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Original Articles

PSYCHOSOMATIC ASPECTS OF NARCOLEPSY*

By

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INTRODUCTION

GELINEAU (1880) introduced the term "narcolepsy" and suggested that it constituted a disease *sui generis*. A careful perusal of the literature, however, shows that the condition had been described previously. Precedence has been claimed for Bright, Thomas Willis and even for Galen! At any rate, Notkin and Jelliffe (1934) were able to unearth 270 cases of narcolepsy and allied conditions in a literature survey covering the years 1813–1931.

Narcolepsy, however, was not even referred to in Osler's *Modern Medicine* (1910) and its incidence almost certainly increased in the 1920s, when most of the classical papers were written (Redlich, 1925, 1931; Adie, 1926, 1930; Lhermitte and Tournay, 1927; Wilson, 1928; Levin, 1929; Thiele and Bernhardt, 1933). Cave's series of 42 cases seen at the Mayo Clinic appeared in 1931 and the fine review of Daniels reporting on 147 cases followed three years later. A new era of treatment opened with the introduction of ephedrine sulphate by Janota and Scala and—independently—by Doyle and Daniels in 1931. In 1935, Prinzmetal and Bloomberg reported that amphetamine sulphate partially relieved the sleepiness. The sympathomimetic amines are still widely used in the symptomatic treatment of the condition though recently it has been suggested (Daly and Yoss, 1956) that methyl phenidyl acetate ("Ritalin") may be superior. Isolated cases of other treatments proving effective have been recorded. Thus Riser and Dardenne (1946) described a case of long-term sleepiness responding to 6 electroshock treatments, while Weitzner (1951, 1952) has described a case of narcolepsy and one of sleep paralysis which responded to insulin hypoglycaemia. Phenurone (Aird *et al.*, 1953) has been used and also glutamic acid (Platania and Mancini, 1955). Potassium has been recommended for cataplexy (Dynes, 1943) but the results have been difficult to evaluate (Aird *et al.*, 1953). It will be recalled that the course of narcolepsy is variable, and it is difficult to draw firm conclusions from small numbers of cases. Daniels

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reported in the era before ephedrine that almost 50 per cent. of his cases showed some improvement on follow-up after two to three years.

There has been much discussion regarding the use of the term "narcolepsy". Some authors feel that it should be restricted to cases showing both the sudden, short, overpowering bursts of sleep ("narcolepsy") and the abrupt attacks of tonelessness with preservation of consciousness ("cataplexy"). Others like Wilson (1928) and Daniels (1934) feel that the lines of differentiation should not be drawn too sharply. At any rate when narcolepsy and cataplexy are present together it is the exception rather than the rule to find definite evidence of organic involvement—hence the terms "idiopathic", "true", or "cryptogenic" narcolepsy. A number of other features may be present also: these include sleep paralysis, sleep hallucinosis, somnambulism, recurrent nightmares, a marked and sudden weight gain, and occasionally, hypogenitality, feminine hair distribution, diplopia and headaches. Most of these phenomena may be regarded as exaggerations of the normal. To this list a number of observers have added paranoid psychosis, a rare accompaniment, which is regarded by them as a development of sleep hallucinosis. Thus Brock and Wiesel (1941) state that the "psychosis is always a development of the sleep hallucinatory state". Drake (1949) similarly observes: "Rarely a chronic hallucinatory state seems to develop on the basis of the dream experience."

There have been many theories regarding the pathogenesis of "idiopathic" narcolepsy. They can be conveniently classified into four groups; these are not necessarily mutually exclusive:

1. That all forms of idiopathic narcolepsy are in fact symptomatic. Lesions of the mesencephalic and diencephalic areas in particular may produce typical narcolepsy or conditions closely allied to it, e.g. epidemic encephalitis (Adie, 1926), trauma (Gill, 1941), tumours (Davison and Demuth, 1945–1946). More recently the "reticular activating system" has been delineated and incriminated (Daly and Yoss, 1957). Certain metabolic disturbances acting through these centres or by some other mechanism may also produce the disease, e.g. hyperinsulinism (Delay, 1942).

2. That narcolepsy is closely related to epilepsy (Wilson, 1928; Cohn and Cruvant, 1944; Roth, 1946; Symonds, 1954). However, the great majority of narcoleptics show no real evidence of epilepsy (Pond, 1952; Daly and Yoss, 1957).

3. That Pavlovian concepts throw light on the narcoleptic symptoms. This view has been maintained by Adie (1926), Levin (1935, 1950, 1953) and Fabing (1945, 1946). These explanations, while perhaps not completely satisfying, have the merit of drawing attention to the role of the cerebral cortex in narcolepsy.

4. That idiopathic narcolepsy may represent a neurotic reaction to mental conflicts. This must be distinguished from the viewpoint that narcolepsy is influenced by the emotional state; all observers agree on this; only some feel that mental conflicts may be a necessary and sufficient cause of narcolepsy. The theories of psychogenesis are discussed briefly below.

It should be added finally that in some cases evidence of hereditary factors has been adduced (Krabbe and Magnussen, 1942; Hoff and Stengel, 1931; Cohn and Cruvant, 1944). Such cases appear to be the exception rather than the rule.

