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BY

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ECHINOCOCCUS MULTILOCULARIS OF THE BRAIN.

By Henry H. Mudd, M.D.,
ST. LOUIS, MO.

Swelling over right Rolandic region; hemiparesis, with tremor; left-sided hemianopsia; choked disk; removal of an echinococcus cyst; hernia cerebri accompanied by high temperature; disappearance of both by pressure; complete recovery.

D. A., an unusually large and well-developed girl, aged twelve years, presented herself, in March, 1891, for treatment. At my request she entered St. Luke's Hospital. Her parents stated that she had always been a robust, strong-willed, and somewhat irascible child. The first evidence of her present trouble was a severe, diffuse headache, which commenced in the latter part of July, 1890. This continued for several weeks. The child would frequently stop playing, lie down until the pain was easier, and then get up to resume her play. She was not confined to bed an entire day during the continuance of the headache.

In the first week of October a small elevation of bone, about the size of a silver quarter, was observed above the right ear. The headache disappeared before the tumor was noticed. The swelling by firm pressure could be indented. On ceasing pressure it returned to its place with a snap, like thick, elastic paper or the bottom of a tin pan when pressure is alternately made and released.

Before this swelling appeared the left hand and arm twitched and trembled. The parents stated that the tremor was confined at first to the hand. Soon after this tremor was noticed the mouth during laughter was drawn to the right side. Later both the head and leg began to twitch and tremble. Thus, according to the history, the twitching commenced in the hand, became diffuse, involving the entire left side, the face, head, neck, and leg becoming implicated. The mother asserts that the walk is better now than it was during the early winter, the twitching in the leg being also much less.

Present condition (March 14, 1891): The child has quick perception and good reasoning powers. There is an active twitching of the left side of the body. This tremor is rarely noticed in the facial muscles, but is pronounced in the platysma and the neck. The head is turned slightly to the right. The left angle of the mouth is depressed. The facial muscles on that side are sluggish and feeble. The impairment is not as marked in the upper part of the face as in the lower. The case is one of well-marked hemiparesis of cerebral origin complicated with tremor.

The pupils respond readily to light, and accommodation is good. There is left-sided homonymous hemianopsia. Smell and hearing, so far as determined, are normal. There is no gastric disturbance; no vertigo and no headache since August. She complains occasionally of the left side being cold.

There is an oval swelling above the right ear. The central point is

elevated fully one-half an inch above the level of the skull. The scalp is normal, and is not infiltrated or inflamed over the protuberant area. A horizontal line around the skull a little above the external angular process passes over the tumor, and the right half of this line is three-quarters of an inch longer than the left. The base of this swelling measures in vertical diameter three-quarters of an inch (two cm.) and its horizontal diameter two inches (five cm.). The apex of the swelling is two and one-half inches above the external auditory meatus.

The motor area first involved by the growth was evidently the centre for the wrist and fingers in the middle third of the ascending parietal convolution. The size of the tumor, the amount of brain-tissue displaced, and the pressure established, implicated the entire left side in loss of motor power. The tumor could be seen and felt. Cerebral localization was unnecessary.

I extract from a report made by Dr. Frank Fry, who carefully examined the child: "Patient has a left hemiplegia; face and neck (slightly) and upper and lower extremities are involved. Eye is slightly affected; the lower part of the face much more. Tongue protrudes straight. She can extend the left hand (wrist) to right line. Relative grip of two hands by dynamometer is: right, 60; left, 40. Has a hemiplegic gait, foot being circumducted and the toe scraping slightly in walking. Can flex the foot (ankle) slightly. There is a slight talipes varus. Measurements of extremities are as follows:

"*Arm:* Dext., 4 inches above external condyle of humerus, 9½ inches; sin., 4 inches above external condyle of humerus, 9¼ inches.

"*Forearm:* Dext., 4 inches above styloid process of ulna, 8 inches; sin., 4 inches above styloid process of ulna, 7¾ inches.

