A CASE OF RECURRENT ATTACKS OF PROLONGED SLEEP *

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SYMPTOMS

1. Attacks of prolonged sleep, lasting from one to six days.
2. Cataplectic attacks, fainting attacks and short attacks of sleep, lasting usually five minutes.
3. Fugues, somnambulisms, and other dissociative phenomena.
4. Vomiting.
5. Epistaxis.

History.—The following account was given by patient's mother. The patient was born in Edinburgh 22 years ago. It was a difficult labour, forceps being used, and there was some doubt whether the child would live. When she was one year old, the family removed to China and remained there for twelve years.

The father was a marine engineer on a Chinese coaster and, because of the nature of his work, was seldom at home. Life in the East proved difficult. At times they were boycotted, and sometimes without food. Fighting was constantly going on, and patient's attendance at school was necessarily irregular; however, these events do not appear to have caused her any particular anxiety and her memories of the country itself are of the pleasantest.

In childhood patient was not subject to any neurotic traits, such as stammering, nail-biting, bed-wetting, etc., but she did, on numerous occasions, walk in her sleep, and the mother recalls one occasion on which she unbolted the door and was discovered in the garden. She was on intimate terms with her mother and confided freely in her. Her father she knew less well, mainly because of his long absences when at sea, but she had a deep affection for him. She was an imaginative child, impressionable, thoughtful, but friendly and sociable.

When patient was aged 10, her mother had a second confinement. Three years later the mother and her two children returned to this country for a holiday. While in this country, news reached them that the father had died suddenly at sea. The man who was looking after their affairs proved to be unreliable and appropriated most of their possessions. The family were left almost penniless, and the mother, in order to earn a livelihood, invested such money as she had in a hairdressing business, which she herself managed. Patient was sent to school, where she found that she was much behind the standard expected for one of her age, but by hard work she soon made good the deficiency, and, in fact, took a high place in her class. She left school at the age of 16 having obtained her Leaving Certificate and commenced work in a large

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warehouse. Since that time she has had poor health and has had several fairly prolonged absences from her employment.

The state of affairs at home was unhappy, mainly because the mother became acquainted with a man in the hairdressing business whom she took into partnership. The mother states that she married this man six years ago, and, although they have never lived together, he has frequently visited her house at nights in a state of intoxication and has caused a great deal of trouble. Moreover, his craving for drink led to his appropriating all the available cash, and it was not very long before he had ruined the business. The mother states that the patient had a certain influence over this man which she utilized to the full in the hope of protecting her mother from any unpleasantness.

Previous Health.—Patient had typhoid when nine, malaria on numerous occasions, dysentery at 11, measles at 12. Since the age of 16 she had vomited practically every morning up to the time she was admitted to hospital, aged 21. At the same time also fainting attacks started and have been present off and on ever since. The menarche occurred at the age of 17. Since then she has been irregular and has been troubled with dysmenorrhœa. At the age of 18 she had an attack of jaundice and was ill for two or three months at that time. Three weeks after the onset of jaundice she had repeated hæmorrhages from the mouth and nose and became extremely weak. A right-sided hemiplegia slowly developed. For two months she was unable to walk unaided out of the room because of the weakness of her right leg. The right arm was also affected. However, patient's doctor has no note of this central involvement, but states that the jaundice itself lasted some five weeks.

When patient was aged 20, following a long history of abdominal pain, she was diagnosed as suffering from chronic appendicitis, and on operation an inflamed retrocæcal appendix was found. The appendicectomy did not improve her general health, and three months later she was admitted to the Royal Infirmary, Edinburgh. A report was received from Dr. J. D. Comrie, in whose ward patient was from October 4 to 29, 1932. She was admitted complaining of vomiting, pain in the stomach and fainting terrors. The vomiting ceased soon after admission. X-ray examination was inconclusive; there was some tenderness over the duodenal region, but no ulcer crater was seen on X-ray. The benzidine test of the vomit and stools was negative. She had a sleep attack which lasted 36 hours, during which there was no response to painful stimuli. She was discharged 'improved' as a case of hysteria.

Eventually, in February, 1933, she was admitted to the Jordanburn Nerve Hospital.