Physiological Studies

In symptomatic narcolepsy the laboratory data may reveal evidence of the underlying disease. In the idiopathic condition, however, the laboratory yield

has been disappointingly meagre. The EEG is typically normal apart from showing evidence of sleep activity (Daly and Yoss, 1957). The B.M.R. is lower, on the average, when compared to a normal group (Daniels, 1934; Pond, 1952). The glucose tolerance was abnormal in six out of twelve cases described by Daniels and is considered to be frequently abnormal by Modlin and Scriver (1951). Autonomic disturbances have been reported by continental workers (Hoffmeister, 1948) but the evidence is not yet completely convincing and recently Roth and Simon (1955) were unable to demonstrate any relationship between the autonomic state of narcoleptics and sleep. Richter (1929) stated that skin resistance was high in narcoleptics. However this finding may have been non-specific; resistance was also found to be high in schizophrenic stupors. Daniels noted polyuria and polydipsia in a few cases, while one case with diabetes insipidus has been described (Modlin and Scriver, 1951). Daniels in a study of forty-four cases could not confirm the alleged frequency of relative lymphocytosis. Many observers have suspected endocrinal abnormalities but there has been no proof.

Psychopathological Studies

Interest in the narcoleptic-cataplectic syndrome by psychiatrists is quite recent. Benedetti (1953) estimated that out of two hundred dissertations on narcolepsy, only twenty touched on the psychopathological side of the problem. In 1934, Notkin and Jelliffe carried out their historic review. Using the term narcolepsy to cover other cases of hypersomnia not showing the full features of the narcolepsy-cataplexy syndrome, they distinguished five groups:

1. Those with a definite psychopathological background.
2. Those with definite cerebral involvement—e.g., brain tumours, trauma, arteriosclerosis, etc.
3. Those associated with chronic encephalitis.
4. Combined epilepsy and narcolepsy.
5. Cryptogenic, or idiopathic.

From the review, it appears that practically all cases in which cataplexy and narcolepsy are present together fall into the cryptogenic category. Their first group contains sixty-four cases out of a total of two hundred and seventy gathered from one hundred and forty-one publications between 1813 and 1931. They say of this group that it includes "Cases of hypersomnia and those of attacks of sleep in persons with a definite psychopathologic background. Those cases may not properly belong to the narcolepsies". Certainly such cases as the "sleeping Effie" of Edward (1848) would appear to be related to the psychotic stupors rather than to narcolepsy.

Daniels in his review of one hundred and forty-seven cases of narcolepsy mentions that while the disease presents the patient with many problems, it rarely results in pronounced personality deviations. However, he describes one girl of fourteen years who became psychotic two years after developing diurnal somnolence and terrifying nocturnal dreams. Her hallucinatory experiences began to persist during the day and she came to believe in their reality. There was some improvement with ephedrine sulphate. Another of his cases became irritable and suspicious and neglected her household duties. Daniels leaves the question open as to whether the psychosis associated with narcolepsy is a reaction of the personality to the illness or an extension of some common underlying organic process. The number of other cases of narcolepsy in which psychotic episodes have occurred is quite small according to the literature.

Such cases have been described by Brown (1908), Froderberg (1930), Thiele and Bernhardt (1933), Locke and Bailey (1940), Brock and Wiesel (1941), Lehrman and Weiss (1943), Drake (1949) and Quensel (1952). Possibly Morton's case (1884) should be included also. All the psychoses were of the paranoidal schizophrenic type, but Locke and Bailey were impressed by the unusual degree of insight shown by their case and questioned whether this was a true schizophrenic illness. Most of these psychoses appeared to develop by way of sleep hallucinosis, the vivid hallucinations eventually persisting into the daytime and attaining the status of reality. The case described by Lehrman and Weiss was atypical in this respect: the authors felt that the paranoid symptoms originated in the patient's self-consciousness regarding her attacks. Narcolepsy appeared prior to the psychosis in all these cases although in Locke and Bailey's case auditory hallucinations had been present fourteen years before the onset of narcolepsy.

In recent years, cases of "idiopathic" narcolepsy have been described in which the illness was felt to be a response to severe emotional conflicts. Sleep used as a resistance to analysis was discussed years ago by Ferenczi (1926) and in 1924, Missriegler analysed a case of morbid sleepiness successfully. (In this case, however, cataplexy was not present.) Missriegler, however, is careful to point out that there may have been an underlying post-encephalitic basis for the condition. In 1944, Langworthy and Betz published a paper based on six cases of "idiopathic" narcolepsy and suggested that the disease represented a personality reaction to emotional conflict and that these patients felt "caught in a life pattern to which they are expected to conform but which they deeply resent". Other papers supporting the importance of the psychogenic factors in narcolepsy have been presented by Myers (1920), Jones (1935), Murphy (1941), Spiegel and Oberndorf (1946), Davison (1945), Barker (1948), Deutsch and Murphy (1955) and Switzer and Berman (1956). The last-named authors summarize the evidence in favour of psychogenicity as follows:

1. Narcolepsy frequently begins at a stressful period in life—that is, in adolescence and early maturity. It could represent an attempt to escape those stresses.
2. The greater incidence (2:1) in men may be related to greater stresses on males.
3. The weight gain could be due to substitutive oral activity.
4. The great variation in the frequency of attacks.
5. The lack of abnormalities in the EEG.
6. The favourable response of the condition to mild stimulants.
7. The favourable response to psychotherapy.
8. The fact that Gelineau emphasized the importance of bolstering morale.

These arguments are all, of course, somewhat double edged. The clear relationship of narcolepsy and environmental stresses is not found at other "stressful" periods of life, e.g. the climacteric. It could be argued that women undergo as much stress as men. The weight gain (in case two for example) is often unaccompanied by any increase in appetite and is very sudden and striking. Great variation in frequency may occur in diseases which are probably not psychogenic, e.g., multiple sclerosis, epilepsy. An abnormal EEG is not an invariable concomitant of brain disease, even quite gross brain disease. Response to mild stimulants does not necessarily favour psychogenicity and in any case the narcoleptic frequently takes increasing doses of the stimulant (e.g. case two). At the present time, too few cases have been treated by psychotherapy for the effectiveness of this to be judged. Also these cases seem rarely to have been

adequately followed up. It would be difficult to find any disease in which bolstering morale is not of value!

