reported to be as high as 85%⁷⁾, and the risk of poor sleep has been reported to be even higher in children suffering global injury⁵⁵⁾. These studies have been by the following. About 41.1% of the studies used a mixture of questionnaire, log, observation, and objective measures with while 20% used a questionnaire or logs and diaries, 18.9% applied merely observation, and 18.9% exclusively used an objective measure such as PSG⁵³⁾. The PSG study was a few. Because, as use of the criterion standard measure of sleep (i.e. PSG) remains a challenge in children with developmental disabilities because electrodes and the clinical setting are often uncomfortable and are not well tolerated, the use of more objective methods is advocated. However, tips on the use of PSG to investigate sleep in populations with developmental disabilities have been published recently^{19), 51)}. It is likely that alternatives to scoring overnight sleep studies are also needed, and examples of this have been published^{9), 17)}. In population with developmental disabilities, in particular, matching the problem, the solution, and the needs of all the people involved is essential⁵³⁾.

To summarize, with this review I aim to provide PSG study of sleep in children and adolescents with developmental disabilities.

II Method

1 Materials

A literature search was conducted using the electronic databases EDS. The publications of Shibagaki et al were, moreover, selected.

The definition of developmental disabilities is primarily a legislative and legal definition, with several criteria stipulated by institutes, organizations, or associations (e.g. the American and Japanese Association on Intellectual and Developmental Disabilities). In general, the criteria for our literature search were a mental or physical impairment or combination of the two; disabilities present before adulthood and expected to continue throughout the person's life; and the need for lifelong support and for educational services.

They were 57 girls and 64 boys, whose ages ranged from 4 mo. to 6 yr., were selected for the study: 4 to 11 mo. (n=50), 1 yr. (n=34), 2 yr. (n=12), 3 yr. (n=10), 4 yr. (n=8), 5 yr. (n=3) and

6 yr. (n=4). They, moreover, included 79 retarded children and adolescents aged 6 months to 20 years: 6 to 11 mo. (n=19), 1 to 1.11 yr. (n=17), 2 to 2.11 yr. (n=11), 3.1 to 8 yr. (n=13), 9.9 to 12.5 yr. (n=4), 14.1 to 16.2 yr. (n=8), and 19.3 to 20.10 (n=6). All subjects were inpatients of the Central Hospital, Aichi Prefectural Colony. Subjects for whom doctors made the diagnosis of etiologically known and unknown developmental disabilities were chosen. Seventy-five subjects had known etiology such as congenital cerebral malformation (n=15) and chromosomal abnormality (n=8). The remaining 46 subjects had unknown etiologies, associated with cerebral palsy (n=19), for example. The subjects were referred with introduction from another hospital. They had a regular checkup. They were wards of the state separated from their families. They were chosen to be free medication and no epilepsy. They were the products of uncomplicated pregnancies with uneventful labors and deliveries. They had no seizure history and no behavioral disabilities. They were only patients. This sample did not represent the general population for developmentally disabled children and adolescents.

A developmental disability was described in the search as mental retardation, intellectual disability, mental deficiency. Sleep was described as generally as possible to allow the inclusion of sleep quantity (e.g. duration, architecture), quality (e.g. problems or disorders), and regularity (e.g. bedtime, circadian rhythm, sleep cycle). No studies were included if an intervention (or treatment, therapy, management) on sleep problem was described.

2 Procedure

The PSG recording (electroencephalogram (EEG), electro-oculogram (EOG), electrocardiogram, respiration, skin temperature, skin potential response (SPR), and electromyogram (EMG)) was carried out in the subject' room (temperature, 25-27°C, relative humidity, 50-70%) after a conventional meal until spontaneous awakening the next morning. EEG was begun before the subjects went to sleep. The experimenter and polygraph machine were located in the room next to the subject's bed. No sedatives were used. The subject had experienced clinical EEG recordings two or three times during the daytime before the EEG recording during sleep in these studies. So measures were apparently taken to prevent novelty effects of apparatus. All subjects had been free of medication, which might affect EEG during sleep for at least 1 mo. before the recording.