Physical State.—Physical examination on admission, apart from the neurological findings, showed nothing definite. She was of the slightly-built, asthenic type. The blood picture was within normal limits. Fractional test meal gave normal figures and benzidine test of the stools was negative.

Sleep Attacks.—From the patient's initial attack of sleep in October, 1932, till the time of her discharge from hospital in November, 1934, there were 12 attacks of sleep lasting anything from 24 hours to six days.

These attacks present a constant picture. There is a preliminary period of malaise, sharp stabbing pain on top of the head, a feeling of fullness in the head, nausea, occasional vomiting, a feeling of constriction in the chest, salivation, occasional sweating. The feeling of fullness in the head is a constant feature. Frequently there is an epistaxis. If this is profuse, there is a marked feeling of relief and the onset of sleep is prevented.

Patient then sinks into a state of sleep, the depth of which varies considerably, from a state of mere drowsiness and muscular weakness to a deep sleep. In the former, patient, though conscious of everything going on around her, is unable to make the

slightest movement, and appears to be asleep. If an attempt is made to rouse her she can recover control of her musculature. In the latter, the corneal reflex is absent and a plantar extensor response is obtained, with sometimes even a crossed response; repeated stimulation produces irradiation of the stimulus, and a flexor response of both limbs. She is entirely insensitive to pain, lumbar puncture producing no response. Tendon reflexes are present. The pupils vary, usually being contracted, but reacting to light and to loud noises. The pulse is usually about 60 to the minute, respiration about 16 to the minute and blood pressure 90/60. Patient may rouse sufficiently during these sleep attacks to take fluids, and attend to her bodily needs. She is never incontinent, but she has no recollection afterwards of these almost automatic acts.

On rousing, she feels extremely unwell. There is marked fatigue and weakness, which persists for 24 to 48 hours. Dimness of vision and diplopia are present. The headache, constriction in the chest, nausea, salivation, etc., which are experienced at the beginning of the attack are still present, but the sensation of fullness in the head is absent. Vomiting frequently occurs. She has no idea as to the duration of her sleep. The eyes are frequently suffused and red. Frequently 24 to 48 hours elapse before patient is able to take any solid food or feels free from the symptoms already noted. It is interesting to note that on two separate occasions the temperature rose to 99.5° F., and subsided to a subnormal level again within eight hours.

It appeared desirable to investigate this case from three different angles: the neurological, the biochemical and the psychological.

Neurological State.—On examining the nervous system on admission, it was found that there was a complete right-sided hemianæsthesia with lack of discrimination and dullness of sensation on the right side of the whole body, including the face. Tendon reflexes were equal and exaggerated on both sides, a flexor plantar response was obtained, abdominal reflex was present. The grip was stronger on the left side than the right and there appeared to be some loss of power in the right leg. The pupils were equal, regular, and reacted to light and accommodation. There was, on rough testing, a concentric diminution of both fields of vision. The pharyngeal and palatal reflexes were absent, as was also the corneal reflex. The optic discs showed no evidence of papilloedema or other abnormal state.

X-ray of the skull showed an unusual appearance, probably developmental in origin; the ridge on the inner surface of the skull corresponding to the attachments of the longitudinal sinus and falx was deviated from left to right in the occipital region. Otherwise the X-ray was negative, and there was no indication of increased intracranial pressure. It was found on subsequent examinations that the degree of anæsthesia varied in different regions. It was most marked in the limbs. For example, testing with a sharp point from fingers to shoulder on the right side, no stimulus was felt; from shoulder to mid-line the stimulus was felt as blunt; and from mid-line to the left hand it was felt as sharp. Degrees of temperature were not appreciated on the right arm. A similar state of affairs was present on the right leg as high as the knee, but above that discrimination was merely blunted. This was the case also on the right side of the body and face. The actual depth of the anæsthesia varied, however, on separate occasions, it being possible on one occasion to draw blood by pricking without causing pain, while on a subsequent occasion the same area might be sensitive. Sensations of pain, light touch and temperature were absent from the most anæsthetic areas, but pressure sensation and muscle and joint sense were nowhere absent.