None of the arguments for or against organicity can be expected to be decisive until more information is available on the nature of the condition. What does seem clear from the review of the literature is that emotional conflicts do affect the manifestations of the condition and the individual's ability to cope with it. The effect of the emotional state of the individual on the condition is particularly obvious in cataplexy (Levin, 1953) but not infrequently actual sleep attacks can be produced by strong emotion (e.g. in the original case of Gelineau, 1880; also in those of Kahler, 1922; Westphal, 1877; Adie (case 2), 1926 and others). The effect of discussing conflictful material on the sleep attacks has been studied particularly in the interesting report of Barker (1948). Barker found that when strong aggressive feelings were aroused sleep attacks tended to intervene.

Case Reports

The two cases to be presented in this paper show pronounced psychological difficulties in addition to narcolepsy. Case 1 showed a definite paranoid schizophrenia and adds another to the small number of cases in the literature. Case 2 showed a neurotic character disorder with paranoid trends. Both displayed "narcolepsy" and "cataplexy" and in neither case was any organic lesion discovered although in Case 2 the illness had followed closely on an attack of "influenza". In addition to their psychological disturbances, they showed some interesting physiological changes.

Case 1

On 30 November, 1955, a twenty-seven year old stenographer, Jean M., was admitted to the Psychiatric Department of the University Hospital, Saskatoon. She had spent the previous six months in the Psychiatric Ward of another general hospital and had received six electro-shock treatments, superficial psychotherapy and largactil. The diagnosis made there was one of paranoid schizophrenia of very recent onset. In addition, she had shown typical features of the narcoleptic-cataplectic syndrome for seven years. For the latter condition, she had been receiving amphetamine sulphate, 5 mg. t.i.d. It was felt that her paranoid schizophrenia had failed to respond to the treatment given.

The patient was a pleasant, well groomed and superficially composed young lady. She smiled frequently and very blandly and at times her smile was quite inappropriate, for example, in discussing her sexual hallucinations, which she claimed she found intensely unpleasant. She showed little concern regarding the future and was herself perturbed by her lack of interest in what happened. She made contradictory statements regarding her hallucinations, saying that the voices she heard were the results of an illness but adding a few minutes later that she suspected a man of her acquaintance of being responsible. These hallucinations were mainly auditory and tactile in character but, in addition, she had from time to time visual and olfactory hallucinations.

The history was obtained from the patient and her relatives. There had been no family history of mental illness although the patient's mother was described as being rather "nervous", while the father and an aunt suffered from "nightmares" but otherwise appeared well adjusted. Jean was the fourth of a sibline of five. During the first few years of her life, she developed normally showing none of the so-called neuropathic traits. At the age of six, she developed involuntary micturition precipitated by emotion. She became extremely sensitive about this and would hide when strangers came into the house. Later, she had to wear pads to protect herself from public embarrassment. She did not suffer from nocturnal enuresis. As she grew older, she avoided mixed company. She herself gave no reason for this. Her sister suggested that it arose from her shyness and self-consciousness over her handicap. She was a conscientious and steady worker at school, completing grade twelve without difficulty. She then left home, took a business course, gained high marks and obtained a job as a stenographer. She proved a skilful and meticulous worker, always paying attention to details, which indeed she had always been noted to do since her early school days. Her work level was maintained until the end of December, 1954, when her psychotic illness appeared in overt form.

In 1948, she had developed the typical features of narcolepsy and cataplexy and the arrival of these two symptoms was followed by the disappearance of her involuntary micturi-

tion. She became troubled by nightmares and these grew more severe as the years passed. The narcolepsy and cataplexy on the other hand were well controlled by amphetamine sulphate, 5 mg. t.i.d. In 1951, she was having frequent nightmares and was afraid of the dark but was otherwise cheerful and well according to her friends. A year later, her nervousness had increased and she was sleeping badly. She went on holiday to her sister's home in 1952 and there she began to complain of mice getting into her bed at night. She would actually see and smell them and would frequently remake the bed during the night. At that time, she also complained of hearing men's voices during the night. In the daytime, she appeared normal except for some nervousness. After a few weeks with her sister, she left for home. She continued to have vivid nightmares but did not mention hallucinations again until December of 1954. At this time, her illness broke out openly. She heard voices by day and by night, accusing her of sexual misdemeanours and became increasingly withdrawn and preoccupied. She suspected men of following her and of spying on her. They seemed to be criticizing everything she did, including her manner of dress, behaviour, appearance, etc. Later she began to feel someone having intercourse with her in bed. She stated that people were masturbating over her clothing and possessions. She became suspicious of her girl friend whom she had known for twenty-two years, and accused her of having sexual relations with a man in her bedroom. This girl was shocked by the change in Jean's mental state. The voices increased steadily in intensity until she began to have long conversations with them. They were usually the voices of people whom she knew at work and were almost invariably men's voices. The "sexual attacks" increased also and she began to wear slacks at night in order to have some protection. Eventually she left her job, went home and would ask her parents to sleep with her because of the frequency of the "assaults". As before, she was most troubled at night but even during the day she would frequently hear unpleasant remarks usually of a sexual nature concerning herself. Her parents were now alarmed and because of this sought psychiatric advice which led to her admission first to the psychiatric ward of a general hospital and then to her transfer to the University Hospital, Saskatoon.