III Results

Twenty-seven studies were reviewed. The earliest report found was from 1977.

1 EEG patterns in sleep cycle

The sleep EEG in healthy infants and children has been classified into several sleep EEG patterns (6 stages). Sleep EEG patterns in 43 mentally retarded children (from 4 months to 8 years of age) were studied throughout nocturnal sleep and following results were obtained⁴⁶⁾. (1) Twenty-two cases evidenced normal sleep patterns that could be classified

into 6 stages. (2) the 21 other cases showed some abnormal sleep EEG patterns as followed: (a) absence of sleep spindle (n=18) included cases of high voltage fast activity (n=2) and cases of low voltage activity throughout nocturnal sleep (n=3); (b) indistinguishable stages 1, 2, 3 and 4 because of little change in delta and theta activities (n=1), extreme spindle-like pattern (n=1), absence of REM sleep (n=1). (3) The severely mentally retarded children under 18 months had definitely decreased spindle activity in comparison to the values obtained by other studies from normal children. (4) The majority of the abnormal sleep EEG patterns occurred during light sleep. (5) A significant decrease in Developmental Quotient (DQ) was found in children with abnormal sleep patterns throughout Non-REM (NREM) sleep as compared with those in whom abnormal EEGs occurred only during stages 1 and 2. (6) It was concluded that the EEG recorded in nocturnal sleep may serve as a useful indicator of abnormality in mentally retarded children.

Sleep EEG patterns in 23 mentally retarded children with cerebral palsy (CP) (from 4 months to 5 years of age) and 39 reference mentally retarded children of no abnormality with the exception of psychomotor retardation (from 4 months to 12 years of age) were studied throughout nocturnal sleep, and the following results were obtained³⁵⁾. (1) Eleven CP cases and 9 reference cases showed normal sleep patterns that could be classified into 6 stages. (2) Twelve other cases in CP and 9 reference cases showed some abnormal EEG patterns. (3) Short sleep and long waking times during the night were noted in 4 out of CP subjects. (4) A long-term rhythm was found in the form of a fluctuation in integrated delta values through the night in 18 out of 23 CP subjects and in all reference subjects. However, such a periodic rhythmicity was not observed in the remaining 5 CP subjects. (5) A significant decrease of DQ was found in CP subjects with all or 1~4 stages indistinguishable, short sleep and long wakefulness and no delta rhythmicity in integrated records as compared to those with all the normal stages.

The PSG sleep studies were conducted with 10 mentally retarded infants (2 months to 4 years of age) suffering from hydrocephaly³⁶⁾. In two cases, it was not possible to distinguish between EEG patterns of wakefulness, sleep stage 1, sleep stage 2 and REM sleep because of abnormal high voltage fast activity during each of these periods. Spindles were observed in the one case but not in the other case. These four stages could, however, be differentiated on the basis of EOG, respiration and submental EMG patterns. One case each of two different etiologies exhibited no spindles. In the remaining 6 cases, the EEG patterns characteristic of the sleep-wakefulness cycle were observed. In one of these cases, extreme spindles were evident. A periodic rhythmicity in the integrated delta levels was found in all cases.

Sleep EEG patterns in 17 infants with cerebral malformations (4 months to 4 years of age) were studied throughout nocturnal sleep and the following results were obtained³⁷⁾. Seven cases evidenced normal sleep wakefulness EEG patterns that could be classified into 6 stages. Ten cases showed abnormal sleep EEG patterns as follows; absence of sleep spindles (n=7) which included cases of absence of EEG patterns characteristic of wakefulness, NREM

sleep and REM sleep (n=5), no characteristic EEG patterns of stages 1~4 (n=1) and stages W, 1, 2 and REM (n=1) and remaining cases with absence of spindles (n=1), and spindles with an extremely low incidence (n=2). Short sleep and long awaking times, and no delta rhythmicity during the night, were noted in 5 out of 17 subjects. A significant decrease of DQ was found in subjects with indistinguishable stages including W, 1, 2 and REM, as compared with patients whose stages were all distinguishable.