On one occasion patient experienced a tingling sensation which involved the left hand, arm, left side of her face, tongue, and finally her throat. Her tongue felt large and she had difficulty in speaking. When the sensation reached her throat, she felt that she was choking and actually lost consciousness for a minute or two. On being examined shortly afterwards, her left arm and left side of her face were

partially anæsthetic, while the sensation of the right side of her body appeared to be normal. The following day anæsthesia had returned to the right side. Further, when first examined, patient answered 'yes' when touched on the sound side and 'no' when touched on the anæsthetic side.

On one or two occasions patient experienced a tingling sensation, starting in the right hand and progressively involving the arm and the right side of the face, the parts at first feeling cold and numb. There was an initial blanching followed by a blue discolouration. These attacks lasted from 10 to 30 minutes.

There have been various phenomena of partial or complete unconsciousness, which started with the syncopal attacks some seven years ago. The syncopal attacks had a gradual onset, with weakness, dizziness and loss of consciousness, and were presumably due to some vasomotor instability; but sometimes consciousness was lost without any warning, 'the ground suddenly coming up to meet' the patient. Such attacks cannot with certainty be differentiated from petit mal, especially as on one or two occasions patient reports that she went to sleep leaning against a wall. The absence of an aura, the lack of rigidity or stereotyped movements, and the variability of the attacks, however, make this unlikely. Neither of these attacks represents a vasovagal type as described by Gowers. Consciousness was definitely lost and was not affected by position.

Since the initial attack of prolonged sleep there have been occasional bouts of sleep lasting five to ten minutes, sometimes accompanied by the usual prodromata of a prolonged sleep attack, but usually coming on without any warning. These are differentiated from the attacks resembling petit mal simply by the feeling of drowsiness which invariably precedes and follows them, and by the fact that patient can be roused.

Finally, without any warning, patient has had sudden attacks of tonelessness. Emotional factors may produce such an attack, but occasionally no factor can be discovered. Patient falls to the ground and is incapable of any movement for five to ten minutes. These cataplectic seizures have occurred on about twelve occasions. They are never brought on by laughing.

During the periods of prolonged sleep there has recently been a tendency to the development of a cataplectic-like state at the beginning of the sleep attack, when patient feels that she could be wakened if she were shaken. This actually has been successful on several occasions. At other times this state has been brought on during sleep by patient having a recurrent dream, which is always accompanied by a feeling of dread. For instance, on one occasion patient went to sleep suddenly. She had complained of pain in the vertex of her head, constriction in her chest, etc. 'I was never really asleep—more drowsy. I realized what was happening in the ward almost all the time, but simply could not move or make a sound. Then I seemed to be on the side of that hill again. The fog was forcing me down and I felt terribly afraid. It seemed to be the end, that this time I was utterly lost, and there was no way out of it at all.' In spite of the dream, she could hear people talking in the ward and felt a longing to get in touch with them. The pain in her head became so intense that she screamed. There was a flash of light through the fog and she remembers nothing else till she woke up, bathed in perspiration and terrified. 'I felt as though I had had a terrible struggle and was quite limp. I felt that if I had not struggled against these two things I would have had no pain and just passed away.' Actually patient did scream, and tore her hair in apparent agony. She fell out of bed, and it was later noticed that she had been biting her lips.

Thus we have various phenomena, ranging from a transitory weakness without actual falling, through various degrees of unconsciousness to prolonged sleep.

A striking aspect of the case has been the predominantly vagal nature of many

of the symptoms associated with the sleep attacks. Patient has invariably experienced a feeling of nausea with salivation and occasional sickness, and a feeling of constriction in her chest, producing a sensation of suffocation. These symptoms were still present when patient awakened. There has been ample evidence of vasomotor instability, and the feeling of fullness in the head which is a constant prodromal symptom is one indication of this. A peculiar vivid purplish discolouration is sometimes present around the eyes, spreading beyond the lids. The eyes themselves at such times are frequently suffused, and we must suppose a marked engorgement of the ophthalmic veins to explain these signs. The pupils, during sleep attacks, do not present a constant picture, but they are never markedly contracted. The blood pressure is fairly low, 90/60, and the pulse about 60.