Progress

On admission, she showed disturbance of volition, association and affect and spoke freely of her auditory hallucinations. She displayed, in fact, the typical features of paranoid schizophrenia, but it was noted that she made better contact than the usual patient and also that, for the most part, she was remarkably detached and objective about her hallucinations. An attempt was made to treat her by superficial psychotherapy. This was difficult. There had been some memory loss following the previous course of E.C.T. and she maintained a peculiarly bland attitude towards her illness and was unwilling to talk about herself and her problems before it began. It was apparent that she had never had any sexual education, the family being somewhat prudish about such matters, but, on the other hand, her siblings had shown no difficulty in making adequate sexual adjustment. No evidence of definite trauma in early life was elicited. The parents were rather proud and somewhat meticulous people, but their relationship with the girl seemed a sound one and the siblings seemed particularly mature and well-adjusted people.

It was difficult to obtain from her the full details as to when her illness really began. This was partly due to the slight amnesia following E.C.T. It was clear, however, that the first symptoms of her disturbance had appeared at night, in the form of nightmares which persisted into the waking state and throughout her illness, it was apparent that the symptoms tended to be intensified at night.

Amphetamine was discontinued, but there was no amelioration of the psychosis. It was then decided to give her an additional course of electro-convulsive therapy and, in addition, to put her on niacin, gr. 1 t.i.d., which has been reported by Hoffer *et al.* (1957) as being of marked value in cases of acute schizophrenia. She received six electro-shock treatments in all on this occasion. Towards the end of her stay, she was put on frenquel, 40 mg. t.i.d., as this had been reported as being of benefit in schizophrenia. During her stay in hospital, there was a slow improvement in her mental condition, the voices remaining but becoming less troublesome and her concentration improving. She continued to have occasional narcoleptic and cataplectic attacks in hospital but no definite relationship of these to stress was discovered. However, it was noticed that the cataplectic attacks increased in frequency at a time when she was impatient to leave the hospital but did not care to express this to her physician; this might, of course, have been coincidental. Her work in the occupational therapy department showed steady improvement and by the end of her stay she was able to carry out stenographic work satisfactorily. She was discharged from hospital on 10 March, 1956 and after a short holiday obtained a job as a stenographer in another town.

It was possible to have her followed up satisfactorily by a psychiatric social worker. She continued to take the niacin and frenquel for six months after leaving hospital and then decided that she did not require them and discontinued both. A year and a half after leaving hospital, she is reported to be functioning well but is still hearing voices daily although they remain much less troublesome than initially. She is able to ignore them and to work steadily, and her physical health is good. She is sleeping soundly and, interestingly enough, her cataplexy has disappeared, while her "sleeping spells" continue but are so mild that she does not have recourse to amphetamine sulphate.

Comments on Case 1

In this girl, the paranoid schizophrenic break seems to have developed out of the vivid nightmares. The pre-psychotic picture shows a shy sensitive person who had denied any conscious interest in the opposite sex. The hallucinatory psychosis appears to assume a compensatory and perhaps wish-fulfilling function. Hallucinations are in the auditory sphere chiefly but are also visual, tactile and olfactory. As in Locke and Bailey's case, the patient shows more insight than is commonly found in schizophrenia but otherwise the picture is unmistakably what would be classified as schizophrenia in the present state of our knowledge. It is interesting that the narcolepsy and cataplexy were well controlled while the nightmares and then the psychosis increased in intensity.

The results of the physiological tests are discussed separately below.

Case 2

The patient, Kenneth R., aged thirty-six years was admitted to the University Hospital in March, 1957. There was a history of the narcoleptic-cataplectic syndrome of eighteen years' duration. During the past nine years he had been under intermittent psychiatric treatment for "depression", instability, a profound sense of frustration and occasional outbursts of rage.

The patient was born six weeks prematurely on a farm. He weighed five and a half pounds at birth and was said to be "black and blue". He experienced some feeding difficulty and when he was a few weeks old his mother developed pleurisy. He was his parents' first child and they had great hopes for him. Just over a year later, a second son was born completing the family.

The parental background was interesting. The paternal grandfather came to England from Germany in the 1870s. He was described as an unstable man and an "alcoholic". He married unhappily; his wife, the paternal grandmother, was a rigid, eccentric woman who became overtly psychotic during her sixties and spent the last twenty years of her life in a mental hospital. Years before she was hospitalized, she would write the patient's parents signing herself "The Countess". Kenneth's father had an unhappy childhood. There was much dissension in the home while at school he was scorned by other boys because he was a "German". He grew up with a chronic sense of inadequacy and resentment and this was increased by repeated occupational failures. Kenneth's mother was four years older than his father and apparently represented a protective figure to him. She had come from a somewhat higher social stratum and before marriage was a children's nurse.

The marriage of Kenneth's parents was not a happy one. His father farmed in Canada and met with frustration and failure. He showed resentment to his wife and was scornful of the children, calling them "sissies names" and making them feel inferior. The mother turned increasingly toward the children, assuming an over-protective role and cherishing great ambitions for them. She used to tell them in childhood that she wished them to become "professional men".

The patient and his brother were very close emotionally. Both were "delicate and fragile", and were "brought up like little Englishmen". This as might be expected did not help them to fit into life on the Canadian prairies. They were derided by other boys because of their accent and manners. They were exceedingly naïve sexually and thought that "children came from Gentle Jesus" and they were repeatedly humiliated by other boys because of this. On one occasion, and there were many such, Kenneth was held down by a girl while her brother made his younger brother eat manure. Kenneth was not surprisingly a shy, insecure and resentful boy. He suffered from recurrent nightmares and somnambulism and he grew up with strong ambitions for himself.

In adolescence he felt, if anything, even more insecure, especially in a sexual sense. He never had a girl friend although he claims he felt interested in girls. There were a number of reasons for this; he lacked both social skills and sexual knowledge and he was particularly sensitive to humiliation and rejection. In addition, the family was poor and he had no money with which to take out girls. He compensated to a considerable extent by working hard at school, but he had difficulties here also (despite an I.Q. of 127) because he was taught one thing at home and another thing at school. Thus he was taught one style of writing by his mother and another by the teacher and he found it very difficult to make a compromise.

