Short Notes and Clinical Cases.

REPORT OF A CASE OF INSOMNIA FOLLOWING ENCEPHALITIS LETHARGICA.

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ALTHOUGH cases demonstrating the various sequelæ of encephalitis lethargica have been recorded in vast numbers in England, yet among them there do not seem to be many with marked insomnia and nocturnal irritability as predominant features. Under the circumstances, the publication of the details of such a case does not seem to be out of place.

C. H., age 8, no previous illnesses, began in April, 1920, to develop drowsiness, manifested by sleep when he sat down, although previously he had been a bright and lively boy. His appetite began to fail and his sleepiness persisted; the patient sleeping immediately after dinner on the night following the commencement of his illness. The following morning, while the patient was asleep, his mother noticed both lower limbs become alternately flexed and extended, the right upper limb adducted and abducted, recurring attacks of twitching of the right side of his face, and eyes which seemed to roll about under half-closed eyelids. She woke him up several times, and spoke to him, but he appeared to be too drowsy to answer, and went to sleep again after the cessation of stimuli. This apathy extended to food, for he hardly ate anything that day. At night he seemed feverish and was given a dose of castor oil. He slept quite well despite the continued twitchings of the right upper limb and the occasional jerkings of both lower limbs.

The following day he was brought to the hospital—the right upper limb still twitching—and, complaining only of a sore throat, was diagnosed as a case of tonsillitis or early scarlet fever, and was sent home with instructions to report himself if a rash appeared. He remained drowsy throughout the day, and vomited once. During the next day drowsiness still persisted, and the arm-jerkings became worse, his upper arm nearly touching his chin during the movements. He was seen a day later, and, the anticipated rash not being present, was sent to an infirmary, where he remained from April to November,

the drowsiness persisting for nearly five weeks. At that institution he was diagnosed as a case of chorea.

Soon after the patient's admission to the infirmary, the twitchings of the lower limbs ceased altogether, whereas those of the upper limb continued unabated; and about three months after the onset of the illness, i.e., about July, 1920, the observant mother once again noticed a change, in that the neck muscles seemed to be weak, the head falling forward and the chin practically resting on the chest. To counteract this weakness a high collar was worn.

During his period of drowsiness, it was with great difficulty that he could rouse himself to answer questions. Even then his reply was an unintelligible sound, and he made no spontaneous remarks. Every little action seemed to be too much trouble.

In August, 1920, when all signs of drowsiness had long been left behind and the patient was running about the wards quite fit, eating well, and putting on weight, there came a remarkable reaction. He could not get to sleep at night. He tumbled and tossed, sat up in bed, played with his toys, and occasionally tried to stand on his head; and it was not till 2 or 4 a.m. that at last sleep came. This state of affairs persisted throughout the remainder of his stay at the infirmary; and at one time the proposition of an operation for cerebral tumour was seriously considered. Thus he continued, too, while he was at a convalescent home for a period of three weeks. From there he returned home, and his mother, describing his first night, said that he started at each sound, and twitched and turned during the whole of the night. He seemed to be in a kind of stupor, being neither asleep nor awake, with both eyes open. He answered when spoken to; and throughout the night he made repeated attempts at blowing out an imaginary candle. That night, too, he had incontinence of urine. He was admitted to Guy's Hospital on April 22, 1921.

On Admission.—The patient is rather small for his age, but quite sturdily built. His gait is peculiar and simulates that of a man with paralysis agitans. His head seems poised rather too forward on his body, and is held rather rigidly in that position, his neck muscles being in a state of hypertonus. There is little expression on his face with the exception of a transient smile which now and again plays about his lips. The upper limb is kept flexed at the elbow and wrist, and the fingers, too, are kept in semiflexion. There is no automatic movement of the limb on walking, the only movements being involuntary twitchings, which are exaggerated when the patient is asleep, and which will be described later. His left arm swings normally. His steps are short and rather rapid, but there is no tendency either to propulsion or retropulsion. On being addressed, the patient will look at his questioner with eyes raised, his head being

kept fixed. His speech is staccato in character. Mentally he is quite bright, though temperamentally he is somewhat nervous.

Examination of Central Nervous System.—

Cranial Nerves.—

I. The patient is able to distinguish different smells.

II. The pupils are equal in size, and react to light but not to accommodation—the reverse of the Argyll-Robertson pupil. The visual fields and the fundi are quite normal.

III, IV, and VI. Eye movements normal in degree and equal in extent. There is definite nystagmus on looking to right and to left, the latter movements being coarser, slower, and of less extent than the former.

VII. There is definite though slight facial weakness on the right side, which weakness becomes intensified and much more noticeable on emotional movements such as laughing, smiling, and yawning.

No abnormality can be detected in the other cranial nerves, except that the tongue, which is very tremulous, deviates slightly to the right when protruded.