2 Integrated EEG

It were accepted that high voltage delta and spindle are the norm in slow wave sleep and light sleep in normal infants and children.

The delta and spindle components in the integrated EEG for 121 infants with developmental disabilities (from 4 mo. to 6 yr. of age: 57 girls and 64 boys) were studied throughout nocturnal sleep^{25), 29), 30), 33), 25)}. In 80 (66.1%, Group A) of 121 subjects, periodic changes of delta and spindle rhythm powers noted in measurement of EEG made during sleep. In 28 (23.1%, Group B), delta but not spindle rhythm power found, and in the remaining 13 subjects (10.7%, Group C) neither delta nor spindle rhythm powers were found throughout measurement of EEG made during sleep. The Tsumori-Inage Questionnaire for Infants was administered to the parents to estimate subject's behavioral developmental level as DQs. Significantly lower mean DQ were found for Group B and C than Group A. These findings suggested that the presence or absence of delta and spindle rhythm powers in EEG measurements made during nocturnal sleep could be taken as an index of the severity of developmental disorders in infants with developmental disabilities.

3 Compressed spectral array

It were accepted that high voltage delta and spindle are the norm in slow wave sleep and light sleep in normal infants and children.

The correlation between the delta and spindle components in compressed spectral array⁹⁾ in 96 mentally retarded children (from 3 months to 12 years of age) was studied throughout nocturnal sleep^{42), 43)}. In 57 (59.4%, Group A) of 96 subjects, periodic changes of delta and spindle rhythm were noted in sleep EEG. In 29 subjects (30.2%, Group B), delta but not spindle rhythm were found, and in the remaining 10 subjects (10.4%, Group C) neither delta nor spindle rhythm powers were found throughout nocturnal sleep. A significant increase in abnormal clinical EEG was found in Groups B and C as compared with Group A. A significant decrease in the DQ was found in Groups B and C as compared with Group A. A significant decrease in DQ was also found in Group C as compared with Group B.

The correlation between the delta and spindle components in compressed spectral array in 21 mentally retarded children with cerebral palsy (from 4 months to 5 years of age) and 32 reference mentally retarded children of no abnormality with the exception of psychomotor retardation (from 4 months to 12 years of age) were studied throughout nocturnal sleep³⁹⁾. In 11 (52%) out of 21 CP and 27 (84%) out of 32 reference cases, periodic changes of delta and spindle rhythm powers were noted in sleep EEG (Group A). In 5 (24%)

other cases in CP and 5 (16%) reference cases delta but not spindle rhythm powers were found (Group B), and in the remaining 5 (24%) CP cases neither delta nor spindle rhythm powers were found throughout nocturnal sleep (Group C). A significant decrease in the DQ was found in Groups C as compared with Group A in both CP and reference cases. Moreover, a significant decrease in DQ was also found in Group B as compared with Group A of reference cases.

The correlation between the delta and spindle components in compressed spectral array in 18 infants with congenital cerebral malformation (from 4 months to 4 years of age) was studied throughout nocturnal sleep³⁸⁾. In 7 (39%, Group A) of 18 subjects, periodic changes of delta and spindle rhythm were noted in sleep EEG. In 5 subjects (28%, Group B), delta but not spindle rhythm were found, and in the remaining 6 subjects (33%, Group C) neither delta nor spindle rhythm powers were found throughout nocturnal sleep. A significant increase in DQ was found in Groups C as compared with Group A.

4 Clinical EEG

In 87 mentally retarded children the daytime EEGs were compared with the nocturnal recordings⁴⁸⁾. Fifty-five out of 87 subjects had spindles, including 3 with extreme spindles, and 32 had no spindles during nocturnal sleep. Abnormal background activity and epileptic form or paroxysmal discharges in the daytime examination were observed more frequently in subjects without sleep spindles than in those with them during nocturnal sleep. A significant decrease in DQ was found in subjects with abnormal background activity (or abnormal background activity and epileptiform or paroxysmal discharges) in the daytime examination and no spindles during nocturnal sleep as compared to those with a daytime normal EEG and nocturnal sleep spindles. A significantly low DQ was found in the subjects without spindles during nocturnal sleep as compared to those having spindles.