In the spring of 1939 when he was nineteen years old and was in grade twelve, he developed "influenza". A few weeks afterwards, he noticed he would become increasingly sleepy. He also started having attacks of weakness. When overcome by some strong emotion, he would just fall to the ground in a flaccid heap. There was also a personality change which he called "a change of nature", and he became increasingly dull, depressed and irritable. Extensive neurological investigations then and subsequently disclosed no evidence of encephalitis. Between 1940 and 1941, he gained over seventy pounds in weight without noticing any

increase in appetite. At the end of 1941 his brother died of pneumonia while serving in the Air Force and Kenneth's loneliness was increased. He, himself, was not accepted for the services because of his illness and he continued to work at home on the farm. This gave the family fair financial support. In 1946, he began to notice a peculiar "indisposition". He would feel powerless to move although he wanted to. He emphasized, however, that it was a disturbance of volition and not a true paralysis. In 1948, because of his depression, irritability and failure to work efficiently on his parents' farm, he consulted a psychiatrist. Previously, he had taken up to sixty or seventy mg. of amphetamine sulphate per day without benefit. Between 1948 and 1952, he was treated by frequent psychotherapeutic interviews in an attempt to enable him to cope more adequately with his problems. During this time, he made an attempt at independence by seeking a job in Regina and supporting himself for a few months. But the improvement did not last and in 1952 he was admitted to the Psychiatric Ward of a general hospital because he felt deeply unhappy about himself and his illness. There he began to feel that he was being neglected. One incident upset him greatly. He confided in a student nurse whom he liked. Later he found that her brother was suffering from schizophrenia and had been admitted to the same ward as himself. He became extremely critical of the staff for allowing a relative of a nurse to be admitted to a psychiatric ward and his manner became increasingly truculent and aggressive. Eventually, he said, he discovered that the nurse whom he had confided in was having an affair with another patient. He apparently felt rejected and in a fit of rage broke all the windows of the sitting room. His conduct had now become so difficult to control that it was decided to transfer him on two medical certificates to the local mental hospital. He remained there for ten months and apparently preferred it, claiming that he had more status there, that the rules were more consistent and reasonable and that his condition appeared to be better understood. He did, however, exhibit peculiar ideas at this time believing for example at one time that he was to be castrated for his narcolepsy. However, close inquiry showed that a patient with similar symptoms had undergone an operation for an unrelated testicular condition. On another occasion, he claimed the staff were ill-treating an elderly patient and struck two female nurses. His behaviour at this time was considered to be quite paranoidal, and on his discharge from the mental hospital the clinical director, a man of wide experience, expressed an opinion that he might be suffering from paranoid schizophrenia. A careful perusal of the records shows however that there is no solid and unequivocal evidence to establish this diagnosis and it seems that the paranoidal features as exhibited were more the manifestations of a neurotic character disorder. This view is substantiated by the results of the Rorschach examination which was carried out then.

He returned to the farm in the summer of 1953 and remained essentially unchanged though perhaps rather more resigned to his condition. Because of the large dosage of dexedrine which he was once again taking, he decided to inquire if anything more could be done for his narcolepsy. As a result, he was referred to the Neurology Department of the University Hospital, Saskatoon, in February, 1957. He was transferred to the Psychiatric Department two weeks later for assessment.

He was treated by superficial psychotherapy and desoxyn 5 mg. b.i.d. which had been substituted for dexedrine. A number of investigations were carried out and are detailed below. He proved to be a deeply insecure man with neurotic paranoid trends and was preoccupied with feelings of humiliation, rejection and inferiority. He showed defensive grandiose trends. It was felt that his ego structure was rather weak and that uncovering psychotherapy might well precipitate a serious breakdown. His age and life situation would have made it difficult to effect any radical change in his character structure. It was quite evident that he had exhibited severe problems long before the onset of his narcolepsy but it was felt that the illness had greatly aggravated these trends. He did gain some relief from brief psychotherapy, talking freely about himself and his problems but seeking to dominate the interviews. It was evident that his feelings toward his mother and brother had been deeply ambivalent while he was frankly hostile to his father. He ventilated many feelings of hostility, inadequacy and inferiority. He had always felt a deep insecurity especially in a sexual sense and had compensated for this by grandiose plans. His difficulties were greatly intensified when the narcolepsy appeared. While the illness permitted an honourable withdrawal and dependency on his parents, it did not, of course, satisfy his needs for achievement and adequacy.

His symptoms diminished markedly while he was on the ward but he continued to take odd "naps" during the day and at times was aware of an indisposition to do anything. His sleepiness was markedly affected by external stimuli. Thus when he conversed with someone he liked he was notably brighter and more alert, though he would invariably have a bout of sleepiness at some time during the day. Discussion of emotionally disturbing material, however, did not appear to precipitate an attack.

He was discharged on 15 April, 1957. He had previously stated that he would feel worse at home. A few weeks later his mother had to write two letters for him because of his "indisposition". Yet while in hospital he had been able to write voluminously. The letters stated that he was finding it very hard to work on the farm.

Comments on Case 2

In this case, there were severe difficulties of adjustment prior to the onset of the narcolepsy. The patient felt emotionally isolated and inadequate. He

found it difficult to identify with the masculine role, and his relationship with his father and his schoolmates greatly increased this problem. The narcolepsy increased his difficulties of adjustment, accentuating the neurotic character trends and making it more difficult for him to follow the maturing process. As a result, he became almost wholly dependent on his parents while bitterly resenting this dependence. He adopted a grandiose irritable manner and rejected attempts to help him find a job commensurate with his abilities. Indeed with an I.Q. of 127 and a deep need to achieve something outstanding on the one hand and his emotional disturbance and narcolepsy on the other, a satisfactory solution of the problem was impossible.