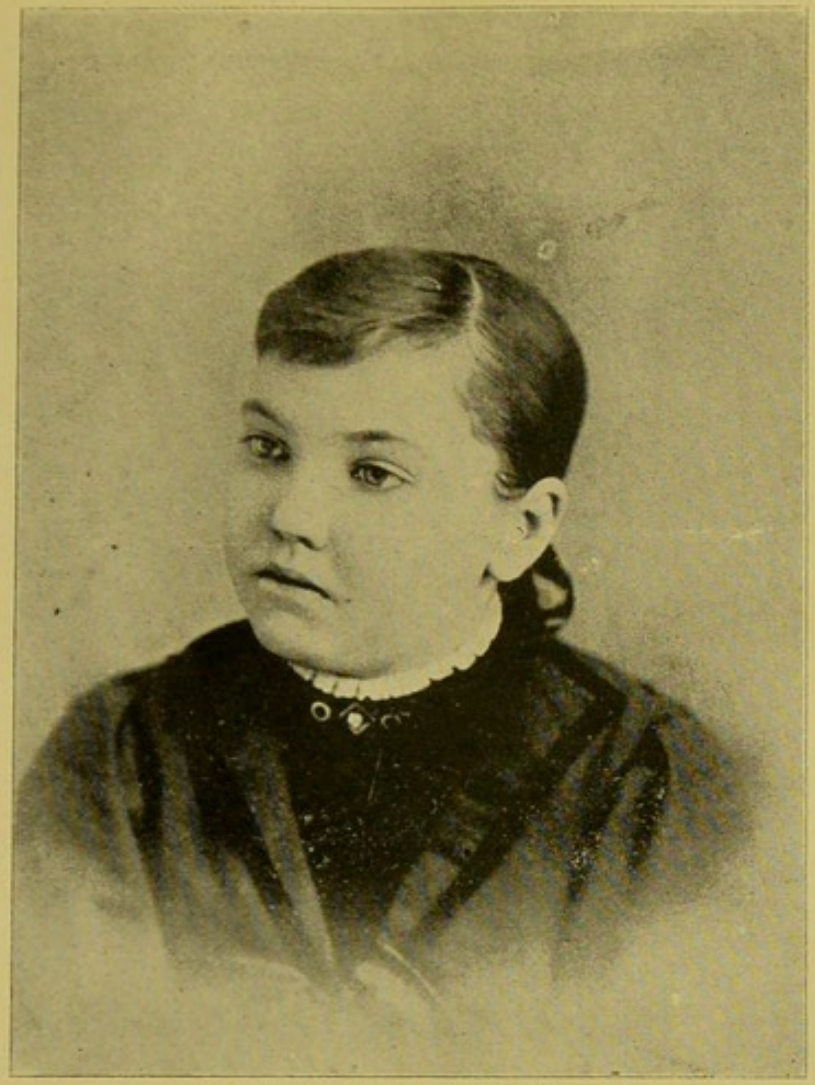
"*Leg:* Dext., 4 inches below head of fibula, 12¼ inches; sin., 4 inches below head of fibula, 12¼ inches.

"The muscles of the neck, of the upper and lower extremities, are in a condition of rhythmic spasm and tremor. This is only slight in the neck, greater in the leg and thigh, and greatest in the arm, forearm, and shoulder (in order named). The spasm is increased by voluntary movement; greatly so in the upper extremity. Knee-jerk slightly increased on the left."