5 Ontogeny of sleep patterns

Development of nocturnal sleep of 79 mentally retarded children and adolescents (from 6 months to 20 years of age) was studied³⁴⁾. The PSG recordings were carried out while the subject was in bed, and routine sleep parameters were measured. Total sleep time and percentage of stage REM decreased and awake time tended to increase with age. These values were similar to those previously found for age matched non-retarded subjects. Results showed that the basic function of the sleep waking system of retarded children seems to develop normally with age.

6 REM sleep latency

While short and zero REM sleep latency are predominant in the first 3 months of age, this class of latency is sharply reduced thereafter²²⁾.

REM sleep latency observed in 61 mentally retarded infants (4-13 months of age) was studied throughout nocturnal sleep⁴¹⁾. Mentally retarded infants revealed a J distribution of short (less than 8 min) and long (more than 8 min) REM sleep latencies. This feature of distribution was similar to that obtained by other authors from normal infants under 3

months of age. REM sleep latency did not depend on the duration of prior wakefulness. Long REM sleep latencies were no more often preceded by long episodes of wakefulness than were short REM sleep latencies. No correlation was found between REM sleep latency and age, DQ or daytime clinical EEG abnormalities.

7 Sleep spindles

The decrease and the absence of sleep spindles have been reported as characteristics of mentally retarded children.

Sleep spindles longer or shorter than 0.4 sec in 90 mentally retarded children (from 6 months to 8 years of age) were studied throughout nocturnal stage 2 sleep^{27), 44), 47)}. The 90 subjects were classified into 5 groups in terms of spindle length: group 1 subjects with the ratio (number of spindles longer than 0.4 sec/number of spindles shorter than 0.4 sec in duration) of more than 2.00; group 2 subjects with the ratio of 1.99~1.00; group 3 subjects with the ratio of 0.99~0.50; group 4 subjects with the ratio less than 0.50; group 5 subjects without any spindles. The following results were obtained. (1) No significant difference was found among group 1, 2, 3 and 4 in the number of spindles shorter than 0.4 sec. However, the spindle longer than 0.4 sec tended to be fewer in order of groups 1, 2, 3, 4. A significant decrease in the number of spindles longer than 0.4 sec was found in groups 2, 3 and 4 as compared with group 1, and in group 4 as compared with group 2. (2) The abnormal clinical EEG tended to be more frequent in the order of group 1, 2, 3, 4, 5. A significant increase in abnormal clinical EEG was found in group 5 as compared with group 1 and group 2, and in group 4 as compared with group 1. (3) A significant decrease in DQ was found in group 4 and 5 as compared with group 1, and in groups 4 and 5 as compared with group 2. (4) It was concluded that the number of spindles longer than 0.4 sec may serve as a useful indicator of abnormality in mentally retarded children.

The occurrence of REM with sleep spindles during stage NREM has never been reported in normal infants and children. Concurrent of REM and sleep spindle in 45 mentally retarded children (from 4 months to 8 years of age) was studied throughout nocturnal sleep, and the following results were obtained⁴⁹⁾. (1) Twenty-five cases showed a single or burst of REMs during stage NREM with sleep spindles. (2) Twenty-nine cases showed sleep spindles at the beginning of toward the end of stage REM sleep. (3) No significant difference in DQ was found between the subjects with and without REMs during stage NREM sleep. The former subjects, however, had more normal clinical EEGs than the latter. (4) No significant difference in DQ or clinical EEG classification was revealed between the subjects with REMs during stage NREM sleep and those with spindles during stage REM sleep. (5) It was conducted that the concurrence of REM and sleep spindle during stage NREM is a useful sign for early diagnosis of mental retardation.

8 REM activity

It has been reported that in person with Down syndrome, a reduction of R index (number of high frequency REMs against the number of low frequency REMs) was positively

correlated with low IQs⁶⁾.