As has been mentioned, it was felt that uncovering psychotherapy might increase rather than diminish his problems. Instead, an attempt was made to help him adjust to his handicap at a superficial level. He developed a strong dependent transference not unmarked, as might be expected, by deep ambivalence.

Physiological and Psychological Investigations

Routine examinations of blood, urine and basal metabolism showed no abnormality.

EEG Studies

The EEG examinations were carried out on a Grass Model 3D eight-channel apparatus. For the parallel E.C.G. examinations, standard lead III was used.

Both cases showed the characteristic short bursts of drowsy EEG activity; there was no evidence of any other abnormality.

When given insulin intravenously, both patients showed remarkable psychological and EEG changes. Hoffer (1957) has been studying the effects of hypoglycaemia on the EEG in an effort to study homeostatic mechanisms. One unit of insulin is given i.v. per eight pounds of body weight. Usually, but not invariably (Ziegler and Presthus, 1957), there is a tendency for slow wave activity to appear (Wyke, 1952). In Case 1, the blood glucose fell over 45 minutes to 38 mg. per 100 ml. During this time, the patient became more alert and the EEG patterns tended to normalize, the sleep activity diminishing. In Case 2, blood glucose fell to 39 mg. per 100 ml. Once again, the patient became more alert and the EEG pattern showed diminished sleep activity. In both cases, drowsiness returned a few seconds after 50 ml. 33 per cent. intravenous glucose was given. The increased alertness with hypoglycaemia was most striking and no clouding of consciousness was present. The EEGs for Case 1 are shown in Figures 1–4. The blood-glucose levels are indicated.

Glucose Metabolism

The Exton Rose Glucose Tolerance Test was used with the following results. These results, being unusual, were checked carefully.

		Case 1 Blood Glucose mg./100 ml.	Case 2 Blood Glucose mg./100 ml.
8.45 a.m.	(fasting)	156	97
9.00 a.m.	50 gm. glucose given		
9.30 a.m.	.. ..	133	134
	50 gm. glucose repeated		
10.00 a.m.	.. ..	82	172
12.00 a.m.	.. ..	116	113
2.00 p.m.	.. ..	113	62

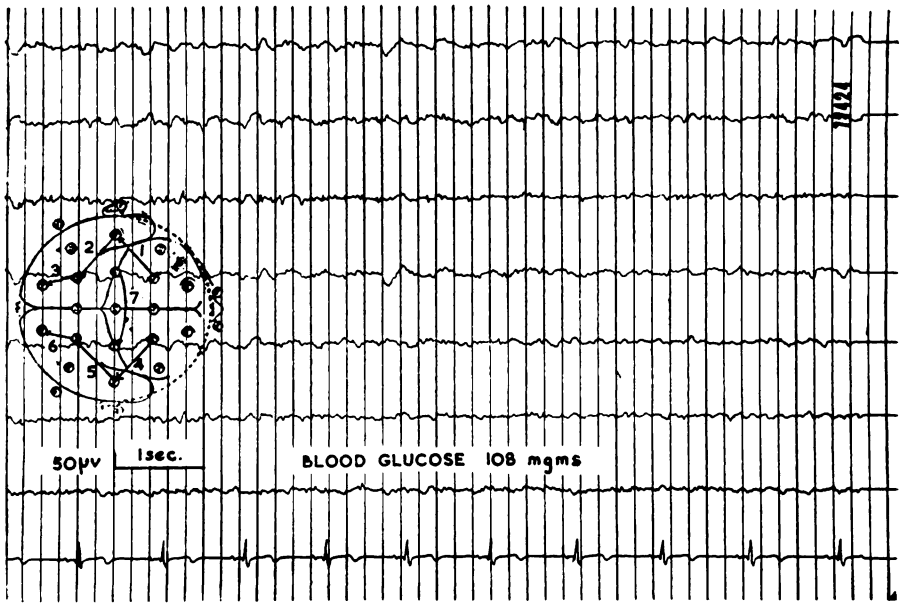

FIG. 1.—EEG on Case 1 immediately prior to injection of 17 units soluble insulin I.V. Time: 8.45 a.m. Note considerable drowsy pattern. Blood glucose level is 108 mg./100 ml.

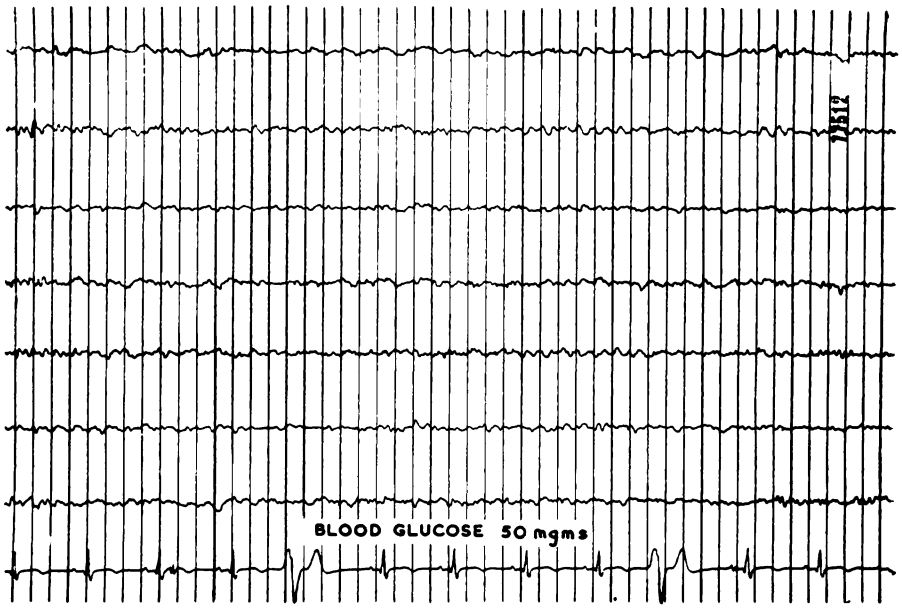

FIG. 2.—Case 1. Leads as in figure 1. Time: 9.10 a.m. Blood glucose 50 mg. There is a little drowsiness.