Biochemistry.—Careful records have been kept of the biochemistry of this case during sleep attacks and also at normal times. They indicate a tendency towards acidosis and probably ketosis, since the ammonia coefficient of the urine, over a period of approximately two months, was consistently raised (5 to 16). The phosphate excretion remained at the normal figure, while all other excretions were greatly reduced. This reduction, however, is explicable by the fact that the basal metabolic rate was low (average — 20 per cent.), and food intake has always been small. Both in the sleep states and in the normal the respiratory quotient was about 0.72, such a figure suggesting some abnormality in the general metabolism, possibly due to carbohydrate lack.

In the sleep state appreciable quantities of acetone bodies were excreted, without, however, any apparent relation between the length of sleep and the amount excreted; for example, in an 18 hours' sleep ++++ of acetone, in 72 hours' sleep ++ (28 mgm. per cent.), in 36 hours ++ (27 mgm. per cent.).

The determinations of the CO₂ combining power gave consistent results throughout (50 to 75 volumes per cent.). No distinction was apparent between the sleep and normal states. This indicates, when taken in conjunction with the urinary findings, a satisfactory ability to combat acidosis.

An attempt was made to discover the nature of the tendency to acidosis. The inorganic phosphate contained in the blood was investigated, but was found to be normal. No acetone bodies could be found in the blood drawn off during a state of sleep, but a high lactic acid content (60 to 107 mgm. per cent.) was found in the blood, both in sleep attacks and in the normal state. The most striking feature of the blood chemistry is the persistently low cholesterol content (70 to 105 mgm. per cent.). This finding bears an interesting relationship to results obtained from a case of typical narcolepsy, with short diurnal attacks of sleep and cataplexy. This case has normally a high cholesterol content (200 mgm. per cent.), while in both sleep and cataplectic states the value falls to below 100 mgm. per cent.

It is interesting that in the case under observation the cholesterol content rose as the frequency and intensity of the attacks decreased. Glucose tolerance tests performed during a sleep state and when awake gave normal curves, and further, the fasting level was the same in both cases. In the already mentioned case of typical narcolepsy the fasting level of the sleep state was lower than when awake.

These findings indicate a tendency to acidosis and ketosis, but do not, however, give an indication as to the cause of the sleep attacks. The cholesterol finding is interesting. According to Duncan,¹ the blood cholesterol is lowered in states of heightened emotion and there is accompanying increase in the basal metabolic rate; however, this case suggests much more a state of emotional inertia and there is a diminished metabolic rate.

Psychology.—It is necessary to condense the large amount of material which has been obtainable by free association and by analysis of the patient's dreams.

Unfortunately much of the material recalled has been of such a nature that it has been impossible to obtain confirmation of it.

Patient believes her mother to have been unfaithful to her father while still resident in China. She maintains that she was present on numerous occasions while intercourse took place between her mother and her father's best friend. The father's prolonged absence at sea favoured such a liaison.

Somnambulisms were frequent at this early age and would indicate that there was a certain amount of conflict going on in the patient's mind. She recalls being greatly upset by her mother's unfaithfulness, the more so as she was constantly in her company and to a large extent idolized her.

Patient believes her younger sister to be illegitimate and states that her suspicions were confirmed when, after her father's death, her mother periodically received money from China, presumably for the upkeep of her younger sister. She maintains, further, that her mother never married her partner in the hairdressing business; that she has confirmed this by perusing the official register, and that, therefore, her youngest sister is also illegitimate. She believes that her so-called 'step-father' and herself are the only two who really know the truth behind her mother's love affairs, and, in order to hide the truth from the world, she is forced to assume a rôle of complete ignorance.

There seems to be no doubt that her insomnia originated shortly after the age of 13 when she returned to this country. She maintains that she formed the habit of lying awake at nights in order to protect her mother if necessary from the advances of her drunken lover.

The patient left school in June, 1928. The vomiting and fainting attacks started about that time. The dates indicate that the mother was about three months' pregnant and, as patient was already quite familiar with the accompanying symptoms, it seems almost certain that her suspicions were aroused. In any case it is certain that patient was troubled with morning sickness from that time.

Between the ages of 16 and 20, patient's life was no happier. The unannounced, nocturnal visits of her mother's lover continued, and at times it was necessary for the mother and daughter to seek refuge in the middle of the night at the grandmother's house. Later the grandmother came to live with the family, and was constantly finding fault and complaining about trivialities. An aunt who lived nearby, by her quarrelsome behaviour, added to the general disharmony. The relatives, who imagined that patient's mother was married, could never understand why she would not get a separation.