REM activity during nocturnal sleep was investigated in 27 infants with developmental disabilities^{26), 50)}. A relationship was found between REM and developmental psychomotor function. Developmental psychomotor scores were correlated with percentage of REM20, the percentage of sleep stage with REM. Percentage of REM to total sleep time were important in association with DQ. The findings are consistent with sleep cognition hypothesis proposed by Espie, Paul, McFie, Amos, Hamilton, McColl, Trassenko, and Pardy^{8), 26), 50)}.

9 SPRs

SPRs of 89 mentally retarded children were studied during their nocturnal sleep^{32), 40)}. Forty-three out of the 89 subjects showed more SPRs (type A) during NREM sleep than in REM sleep. The opposite was observed in 10 cases (type C), and 4 had evenly distributed SPRs during both sleep phases (type B). The remaining 32 subjects had mixed types AB (n=19), AC (n=6) or BC (n=7). Type B and C (including the mixed type) were observed more frequently the subjects with low DQ and abnormal clinical EEGs than for those with high DQ and normal clinical EEGs. Since it has been well established that normal subjects of 3 months and over exhibit exclusively type A, SPRs may be used as an additional tool for the diagnostic assessment of mental retardation in early infancy.

10 Skin temperature

In healthy infants less than six months old the hand dorsum temperature rises just before and during stage REM compared with respective preceding level (immature pattern), whereas in subjects of six months over it falls just before stage REM and continues to fall during stage REM (adult pattern by Matsumoto, Morita, & Kiuchi¹⁴⁾). The dynamics of hand dorsum temperature during nocturnal sleep have been studied in mentally retarded infants aged 0-5 years (n=25)^{31), 45)}. All subjects revealed both adult and immature patterns. The adult pattern tended to increase with age from 0- to the 2 year old group.

11 Heart rate

Heart rate variability in four normal and 30 mentally retarded children was investigated during nocturnal sleep²⁴⁾. In four normal and 25 mentally retarded subjects, a high frequency component defined as spectral activity between 0.15 and 0.5 Hz was found in the compressed spectral array of power spectra. A periodic change was seen in the high frequency component which corresponded precisely to the sleep cycle. However, four children with mental retardation did not show normally expected change in the high frequency components specific to sleep stages; a quantity of the high frequency component in only one sleep cycle (n=2; 6 mo. and 3 yr. 8 mo.), a decreased quantity and discontinuity of the high frequency component in only the second sleep cycle (n=1; 1 yr. 2 mo.), and no high frequency component in any sleep cycle (n=1; 3 mo.). These findings suggested that some mentally retarded children have an abnormal parasympathetic nervous activity.

12 Respiration

Respiratory disturbances during sleep have never been reported in mentally retarded

infants.

Respiratory pauses (ResP), apnea (Ap) and periodic respiration (PerR) observed in 42 mentally retarded infants (from 9 to 50 weeks of age post term) were studied throughout nocturnal sleep²⁸⁾. All subjects had ResP in the recordings. The range of ResP rates was 4.1-42.1/h, and the mean was 13.5/h. Ap occurred in 22 out of 42 subjects. The range of Ap occurrence (10-15 sec duration) among subjects was 1-10, with a mean of 4.1. Moreover, in 4 out of 22 subjects, Aps of greater than 15 sec duration were seen. In 30 out of 42 subjects, PerR occurred. The range of occurrence of PerR among subjects was 1-35, and the mean was 9.6.

IV Discussion

1 EEG patterns in sleep cycle

Since this study had no age and sex matched control, results obtained by several other authors were referred to as normal data. The hypnagogic activity was observed in 18 out of 22 subjects with normal EEG, and in 12 out of the 21 other subjects with abnormal EEG. The diffuse rhythmic theta waves were observed in 17 out of 22 subjects with normal EEG, and in 10 out of 21 other subjects with abnormal EEG. They classified the hypnagogic activity into stage 1 and diffuse rhythm theta waves into stage 2. These cases under 18 months definitely had decreased spindle activity in comparison to the values obtained by Tanguay, Ornitz, Kapiian and Bozzo⁵⁴⁾.