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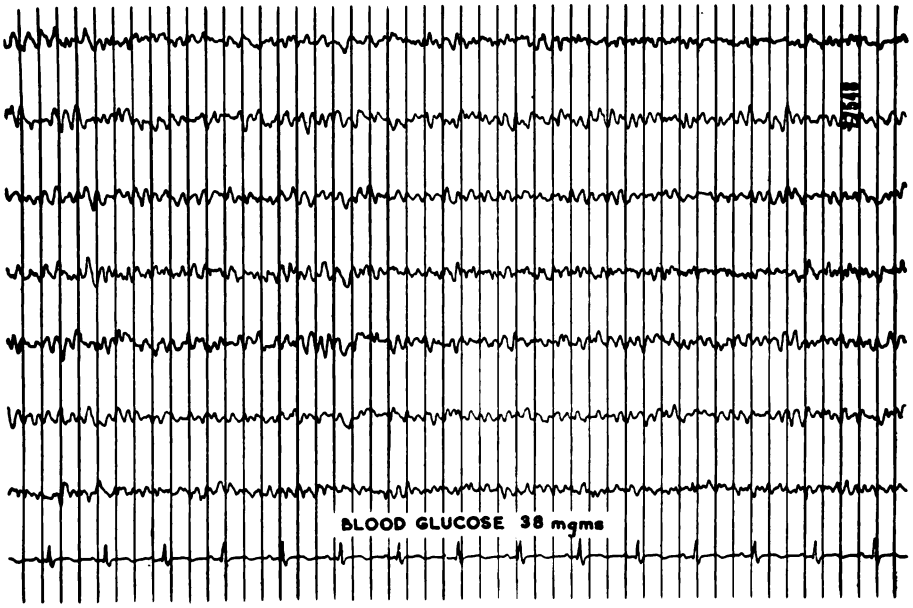

FIG. 3.—Case 1. Leads as in figure 1. Time: 9.25 a.m. Blood glucose is now 38 mg. There is very little drowsiness and alpha activity appears fairly well maintained.

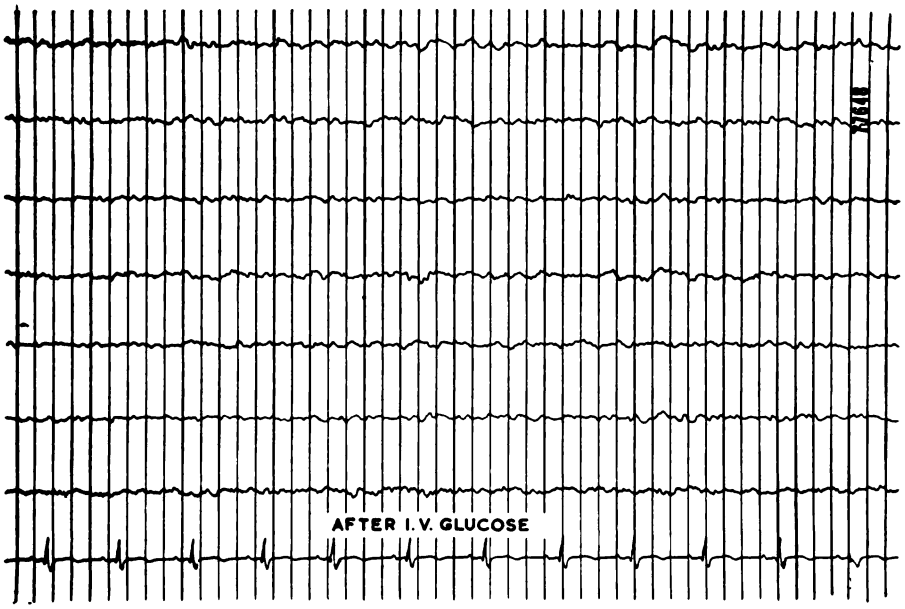

FIG. 4.—Case 1. Leads as in figure 1. Time: 10.0 a.m. 50 c.c. of 33 per cent. glucose has been given I.V. The drowsy pattern is markedly increased.

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Response to Autonomic Drugs (1954)

Hoffer in studying the effect of para-sympathetic activity on sleep has been using the following combination of drugs: atropine, 3 mg. i.m. followed by physostigmine, 2 mg. i.m. one half hour later; finally 200 mg. acetylcholine is given i.v. after a further 15 minutes. After the injection of acetylcholine, the subject may often feel sleepy and show sleep activity on the EEG. The results in the two subjects were not striking though they both reported some slight increase in alertness following acetylcholine.

Both subjects were given Funkenstein's mecholyl test. Neither re-established homeostasis in 30 minutes. Case 1 showed a "hyper" type of reaction (but the E.C.T. given a few weeks previously may have affected this result). Case 2 showed a "hypo" reaction, the blood pressure remaining below the initial base line.

Methedrine, 30 mg., was given on a separate occasion to Case 1. A striking activation of the psychosis occurred and subsequently auditory hallucinations bothered her all night. Case 2 was given adrenaline by inhalation. He claimed that the drug relieved his drowsiness for about one hour but to produce this effect he had to inhale 500 micrograms. He was also, on another occasion, given 1 mg. of physostigmine salicylate intramuscularly; this made him feel sick but had no effect on his drowsiness.