When patient was 19 she became attached to a medical student. It was her first serious love affair and lasted 18 months. They were not actually engaged to be married, but there was a mutual understanding. The romance, however, ended suddenly. The young man had asked patient to go to a party. She did not feel well enough to accompany him and he promised not to go without her. However, he did not keep his promise, and at the party he proceeded to get drunk. He had intercourse with a girl whom he barely knew and later learned that she was pregnant. He felt it his duty to marry the girl, but the couple were unhappy together. Patient feels certain that she would have been happy living with this man, and this misfortune merely tended to confirm her distrust in humanity. Her friendship with the young man ended in November, 1932.

The mother confirms the fact that the unhappy domestic situation, together with patient's poor general health and her disappointment in love, had combined to make the situation an intensely unpleasant one. Patient herself maintains that, prior to being admitted to the Edinburgh Royal Infirmary in October, 1932, she frequently contemplated suicide and prayed fervently for death.

Three months elapsed from the time of her discharge from the Royal Infirmary

till the time of her admission to the Jordanburn Hospital, and during that time she had several attacks of prolonged sleep.

DISCUSSION

At first it was considered that there was some organic change, encephalitic or neoplastic in origin, situated in the brainstem, to explain the various symptoms occurring in this case. The attacks of sleep, the vasomotor disturbances and the metabolic upset might all have been explained on this basis. However, the variability of the anæsthesia, the presence of the 'Yes-No' phenomenon, the absent palatal, pharyngeal and corneal reflexes, the absence of any evidence of increased intracranial pressure, the concentric diminution of the fields of vision, the fact that the condition progressively improved with psychological treatment, and finally the temporary transference of the anæsthesia from the right side to the left side, all tended to throw the emphasis on a psychological explanation of the condition.

Turning now to the question of the sleep attacks, Kinnier Wilson² includes under the heading of narcolepsy two definite forms: (1) short attacks of sleep, associated with cataplexy; (2) attacks of prolonged sleep, among which he specifies those caused by hysteria, cerebral tumour or abscess, epidemic encephalitis and trypanosomiasis. This author makes several references to the as yet little understood accompanying symptoms of epistaxis and diplopia. In the present case a previous epistaxis, when it occurred, appeared always to prevent the onset of a sleep attack.

The cataplectic-like states already described, which occur either at the beginning or end of a sleep attack, correspond very closely to the pre-dormital and post-dormital types of sleep paralysis described by Levin.³ The latter are brought on by the emotional experience of an unpleasant dream, and this was found to be present in this case. In his paper the author makes use of Pavlov's concept of spreading inhibition to explain sleep, and suggests that the differentiation between the sleep narcolepsy and the states of cataplexy and sleep-paralysis really depends upon the fact that in the one case the inhibition is confined to the substrate of consciousness, whereas in the other it is confined to the motility substrate. In the former, although the patient is apparently asleep, automatic acts can be performed; in the latter, although patient is unable to move, consciousness is retained. These various points are well illustrated in this case. Patient was able to take food and attend to her bodily needs during a prolonged sleep, while on other occasions, although unable to move, she was fully conscious of her surroundings. However, the phenomena observed in this case, although they embody most of the features of narcolepsy as described by these authors, do not show a sufficient uniformity to fit into any one picture.

The question of the autonomic imbalance would appear to be of particular importance in this case, but in this connection it is impossible to attribute the condition wholly to either the organic or functional aspects of the case.

It is possible that some organic change in the region of the hypothalamus may have been responsible for the various symptoms of parasympathetic overaction, but it is much more probable that it is the result of psychic conflict. The somatic response was obtained through the medium of the parasympathetic and patient tended to become conditioned to such responses.

The objective evidence of cerebral engorgement in this case suggests the possibility of raised intracranial pressure resulting from dilatation of cerebral vessels. There is also the evidence already referred to of engorgement of the ophthalmic veins and the subjective sensation of fullness in the head which frequently leads to an epistaxis. If an epistaxis does not occur, sleep sometimes ensues; a loss of blood is invariably accompanied by marked improvement in the clinical condition.