Musculature.—The muscles of the right upper limb are in a state of tonic contraction of slight degree as compared with those of the left side. The right upper limb is held flexed at an angle of 150° at the elbow and at the wrist; and there is semiflexion of the fingers, with the thumb approximated to the index finger and the forearm in a semiprone position. This limb is periodically adducted and the fingers flexed through the clonic contractions of the following muscles :—

1. Pectoral group of muscles ; 2. Latissimus dorsi. These muscles are most pronounced in their contractions, which can readily be seen and felt.

3. Biceps brachii. Tendon can be palpated readily in the antecubital fossa.

4. Pronator radii teres, which keeps the forearm in the semi-pronated position.

5. The flexors of the fingers and wrist. The fingers and thumb can readily be seen to contract clonically at the metacarpophalangeal and interphalangeal joints.

6. The rhomboid muscles. Their action in drawing the scapula inwards can be appreciated by placing the hand on the superjacent skin.

Other muscles most probably play their part, too, but it is very difficult to be certain. Of these, the small muscles of the thumb and of the little finger, the muscles of the hand, and perhaps the supinators, are the most important.

These movements seem to be due to clonic contractions of the above-mentioned muscles, and there does not seem to be alternate contraction of their antagonistic muscles. No contractions can be detected in the deltoid, triceps brachii, or extensor muscles of the hand. The rate of the contractions varies between 80 and 90 per minute, being greater at times of emotional stress than under normal conditions. No fibrillary twitchings could be detected.

On testing both grips, it is found that the right grip is definitely the weaker, and the flexors of the forearm and the adductors of the upper arm are also slightly less powerful than the corresponding muscles of the opposite limb.

Reflexes.—The reflexes of the upper limbs are present, though diminished in extent, and equal on the two sides. The abdominal reflexes are brisk and equal. The knee- and ankle-jerks are brisk on both sides, though slightly more so on the left. The plantar reflex is definitely flexor on the right side, whereas a doubtful extensor is obtained on the left side.

Cutaneous and deep sensibility and sense of position are all normal; as are the cardiovascular, respiratory, alimentary, and genito-urinary systems.

The patient's first night at hospital was typical of practically all his nights. At 10.30 p.m., when all the other patients were asleep, he simulated a boy suffering from a very bad attack of chorea. With eyes practically closed, and unconscious of all external surroundings, he wriggled about in bed, lying first on one side and then on the other. Shooting out both legs alternately, thus kicking the bedclothes from off him, he would pursue his manœuvres covered by a nightshirt only. At times he would be in a position of opisthotonus, followed shortly afterwards by the praying attitude of the follower of Islam. At times his movements would become so violent that he would bump his head against the sides of the bed, and it required a very vigilant nurse to prevent his falling on to the floor. During the whole of this period the twitchings of the right upper limb would continue unabated, and after a varying interval the patient would suddenly become conscious of his environment, sit up in bed, and smile. Incontinence of urine was a not uncommon accompaniment.

Attempts to determine reflexes, especially the plantar reflexes, were made during sleep, but were very soon followed by violent movements, making further tests impossible. As the result of many such attempts, I am inclined to say that the right plantar reflex is definitely flexor, and the left most probably extensor.

The patient was warded for over a month, and despite the fact that an undisturbed night's rest was a rare event, he seemed to suffer

no ill effects, beyond the fact that he felt sleepy in the morning, which is not at all surprising, as he did not start his night's rest till 2 a.m. Throughout the day he would be quite cheerful and run about the ward, but, if he were given an opportunity to lie down, he would soon be asleep and would sleep well, the clonic twitchings of his right arm being the only movements present.

Treatment was very disappointing. As a first resource hypnosis was attempted. This proved tolerably successful the first night, less so the next, and finally it had to be given up, as the patient would not keep still for any length of time. Following this, medication by chloral and bromides by night and increasing doses of hyoscine hydrobromide by day was tried, with somewhat better results. If the chloral were given in the early hours of the evening, the period of nocturnal activity seemed to be diminished, but the twitchings were not appreciably affected by the hyoscine. Leahy and Sands¹ in America reported a number of similar cases in which they found that "a warm pack in the milder cases and a wet pack in the more disturbed, was conducive to sleep and allayed some of the irritability. Hydrotherapy and massage were also utilized freely." Hydrotherapy was not employed in this instance, because that therapeutic measure was found to be unavailing in similar cases. I examined the patient again two months after his discharge from hospital, and he was not one whit the better. There must be many such cases in the country, and one wonders what is the ultimate prognosis.

In conclusion, I wish to express my indebtedness to Dr. Symonds, under whose care the patient was in hospital, for his permission to publish the details of the case.

REFERENCE.

- ¹ LEAHY, SYLVESTER, R., and SANDS, IRVING J., *Jour. Amer. Med. Assoc.*, lxxvi, No. 6.