MAJOR HYSTERIA WITH ATTACKS OF IMPAIRED CIRCULATION OF THE LEFT UPPER LIMB.*

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THE case reported in this paper is of importance because of attacks of grave hysteria occurring after severe head trauma and associated with attacks of disturbed circulation of the left upper limb. It leads to a consideration of some of the peculiar clinical conditions attributed to the thalamic and striate regions. The extraordinary periodic alteration of the circulation of the left upper limb in this patient is an unusual phenomenon and probably was partly under the control of the will.

The patient, male, 19 years of age, was referred to me by Dr. A. C. Trapoid, of Wilkes-Barre, April 28, 1927.

CLINICAL HISTORY.

In February, 1926, the boy was struck from behind by some unknown assailant while returning from his daily work as a stenographer. He laid unconscious in the snow for a period of two hours before regaining consciousness. He was then taken home in an automobile, but was able to walk and complained only of a slight headache. He ate supper and retired. The next morning he complained of a severe frontal headache, felt dizzy, fell over unconscious and awoke in bed about an hour later. He was sent to the hospital complaining of severe headache in the top of the head. This gradually disappeared and he was well at the end of ten days except for numbness and swelling of the left upper limb. He could raise the upper limbs in all directions, but he tired easily. The numbness disappeared at the end of six weeks.

Following this period he was well until June 1926, when he was struck on the head by a falling baseball. He was temporarily stunned but did not lose consciousness. The next day while at work he had what he termed a "weak spell," which lasted about one half hour. In this attack he fell over unconscious, but did not have convulsive seizures. He complained of severe headache (apical and frontal) for two days. After this he was in good health until April 16 1927, except that since his original injury he had frequent swelling of his left upper limb; the limb turned a purplish colour and was cold from the elbow down. He seems to have had no such attacks before his injury.

On April 16, 1927, he arose as usual, felt perfectly well, but while eating breakfast complained of severe frontal headache, later becoming occipital. In attempting to pick up his lunch basket from the table it fell to the floor and

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on stooping over he walked several feet and fell forward on the floor. He was unconscious for several hours but had no convulsive seizures, until he was apparently regaining consciousness, when twitchings of the muscles were noticed over the entire body. Following this he had similar convulsive seizures at short intervals in which his head and body assumed a position of opisthotonos. He had no further seizures from 8 p.m. until 9 a.m. the following morning, when similar attacks occurred at very short intervals; in these his arms and legs were extended, accompanied by slight jerking and marked retraction of the head. His father states that during an attack the left arm and forearm became markedly reddened and swollen, and the veins of the forearm stood out very prominently. Upon recovering from this seizure he complained of severe frontal headache, changing to occipital. During the attacks he never frothed at the mouth or bit his tongue, and he had no incontinence of urine or faeces.

DESCRIPTION OF ATTACKS.

Note of observation by myself, May 23, 1927.—He has been having attacks about every five minutes. The attack begins with simultaneous fine tremor of the fingers of both hands, tremor of the eyelids of each side, followed almost immediately by great spasticity of all four limbs. The fingers of each hand are over-extended; the feet are contracted into the position of talipes equinovarus. The body is arched in opisthotonos, and towards the end of the attack there may be one severe jerking of the trunk upward in more pronounced opisthotonos and the attack is over. The entire attack lasts scarcely more than one minute. Towards the end of the attack there is jerking of the muscles about the mouth and in the cheeks. In the intervals between the attacks the muscles are very relaxed and the man is slightly cyanosed. His pulse is rapid and his blood pressure is 165/80. The attack frequently ends with a violent contraction in opisthotonos, followed by sudden relaxation, so

Photograph taken during a convulsive attack. It shows the opisthotonos, the overextension of the fingers, and the position of the upper limb most favourable for the circulatory arrest.
(From a moving picture film made by Dr. A. M. Ornstejn.)

that the body falls heavily on the bed. He has repeatedly come out of the attacks suddenly with complete consciousness, and then suddenly with a severe extension movement of his upper limbs has returned into an attack. The moderate cyanosis after an attack seems to be particularly marked in the distal parts of all four limbs; the distal parts of the lower limbs are moist and cold. This is different from the cyanosis of the left upper limb alone. Dr. Loe, my interne, observed that the pupils were dilated markedly and seemed to react very slightly to light. The attack is typically one of major hysteria.

We attempted to bring on a vasomotor discoloration of his left upper limb by suggestion, placing a hypodermic needle under the skin of the left arm, but it had no effect even with repeated suggestion. Pressure was made behind and below the tip of the right mastoid process. This spot is a hysterogenic zone and corresponds to the place where the man received the blow. Pressure immediately produced an attack exactly like those previously described, but the opisthotonos was more pronounced, and was so extreme that the top of the head rested on the bed. The arching of the body once or twice assumed a forward type which was never very pronounced, but the lower limbs were forcibly flexed at the hips to a considerable degree and then suddenly allowed to fall.