Several workers have suggested that engorgement of the cerebral vessels predisposes to the onset of sleep, but recently F. A. Gibbs, E. L. Gibbs and Lennox,⁴ using a thermo-electric blood flow recorder, inserted into the internal jugular vein, were unable to demonstrate any significant change in the flow of blood through the brain at the onset of sleep.

Hess⁵ suggests that sleep is reflex in character, with the autonomic nervous system as mediator, a sleep centre probably being situated near the aqueduct of Sylvius. He has produced sleep artificially in cats by injecting ergotamine into the third ventricle. This damps the sympathetic and stimulates the parasympathetic. Electrical stimulation of the brainstem in the region of the supposed sleep centre produces a like effect. Dikshit,⁶ in Edinburgh, has recently produced sleep experimentally by injecting acetylcholine into the lateral ventricle of cats.

The peculiar vascular disturbance confined to the right side of the face, arm and hand has already been referred to. The distribution suggests some connection with the hemianæsthesia, the afferent impulses concerned in the maintenance of the sympathetic tone of the capillaries being deficient, and an atonic condition resulting.

One can trace a growing tendency towards dissociation. There is a long history of somnambulism, and, more recently, of fugues. Other less complete dissociative phenomena are demonstrable. For instance, when in hospital, patient was walking in the grounds with two other patients when she suddenly became aware of the fact that she had no conception of her companions' identity. She continued to converse and eventually left them without having aroused any suspicion in their minds. Inside the hospital people appeared to be vaguely familiar, but she had forgotten their identity. She lay down and slept for an hour, and on waking, except for a slight feeling of confusion which soon wore off, she felt quite well.

The hemianæsthesia also demonstrates her dissociative phenomena. Patient said 'Yes' when touched on the sound side and 'No' when touched on the anæsthetic side. Bernard Hart⁷ suggests that in such a condition 'Yes' and 'No' indicates a connection between a dissociative consciousness

which is aware of the sensation, and a normal consciousness which ignores it.

The cataplectic attacks and similar states closely resembling the pre-dormital and post-dormital types of sleep-paralysis described by Levin may be regarded as further examples of dissociation.

It seems reasonable to suppose that the sleep attacks have developed from the previous less absolute states of dissociation. The faints, cataplexies, amnesias and sleeps may then be regarded as fulfilling much the same function in giving the patient a temporary escape from reality. They would represent a solution, though an unsatisfactory one, of her problems, and the symptoms being taken by her to represent some ordinary illness would not be accompanied by a feeling of concern and would produce the appearance of indifference in this case.

There would appear to be a strong purposive aspect in this case, and thus the personality being disinterested in the removal of the symptoms, they tended to become reflex and autonomic. Kretschmer⁸ indicates how fainting, etc., may be divided biologically into three stages: firstly, the stage of acute affective reflex which is followed by the stage of voluntary reinforcement which falls gradually into the stage where the reflex is chronically impressed on the mind. In other words, the disgust and conflict associated with the early history of this case reflexly produced vomiting and fainting, but the absence of any strong wish to get well, and, indeed, a probable desire for illness, led to a reinforcement of the reflex which thereafter functioned with extreme ease and readiness. Subjective over-emphasis of such tendencies led to more profound manifestations, as, for instance, the varying degrees of unconsciousness. Finally, within recent years, the idea of death has come to have a strong appeal for the patient. It has come to be closely associated in her mind with the idea of sleep, both offering an escape from her conscious worries. Her attitude towards death, however, is ambivalent and the uncertainty of death frightens her. Her insomnia may be partly due to her associating sleep with death and the resulting conflict over her desire and fear of death.

In conclusion, it is now five months since patient was discharged from hospital and she has been free from symptoms except for occasional headaches, insomnia and fatigue.

SUMMARY

An attempt has been made to study from various angles the factors leading up to and found during attacks which are indistinguishable from normal sleep and last for several days at a time. The evidence in favour of an explanation along organic lines was rejected and a psychological explanation is offered.

I wish to express my indebtedness to Professor D. K. Henderson, Physician Superintendent of the Royal Edinburgh Hospital, for permission to publish this case and for much invaluable help and criticism. I am also

greatly indebted to Dr. Henry Tod, Biochemist, Royal Edinburgh Hospital, for his ready co-operation in the biochemical investigations of the case.

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