A significantly low DQ was found in these children with abnormal EEG patterns throughout all NREM stages as compared with abnormal patterns during stages 1 and 2. The range of EEG abnormalities, as could be expected, was greater in former than in the latter.

Polygraphic study on mentally retarded children⁴⁸⁾ did not reveal abnormal sleep EEG patterns such as indistinguishable stages 1~4 due to little change in delta and theta activities, low voltage activity throughout nocturnal sleep, absence of REM sleep, and the abnormalities in light sleep, simply because this study have been based almost entirely on EEGs recorded in the short sleep time. The significance of EEGs recorded all night should be noticed, at least, in the mentally retarded children.

2 Integrated EEG

The hypothesis was for significant differences among the sleep types in DQ; this is, that there should be significant differences in the mean scores for developmental disability, expressed as DQ, across the three groups defined by the different EEG patterns during sleep. This was what the analysis of variance tested. Different sleep patterns appeared with different magnitudes of disability.

There were no age-matched controls. The lack of control would call the significance of any findings into question, unless it were accepted that high voltage delta and spindle are

the norm in slow wave sleep and light sleep in infants. There is, however, a great difference between obtaining statistical significance in mean levels and having tests of diagnostic utility. This study involves the problem of delta rhythmicity in infants with developmental disabilities, which can only be resolved by further research.

In the present study, this method of assessing the severity of the infants' developmental disorder used estimates of DQ. These DQ values were reduced in infants with developmental disability in proportion to severity of the developmental disability. In the present study, lower mean DQs were found in Groups B and C than in Group A. These findings suggested that the presence or absence of delta and spindle rhythm powers on the EEG during nocturnal sleep could be taken as an index of severity of developmental disorders in infants with developmental disabilities.

3 Compressed spectral array

In 10 (10.4%) of the present cases, no variation of delta and spindle components in compressed spectral array was found. It is suggested that, in certain mentally retarded children, delta components do not increase during NREM sleep, and spindle bursts do not develop throughout nocturnal sleep.

Children with abnormal sleep EEG patterns of indistinguishable stages 1~4, due to little change in delta and theta activities throughout NREM sleep, tend to have a lower DQ than those in whom abnormal EEGs of very high voltage activity occur only during stages 1 and 2⁴⁶⁾. Moreover, children without sleep spindles tend to have a lower DQ than those with spindles, and a significant decrease of DQ was found in cerebral palsy infants with all indistinguishable or 1~4 stages, short sleep and long wakefulness, and no delta rhythmicity in integrated records as compared with those with all the normal stages⁴²⁾. In the present study, abnormal clinical EEGs and low DQ scores were significantly more frequent in groups B and C than in group A. Moreover, lower DQ scores were significantly more frequent in group C than in group B. These findings suggested that the correlation between the presence or absence of delta and spindle rhythm powers in nocturnal sleep EEG could be taken as an index of the severity of developmental disorders in mentally retarded children.

4 Clinical EEG

Children without spindles during nocturnal sleep tended to have a lower DQ than those with spindles. In the present report, significantly low DQs were found in subjects without spindles during nocturnal sleep as compared to those with them. Moreover, the former subjects had more abnormal EEGs in daytime examination than the latter. These findings confirmed again our previous suggestion that the absence of spindles during nocturnal sleep is an effective indicator of the severity of developmental disorders in mentally retarded children.

5 Ontogeny of sleep patterns

Notwithstanding varying etiologies and varying degrees of retardation of the present subjects, the main finding was that the effects of age were sufficiently strong to produce a

significant correlation with age in the direction expected from studies of non-retarded subjects. Moreover, this study had many methodological difficulties that are virtually inherent in this type of study; the heterogeneity of the subject sample, the use of the first night data, and the absence of direct non-retarded control subjects. This study would have to question this results if this study had found marked differences between this subject group and non-retarded control groups reported on in the literature that were at the same time inconsistent with previous reports of differences between retarded and non-retarded subjects such as the absence of sleep spindles in retarded subjects²⁷⁾. Despite methodological difficulties, however, main findings of this study show a approximately similar ontogeny of several sleep patterns between retarded and non-retarded subjects.