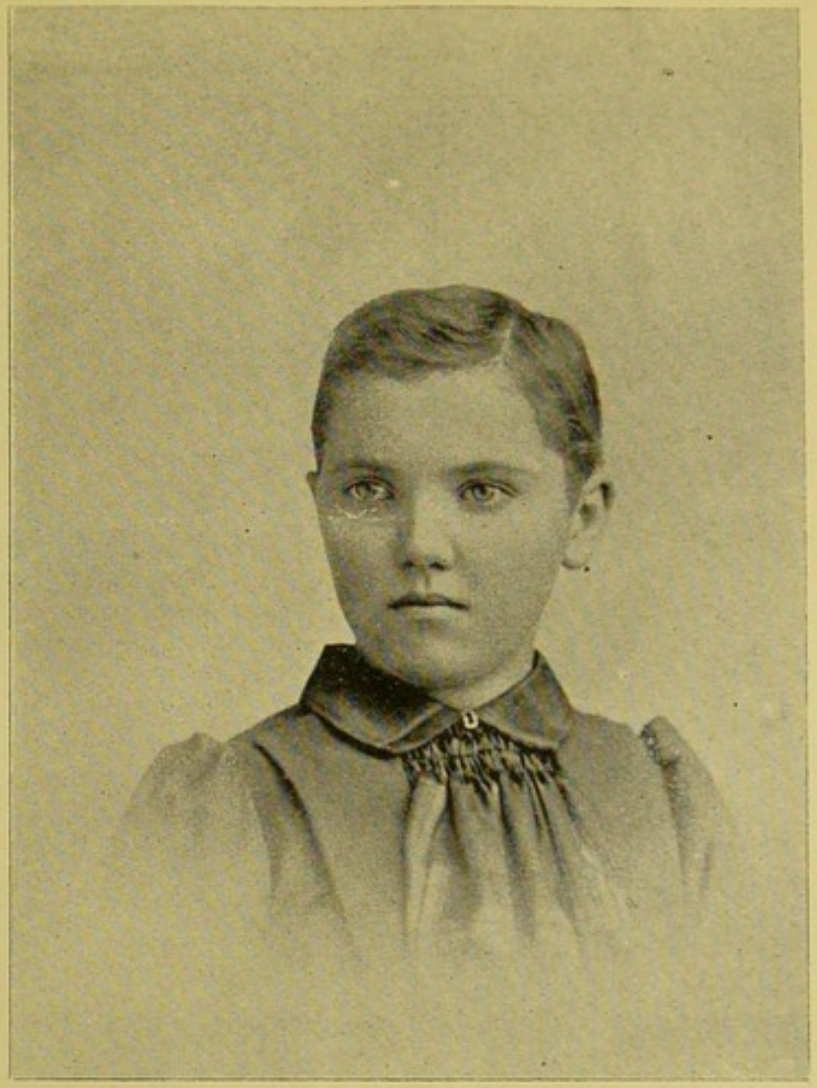
Dr. J. B. Shapleigh, examining the ears, reports them almost normal, and says that "there is no evidence that the function of the auditory nerve is at all interfered with by the cerebral trouble."

Dr. M. H. Post found "optic neuritis, the swelling of each disk amounting to four dioptrics. Also, a loss of vision in the left half of each field (homonymous hemianopsia) corresponding to the right half of each retina to the vertical median line. Nothing in the condition of media or fundus of either eye to account for this defect. (Before the operation vision in right eye $= \frac{14}{XVI}$, some hesitancy, and in the left eye $= \frac{14}{XVI}$, prompt and accurate.) Pupils react well both to light and accommodation.

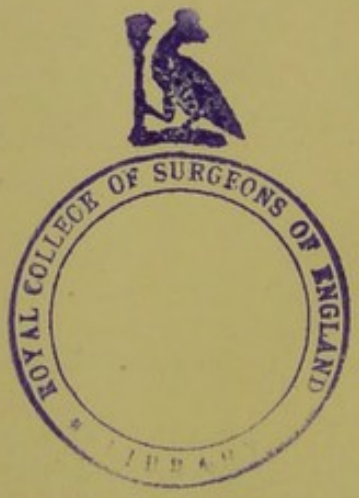
Ten days after the operation, vision in right eye $= \frac{14}{XII\frac{1}{2}}$, and in the left eye $= \frac{14}{XII\frac{1}{2}}$. Condition of each fundus as at first examined, and hemianopsia still present.



ONE MONTH BEFORE OPERATION.



FIVE MONTHS AFTER OPERATION.


ROYAL COLLEGE OF SURGEONS OF ENGLAND

The anatomico-pathological diagnosis was uncertain, but we suspected a sarcoma. The temperature on the day before the operation was 100.4°, though she appeared well in other respects.