The patient said that he was told on May 21 that as he had had no attacks since he came to hospital he might as well go home. He was overjoyed by this and was thinking very much about returning to his home. Two days later he began to have the convulsive attacks described above. They have been very frequent and are all of the same type.

The eyes were examined and reported negative. The blood Wassermann was negative in all antigens. The head was reported negative by X-ray examination, and X-ray examination for cervical rib showed no evidence of this anomaly.

Results of examinations made by my interne, Dr. J. W. Clark, July 5, 1927, on the condition of the blood pressure and of the pulse of the upper limbs during an attack of congestion of the left upper limb, testing the limbs alternately and quickly, are as follows:

Blood Pressure.

Right Upper Limb	Left Upper Limb
130/86	90/54
132/90	90/54
132/96	80/45
131/95	128/78
130/100	124/85
130/75-Later	122/85

Pulse.

86.

86.

FURTHER CLINICAL OBSERVATIONS.

On May 30, 1927, I had the opportunity of seeing one of the attacks of cyanosis of the left upper limb. The discoloration was very intense. The whole left upper limb was a reddish purple which shaded off into the normal appearance near the axilla. The man believes, and his belief seems to be well-founded, that the attack is more likely to develop when he is lying on his right side and his left upper limb is fully extended. The attack this morning seemed to be distinctly influenced by this position and has lasted during the entire time in which this position was maintained, but this is the position of the upper limbs during the convulsion. Blood pressure observations made during periods of relaxation and unconsciousness revealed the following:—

Left upper limb blood pressure 156/80 ;

Right upper limb blood pressure 134/62.

The blood pressure was less in the left upper limb during an attack but greater in the period of relaxation following.

The patient assumed the position necessary to induce an attack of cyanosis and swelling in the left upper limb. The required position is as follows: he lies upon his right side and allows his left upper limb to be extended either along his body or a little behind his trunk. From near the elbow the limb became a deep reddish blue colour. The depth of colour increased towards the fingers. The left hand and forearm appeared to be swollen; the skin was tense. The biceps muscle was apparently in unusual tension. The veins of the hand, forearm and upper arm were distended with blood; those of the forearm stood out prominently.

In one unusually severe attack the following observations were made. Right radial pulse 88; Dr. J. W. Clark thought that he obtained the left radial pulse once and counted it to be 88. However, upon further repeated observations he was unable to detect the pulse at the radial artery or in the brachial artery at and above the elbow in the left upper limb. Blood pressure right upper limb 126/82. In the left upper limb no sounds were audible upon repeated attempts. The observer caused the mercury to rise to at least 180 mm. and listened carefully down to 30 mm. When the left upper limb was elevated and the attack was passing off, he noted that the left hand and fingers felt quite cold and clammy. The temperature of the forearm and arm seemed to be normal. There was no apparent abnormality in the right upper limb. In the upper arm, as the normal colour returned, there remained numerous little punctate areas of discoloration, as though the areas around the hair follicles were the last to lose it. About twenty minutes after the attack the following observations were made. Blood pressure, right upper limb, 126/84; left upper limb, 112/70. The auscultatory sounds incident to the reading of the blood pressure in the left upper limb were at the time of the reading 112/70 relatively faint. Those in the right upper limb were loud. When the blood pressure was taken in the left upper limb ten minutes later it was 126/80. The

auscultatory sounds in both arms were then equal. Radial pulse, right upper limb, was 72; left upper limb, 72. The two pulses were synchronous. Dr. Clark reported that at first practically none of the attacks of unconsciousness were accompanied by discoloration of the left upper limb, whereas during the past two or three weeks about 75 per cent. of the attacks of unconsciousness were accompanied by a variable discoloration of it.

August 3, 1927. The patient says the cyanosis of the left upper limb is produced when he puts "stress into it," as in eating or typewriting; he does not seem to have it unless he increases the tonicity of the muscles of the limb. He has it in typewriting when he strikes the keys hard in making several carbon copies. The cyanosis suggests a mechanical origin, as though caused by contraction of muscles of the upper arm interfering with the circulation. He has no power of doing this in his right upper limb.

DISCUSSION.

Rudolf Schindler¹, in a recent monographic study of the relation of the nervous system to spontaneous hæmorrhage, concludes that small cutaneous ecchymoses are very frequent in hysteria and in some cases these ecchymoses may be enormous. The condition is curable by psychotherapy. If ecchymoses may occur as a part of hysteria, as we now understand this disorder, cyanosis would seem to be possible also as a result of hysteria, and then the question arises whether both hysteria and ecchymoses or cyanosis may not be the result of thalamic, subthalamic or substriate lesions. We failed entirely to produce the cyanosis of the left upper limb by suggestion.

Kinnier Wilson² has mentioned the resemblance of grave hysterical convulsion (*arc de cercle*) to certain symptoms produced by mesencephalic transection.

In my paper³ on subcortical epilepsy I referred to the possibility that the striate body may be an important part of the sympathetic supply, and may contain vasomotor centres. It is possible that certain manifestations of epidemic encephalitis which have been regarded by some as suggestive of hysteria may be related to the thalamic or subthalamic region, and vasomotor disturbances occur in epidemic encephalitis which affects this part.