6 REM sleep latency

While short and zero REM sleep latencies are predominant in the first 3 months of age, this class of latency is sharply reduced thereafter²²⁾. Although the mean REM latency increases with age, there is no continuous shift from short to long REM sleep latencies, since the distribution of REM sleep latencies turns out to be bimodal with a gap in the time interval 8-16 min²²⁾. Because all subjects in the present study were 4 months or order, they expected a certain bimodal distribution of REM sleep latency similar to normal older infants (over 4 months of age) reported by Schlz, Salzarulo, Fagioli, and Masstani²²⁾. The REM sleep latencies showed a J distribution, and this feature was similar to that of normal young infants under 3 months of age reported by Schlz et al.²²⁾.

7 Sleep spindles

The decrease and the absence of sleep spindles have been reported as characteristics of mentally retarded children. As for short spindles in duration, Metcalf and Jordan¹⁶⁾ have reported an increase in the number of short spindles (about 1 sec in duration) during the second year of life, suggestion that by 18 months of age there may be as many as 4~5 spindles per 10 sec of EEG recording. However, spindles shorter than 0.4 sec have never been studied in normal subjects.

Spindles longer than 0.4 sec tended to be fewer in the order of group 1, 2, 4, 4. It is suggested that for the classification of mentally retarded children in terms of spindle length, an important factor is not the number of spindles shorter than 0.4 sec, but rather of those longer than 0.4 sec.

It has been reported that in normal infants, the presence of a spindle should not be defined unless it is of at least 0.4 sec in length⁵⁶⁾. Since the number of spindle shorter than 0.4 sec in duration has never been reported in normal infants, one cannot say that the number of spindles shorter than 0.4 sec in the present cases is larger or smaller than in normal infants. It seems, however, that the spindles shorter than 0.4 sec or the spindles lower than 15 μ V in amplitude are similar to the pre-spindles lasting a few days and grade 1 spindles persisting for approximately 7~10 days after birth reported by Metcalf¹⁵⁾.

Children without sleep spindles tend to have a lower DQ than with spindles^{27), 48)}.

Moreover, children with abnormal sleep patterns throughout NREM sleep tend to have a lower DQ than those in whom abnormal EEGs occur only during stages 1 and 2⁴⁶⁾. In the present study, significantly fewer spindles longer than 0.4 sec were found along with lower DQ scores, and a significantly more abnormal clinical EEG was found in group 4 than in group 1. Furthermore, significantly fewer spindles longer than 0.4 sec were found along with lower DQ scores in group 4 as compared with group 2. These findings suggested that the number of the spindles longer than 0.4 sec can serve as an effective indicator of the severity of developmental disorders in mentally retarded children.

The occurrence of REM with sleep spindles during stage NREM has never been reported in normal infants and children. The occurrence of REMs with spindles during stage NREM sleep is a useful sign for early diagnosis of mental retardation, especially in early infancy when the diagnosis is not easy. It should be added that the event itself does not mean that the given case is more severe than those without REMs during stage NREM sleep.

8 REM activity

Out of 14 sleep measures, Duration of Awakening Time, Duration of REM Stage, and Percent REM20 are important in their association with the DQ^{26), 50)}. It has been reported that in person with Down syndrome, a reduction of R index (number of high frequency REMs against the number of low frequency REMs) was positively correlated with low IQs⁶⁾. The present finding that the correlation was between the DQ and Percent REM20 activity was consistent with the previous report.

It is known, at least for healthy infants, that type of sleep changes developmentally during the first year. Duration of REM Stage is associated with developmental level¹²⁾. This study's finding of longer Duration of REM Stage associated with higher DQ might be taken as an indication of the development of a sleep pattern. However, since the difference in the DQ of this study's infants reflects the severity of disability, the longer Duration of REM Stage may be also the reflection of the lesser disability of these infants.