Operation. The patient having been properly prepared—antiseptic precautions being taken—the operation was performed March 20, 1891. A large, oval flap of the scalp and periosteum, with its base above the ear, was reflected from the skull. The bone was incised by a chisel, and an oval flap outlined with the bone-pliers. This flap was turned downward; the bone was thin and pliable and did not break. The periosteum was closely adherent to the elevated area. The dura was attached to the bone, but was loosened; it was evident that a fluid separated it from the brain. There was no pulsation perceptible either to sight or touch. The dura was incised, and a clear, watery fluid escaped. The opening was enlarged, and a collapsed cyst was lifted from the opening. Many cysts of varying sizes were removed, with their contents. Finally, a translucent membrane was exposed at the deepest part of the depression in the brain; it looked like another cyst, and was purposely punctured. It was probably the ependyma ventriculi, for the lateral ventricle was opened and drained; the flow of serum was free, and continued seventeen days.

The excavation in the brain left by the removal of the cysts was cleared of the fragments of the cysts-walls by a blunt curette used as a scoop. The brain was not bruised. This deep excavation, as large as a hen's egg, retained its outline. The cavity of the lateral ventricle being dilated was well exposed. The outlines of the convolutions were obscured; but, as nearly as I could determine, the temporo-sphenoidal convolutions were pushed downward and backward. The cavity occupied the greater part of the lower two-thirds of the Rolandic region.

A gauze drain was left in the opening in the dura and carried out through the bone and the scalp. The bone-flap was put into position, and the scalp sutured in place. The abundant antiseptic dressings put over the wound were held by a roller bandage which exerted moderate pressure.

The patient bore the operation well. There was no apparent disturbance from the escape of the cerebro-spinal fluid.

There was no evidence of inflammation of the parts involved, except in the adhesion of the dura mater and pericranium to the thin and distended parietal bone.

Notes from Case Record. March 20, operation at 1 P.M. Present: Drs. Gregory (senior and junior), Dr. N. B. Carson, Dr. Frank Fry, Dr. L. Bremer, Dr. E. F. Smith, Dr. Harry Hodgen, and Dr. Harvey Mudd.

20*th* (five hours after operation). Dressing is saturated with moisture. Left foot and heel itching. Less motion in arm. Sweating.

21*st*. Arm quiet, except as muscular effort brings twitching. Tent removed. Pulsation evident over the site of the protuberance.

23*d*. Vomited several times to-day.

24*th*. Can move arm without tremor.

25*th*. Moves arm without tremor.

April 4. Twitching of arm more marked during last day or two. Serous discharge has been less.

7*th*. Dressings dry. There has been a diminishing discharge of serum. Has been no suppuration. A soft swelling appeared at the

site of opening in the skull two or three days after the operation. It is now quite marked, more tense, and pulsates.

20*th*. Arm twitches even when patient is quiet.

21*st*. Arm twitches almost constantly.

24*th*. Right side of face and scalp much swollen.

25*th*. Wound was reopened. The mass proved to be a hernia cerebri protruding through the opening in the skull. The hernia was explored with the aspirator. No fluid was obtained, and no evidence of a re-development of the cystic growth was discovered. The scalp was again sutured and the wound entirely closed after freshening the edges. Firm pressure was put upon the hernia. This exploratory operation did not disturb the patient. The wound healed perfectly in a few days. The firm pressure put upon the hernia soon controlled it, and the patient's general condition rapidly improved. The hernia had a base of nearly two inches diameter and an elevation of at least an inch.

30*th*. Patient for first time wants to get out of bed. Walked about the room.

May 1. Patient is hungry all the time.

7*th*. Discharged from the hospital.