Psychological Investigations

The report of the Rorschach examination on Case 1 (Mr. W. J. Craig) stated: "This is a schizophrenic type of record. Scoring in Piotrowski's (1950) objective manner, five signs of C.N.S. disorder are found. Score for formula alpha was three and the test was within validation limits indicating schizophrenia rather than neurosis. Of the four signs differentiating schizophrenia from organic conditions, the patient has two favouring schizophrenia. The prognostic indication gave a score of five which predicts little change or perhaps some improvement in the schizophrenic condition over the next three years." It was felt that the E.C.T. previously given would account for the organic signs. The M.M.P.I. yielded the following profile:

Scale	L	F	K							
Raw Score	5	4	15							
Scale	Hs	D	Hy	Pd	Mf	Pa	Pt	Sc	Ma	Si
T Score	50	77	57	43	55	70	78	74	40	78

The Rorschach report (Mr. W. J. Craig) on Case 2 was as follows: "This patient has adequate control but shows depression and anxiety plus considerable fluctuation in mood. He prefers life to be well organized and secure and in this setting would like a dependent role. He is uncertain of his masculinity and his father relationship is seen as hostile." The M.M.P.I. profile obtained was:

Scale	L	F	K							
Raw Score	2	10	12							
Scale	Hs	D	Hy	Pd	Mf	Pa	Pt	Sc	Ma	Si
T Score	59	94	71	66	61	56	80	96	58	48

Discussion

In both cases, the illness had had striking repercussions on the life of the patient. In Case 1, adjustment before the illness was somewhat precarious, the personality showing obsessional trends and sexual feelings being denied. When the psychotic illness developed, apparently as an extension of the sleep hallucinosis, the content consisted of material which had never gained conscious expression. In Case 2, the previous adjustment was poor and the illness facilitated regressive trends already present. This patient's desire for independence, maturity and achievement remained unsatisfied, however, and severe conflict remained.

The question arises, of course, whether the narcolepsy arose solely as a response to the pre-existing conflicts. While this cannot be answered definitively, there are a number of points against a purely psychogenic aetiology. The results of the physiological data suggest some underlying metabolic dysfunction. The peculiar alerting response to hypoglycaemia is quite unusual. Excitement is, of course, common with hypoglycaemia, increased alertness is not; in fact, hypoglycaemia on occasion has simulated narcolepsy (Delay, 1942). It has been suggested that narcolepsy is a reversible disturbance of the reticular activating system (Daly and Yoss, 1957) and it is interesting to note that the reticular system contains components sensitive to acetylcholine (Rinaldi and Himwich, 1955a, 1955b) and adrenaline (Rothballer, 1956). A possible explanation of the alerting effect with hypoglycaemia could be a central liberation of adrenaline with stimulation of the adrenaline sensitive part of the reticular activating system. A disturbance of this system might also explain the beneficial effect of the sympathomimetic drugs in narcolepsy. Another possibility is that the rise in free glutamate which occurs in hypoglycaemia (Himwich and Sullivan, 1956) may have an ameliorating effect on the narcolepsy. The atypical responses to the Exton Rose Test also suggest that factors other than psychological are implicated. Again, the sudden marked gain in weight (without increase of appetite) in Case 2 would be consistent with involvement of the hypothalamic brain stem region. Finally, if the sleep attacks are regarded as ego defensive manoeuvres, the question still arises as to how this particular defence comes to be selected. So far, purely psychological studies have shed no light on this problem.

There are additional general considerations which would favour the existence of some underlying organic disturbance. First of all, the characteristic short bursts of drowsy activity seen on the EEG often come and go suddenly without any apparent relation to stress. This was certainly true in both the cases described. Secondly, skilled clinicians with wide experience of narcolepsy have not been struck by the neurotic trends of these patients (Adie, 1926, 1930; Cave, 1931; Daniels, 1934) and it is possible that the cases reported by psychiatrists may represent a biased example of the total narcoleptic population. Thirdly, other features suggestive of hypothalamic involvement have often been reported in narcolepsy (sudden changes in weight, hypogenitality, disturbance of carbohydrate metabolism, etc.). Fourthly, it may be pointed out that in the psychosomatic disorders, it is usually held that, in addition to the conflicts involved, some specific additional factors must be invoked (Alexander, 1952); similar considerations would apply to narcolepsy.

Psychological factors are certainly relevant however to the expression of the condition. This is obvious enough in relation to the cataplexy and also frequently to the attacks of sleepiness, as was observed previously. It has been

noted repeatedly too that factors normally producing boredom and fatigue in normals act with increased force in narcoleptics inducing attacks of somnolence. Finally, it is clear that the illness may present the patient with serious problems of adjustment to his altered function and role and that psychotherapy may be indicated.

The relationship of the psychosis which rarely develops in narcolepsy to schizophrenia must remain obscure while so little is known about both disorders. It appears, however, as if the psychosis may either develop in intimate connection with the sleep hallucinosis (the more common way) as in Case 1 or else independently and perhaps fortuitously.

Summary

The literature relating to narcolepsy and associated emotional disturbance is briefly reviewed. The rarity of associated overt psychosis is pointed out.

Two cases of idiopathic narcolepsy are then presented. The illness in the first case was accompanied by paranoid schizophrenia, in the second by a neurotic character disorder.

Unusual neurophysiological findings are presented. The chief of these was a marked increase of alertness following hypoglycaemia. The possible significance of this and other findings is discussed.

Finally, it is suggested that psychological factors have a modifying influence in the narcoleptic phenomena but probably do not act as a necessary and sufficient cause of narcolepsy. The importance of psychotherapy in some cases is stressed.

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