9 SPRs

The SPR frequency of REM sleep in non-retarded children increases significantly during the first month of life and does not change subsequently. On the contrary, SPRs in NREM sleep increase from 3 days to 25 weeks after birth, so the SPR frequency in newborns is slightly higher in REM than in NREM sleep. From 1 to 8 weeks this tendency persists but becomes less remarkable. For 9~12, and 13~25 week old infants, on the contrary, SPRs are more frequent in NREM than REM sleep⁴⁾. This distribution is the same as for children 2 years of age and older²⁾ or adults¹²⁾. Because all subjects in the present study were 3 months of age or older, the SPR distribution was expected to be type A; although 43 out of 89 subjects were type A, the remaining 46 subjects were type B and C, which corresponded to normal subjects of less than 3 months old. The results suggested that the presence of types B and C in the subjects indicated abnormality.

In the present study, abnormal clinical EEGs and low DQ scores were significantly more

frequent in type B and C than in type A. These findings suggested that the type of SPR distribution during sleep could be taken as an index of the severity of developmental disorders in mentally retarded children.

10 Skin temperature

Since the subjects of this study were few, this study could neither establish a consistent developmental tendency with age, nor find any correlation between any etiology and the dynamics of skin temperature during sleep. Those results corroborate the suggestion of Matsumoto, et al.,¹⁴⁾ namely, that the temperature change before and during stage REM sleep serves as a new and useful indicator for the development of brain function.

11 Heart rate

Mentally retarded subjects have wide ranging age and various (or unknown) etiology. Various factors (age, etiology, sex, body weight, etc.) are considered to affect the results, especially on the autonomic function. This research should be continued by investigating more cases because classification of the subjects will make the results more clear. Aside from this small sample, data were not separated by sex because no sex difference in sleep parameters has been reported for non-retarded and adolescents⁵⁷⁾.

It is necessary to discuss why four subjects showed the differences in the high frequency component because this point is a main topic of this study; however, discussion is not possible for there were too few data.

The low frequency component decreased during sleep with minimal values in slow wave sleep, whereas the high frequency component displayed an opposite trend with increased values during sleep, reaching a maximum at the time of the low frequency nadir in slow wave sleep. At each epoch studied, the variation in low frequency and high frequencies were reciprocal. This suggests that the increase in high frequency components represent mainly surges in parasympathetic activity and the decrease in low frequency express a declining parasympathetic activity. These results indicate a clear increase in parasympathetic activity during sleep, with the highest values being reached during slow wave sleep. During REM sleep, a decrease in the over-all parasympathetic activity occurred very near the level of awaking. In the present study, four normal subjects showed changes in the high frequency components specific to the sleep stages. It is suggested that at least children of ages between 9 months and 11 years show changes in the high frequency components specific to the sleep stages. However, three mentally retarded subjects showed did not show normally expected changes in the high frequency components specific to the sleep stages. In one of them (3 yr., 8 mo.), a high frequency component was found in only one sleep cycle, and it was smaller and discontinuous. In another (1 yr., 2 mo.), a decreased quantity and discontinuity of the high frequency component was found only in the second sleep cycle. These suggest an abnormal parasympathetic activity. The remaining mentally retarded subject (3 mo.) showed no high frequency components in any sleep cycle. Previous studies have shown that heart-rate variation at the respiratory frequency (respiratory sinus arrhythmia)

predominates during epochs of quiet sleep, particularly after 2 months of age, whereas during REM periods lower frequency variation is produced²¹⁾. In the present case, abnormal parasympathetic activity was concluded, since the subject was 3 months old.

The approach used in this study is a noninvasive method and allows continuous monitoring of autonomic cardiorespiratory control throughout nocturnal sleep.

12 Respiration

It has been reported that normal infants show very large changes in respiratory rate during the period of 9~19 weeks after birth¹³⁾. In the present study, the number of subjects aged 9~19 weeks was small (n=4) and their results were added to those of the subjects aged 20~50 weeks, since no definite difference in respiratory interruption was found between the former and the latter groups.

Respiratory disturbances during sleep have been reported in normal and premature infants²⁰⁾ as well as in sudden infant death syndrome²³⁾. However, respiratory disturbances during sleep have never been reported in mentally retarded infants.

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