The surface temperature of the body and extremities was not taken. The temperature for a few days before the operation was recorded. Only once was it observed to rise above 100°; this was on the day before the operation. On the day of the operation the morning temperature was 99°; at 9 P.M. it was 102.4°. Most of the time for the succeeding thirty-five days it fluctuated between 100° and 103°. The changes were at times rapid and always uncertain. On the twenty-second day, for the first time, a normal temperature was recorded, but on the same day it reached 105.4°. This fever did not appear to distress the child; the tongue remained clean, the appetite good, and the loss of flesh was not great. The child slept much, but was bright, though irritable when disturbed, and she had no desire to get up. Pupils reacted well, and were rather large.

The swelling at the site of tumor reappeared after the first operation before the primary dressing was removed; it pulsated, and was soft; it increased in size, and at points the scalp appeared thin, and fluctuation was evident; I punctured at different points, and serum escaped freely; drainage from under the scalp and from these punctures did not cease until the seventeenth day. I feared the fever was due to pressure from a serous exudate from the ventricles or to an inflammatory process, though no other evidence of wound disturbance was present. Hence, the support given to the hernial protrusion was insufficient and more or less intermittent. The patient was irritable, and complained bitterly of a tight bandage. On the thirty-fifth day, with a temperature of 102.8°, the scalp was lifted from the skull and the tumor sufficiently exposed to reveal its true character as a hernia of the brain. The aspirator was used at several different points with a negative result. Being now satisfied of the condition, a pad, with firm pressure from a bandage, was applied. The temperature promptly subsided, and the next day reached 99°. Thereafter it was normal, and the patient was discharged well eleven days later, or May 6, 1891.

The prompt disappearance of the fever was evidently due to the pressure and control of the hernia cerebri. It is now evident to me that

prompt and efficient pressure would have hastened the recovery of my patient. It is probable that irritation of the motor area and the corpus striatum determined the abnormal heat.

Etiology of tumor. The patient lived in the country, and had for a playmate a shepherd dog with which she spent much of her time. The dog was killed in 1889, so that the infection of the patient, if from this source, must have occurred at least one and one-half years before the operation. But it was only eight months from the initial symptom to the date of the operation. It is possible that some of the larvæ may still remain as a focus for a redevelopment of this parasitic cyst.

The prognosis is, however, good, as other cysts of the same character have been cured simply by aspiration.

A considerable quantity of the fluid from the cysts was lost, but the portion saved, together with all of the cyst-walls, was given to Dr. Ludwig Bremer for examination. The specimen removed and some microscopic slides were presented for inspection.

Dr. Bremer's report, together with remarks on the neurological aspect of the case, he has kindly added to this history.

September 16, 1891. The patient presented herself for examination to-day. She has worn a truss over the defect in the skull up to the present time. There is no disposition to protrusion of the brain, and the place of operation is marked by a slight depression. The thin flap of bone which was preserved and turned back is still perceptible to the touch in its original size. The pulsation of the brain can be felt at the upper crescent of the defect through the integument whenever the head is in a dependent position.

The paralysis and irritative disturbances have disappeared. Her gait is perfect; she is able to hop on the formerly affected (left) leg. Uses her left hand with ease and strength. Facial expression is natural. No trace of tremor. Reflexes normal.

Dr. Post, after a final examination, reports that the optic neuritis has disappeared; bloodvessels still a little enlarged. The hemianopsia is disappearing, the field of vision being nearly normal.

This complete recovery from the effects of a defect of such size as that described above is, perhaps, without a parallel in cerebral surgery. If we consider that a prolapse of the brain of such magnitude always means more or less necrosis, and occurring in a highly differentiated region of the brain, as this did, it is certainly remarkable that a complete restoration to perfect health should have taken place as evidenced by the absolutely normal condition of the organs affected by the morbid process.

Literature (by Dr. Bremer).—Hydatids of the brain are comparatively rare. T. S. Cobbold (*Parasites of Man and Animals*) found that among 327 cases of hydatids known in literature, 22 occurred in the brain. The organ of predilection for the parasite is the liver.

Up to the year 1883, 19 cases of echinococcus of the brain had been

registered by A. Steffins as post-mortem findings. Since then about 9 others have been reported, 3 of which are not based on autopsies only, but gave cause for operative interference. Of these, as will be seen, only 1 recovered.