Cases such as those reported by Sokolansky⁴ are very difficult to interpret. One patient, for example, had definitely had epidemic encephalitis followed by Parkinsonism. He had frequent attacks of the following character: he complained in an attack of pain about the heart, of a feeling of great anxiety; he longed to die and begged for poison. This emotional disturbance ushered in an attack. Tonic spasms of the eyes then caused the eyeballs to be drawn upward—a condition frequently observed in epidemic encephalitis. These attacks were followed by severe myoclonia of the whole body. Saliva flowed, perspiration was free, and the pulse was between 110 and 120 per minute. The patient under ordinary conditions did not have the power to raise himself in bed, but in the attack he sprang from the bed with extraordinary speed and

ran along the corridor, but would fall. He attempted to bite or spit upon anyone who came near him. During the attack consciousness was preserved, his behaviour was impulsive or emotional, he acted contrary to his will, he said he was controlled by some evil force. He begged to be fastened to the bed as the attack was coming on, and during it warned all to keep away from him. The emotional element in these attacks suggests the optic thalamus. The attacks were thought possibly to be the result of some biochemical process affecting the basal ganglia.

Foerster⁵ states that the optic thalamus is under the control of an inhibitory tract from the cortex and also of one from the corpus striatum. When these tracts are destroyed in lesions of the thalamus a very limited irritation of some part of the body may cause an over-reaction referred to the entire half of the body on the side of the lesion (hyperpathia); in some instances it may be referred to the entire body. These phenomena are seen in highly neurotic individuals. In this way also the over-reaction from stimulation of special senses in thalamus lesions, so that this stimulation produces pain, is explained, for all sensations and special senses depend on the integrity of the optic thalamus.

Accepting these statements made by Foerster we may assume that in a case such as that reported by Sokolansky the emotional disturbances were caused by lesions of the optic thalamus resulting from epidemic encephalitis.

Shall we conclude that the convulsive attacks in my patient were the result of the severe head trauma and dependent on such a lesion (possibly hæmorrhagic), as that producing mesencephalic transection, and that the cyanosis of the left upper limb was caused by striate lesions, or shall we seek further for an explanation? The convulsive attacks were undoubtedly influenced by suggestion.

That hysteria may be associated with epidemic encephalitis is shown by Marinesco⁶, as he was able to remove the hysterical element by psychotherapy. He says his observations demonstrate that epidemic encephalitis favours the appearance of hysteria because a relation exists between the mechanism of these two pathological states, and that recently Binswanger reported a case of traumatic hysteria with the appearance of postencephalitic Parkinsonism. Marinesco reports a case of epidemic encephalitis with Parkinsonism, also with hysterical hemiplegia and hemianæsthesia and other hysterical signs, cured by suggestive and electrical treatment under the form of torpillage, but the encephalitic manifestations persisted.

There is an explanation for the cyanotic attacks in the left upper limb of my patient which demands attention. Dr. J. B. Wolfe has referred me to a paper by Hyman⁷ on voluntary pulse control in the radial artery at the wrist. Hyman has seen three patients in 1926 who had the ability to regulate or stop their pulse at will. He could find nothing on the subject in the literature as late as 1926. It was known to physicians during the world war that certain soldiers attempting to malingering could alter the activity of their radial pulse

by contracting the adductor muscles of the shoulder joint when a rolled towel or similar object was placed in the axilla, and in this way make pressure on the axillary artery.

Deliberate interference with the pulsations in the axillary artery has been noted in certain individuals with 'loose joints'; in such persons extreme rotation of the humerus winds the artery round the bone, with subsequent compression of the vessel.

A more subtle mechanism is that wherein the pulsations in the radial artery are controlled by voluntary muscle contraction either consciously or unconsciously on the part of certain persons who possess unusual neuromuscular reflexes. Hyman examined a man who could "will" his radial pulse to become smaller, and if he "willed it hard enough" he could stop the left radial pulse for about fifteen seconds without appreciable effort. The heart rhythm and the pulsation of the right radial artery were not affected. The "block" was located by Hyman in the left subclavian artery where the artery passes over the first rib and under the insertion of the scalenus anticus muscle. Examination under the fluoroscope showed a small movement of the soft tissues lying under the outer end of the clavicle. The patient was not aware of muscle movement but he experienced muscle fatigue in the deep neck muscles of the posterior group.

Two explanations are regarded as possible: the first, that the subclavian artery perforates the scalenus anticus muscle and because of its peculiar development this muscle exerts a sphincter-like action when contracted, shutting off the pulse wave in the artery. The second possibility is that the left subclavian artery has an unusually well marked isthmus which is readily constricted when the scalenus anticus muscle in contracting presses it against the tubercle of the first rib. The longest period Hyman's patient could "block" the radial pulse was 18.6 seconds.

Since this paper was read an interesting study on the diagnosis between hysteria and extrapyramidal disease has been published by Rothfeld⁸, with references to the literature.

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