Dr. A. Castro,[1] of Buenos Ayres, trephined a boy of fourteen, with localizing symptoms, over the Rolandic region, for a progressive atrophy and paralysis of both extremities of the right side; pain on left side of the head. Two cysts were found—one the size of a walnut, the other that of a hen's egg. The patient died from basilar (?) meningitis.

Another case was operated on by J. C. Verco.[2] The patient was a boy of eleven years. At first, pain in right temporal region; later on, protrusion of bone. Choked disk, mydriasis, attacks of unconsciousness, with spasms in the left arm. The patient was already much reduced in strength when consent to operation was given.

After the cyst had been removed a very large cavity remained. The wall of the cyst was formed by a thin layer of brain substance. The child died from purulent meningitis, which probably had existed before the operation.

The third[3]—and, previously to the one under discussion, the only case recovered after operation—is reported in the *Australian Medical Journal* for July, 1890. Dr. Graham and Mr. Grubbe diagnosticated, in a boy of sixteen years, presenting the symptoms of headache, nausea, vertigo, vomiting, loss of memory, mental dulness, and paresis of the right arm, a tumor in the left motor area.

The bone when removed was only one-sixteenth of an inch thick, and a cyst four inches long was found upon the pia mater, and removed. The brain itself was not involved; the boy recovered and remained well, except for his blindness.

Point of Colonization and Development of the Parasite in Dr. Mudd's Case.

The starting-point of the parasitic growth was most probably on the convexity of the brain, over the centre for the wrist and fingers, in the subarachnoidal space. Since the sac extended through the whole of the centrum ovale into the lateral ventricle, it might be assumed that possibly it developed from the latter, and thence grew and spread toward the convexity, for the ventricles form a seat of predilection for echinococcus of the brain; next comes the subarachnoid space. But the intra-ventricular origin of the growth is rendered improbable by the clinical course of the case.

Localized irritative symptoms (twitching of the hand) manifested themselves first, pointing to a circumscribed area of the cortex; with

[1] Jahrb. ür Kinderheilk., Bd. xx, Heft 1.

[2] Annales del Círculo Medico Argentino, October, 1889.

[3] "Hydatid of the Brain; Removal by Operation; Death after Four Days." Intercolonial Medical Congress of Australasia: Transactions of Second Session, held in Melbourne, Victoria, January, 1889. (After Neurolog. Centralbl., 1889, p. 443.)

the encroachment of the increasing tumor upon the subjacent conducting fibres paralytic symptoms set in. Again, the shape of the defect speaks in favor of a development from the periphery to the mesial plane. The shape of the sac, as outlined by the hole in the brain, was that of an egg—oval in the strict sense of the word—the apex being toward the ventricle, the base toward the convexity. Thus, for simple morphological reasons, the subarachnoidal origin of the tumor seems to be the more probable one. Growing from the periphery it pushed its way toward the centre. Whereas, during the first period of its development, there was an effort on the part of the tumor to corrode and discharge through the skull-bone, this outward march was checked for some unaccountable reason, after the inner table and the diploë had been destroyed and the outer table had been reduced to a thin lamina not much thicker than an eggshell.

Definition and Causation of the Clinical Features.

It might appear singular that a tumor larger than a large-sized hen's egg should produce no graver symptoms than it did, and that the hand and arm should be found only in a state of paresis, when the region of the arm and hand centres, and apparently their conducting fibres, were occupied by a foreign mass. Another question is the nature of the irritative symptom—the tremor.

The first peculiarity, the partial paralysis, seems to be explicable on the following grounds: The tumor grew very slowly; it did not involve the brain directly, but, with its pointed extremity directed in the axis of its growth, it pushed the fibres of the brain substance asunder, compressing rather than destroying them. Supposing even that the cortical centres for the fingers, hand, and arm were destroyed, the slow growth of the parasite gave the surrounding cortical areas a chance to vicariously[1] assume the functions, at least partially, of the destroyed centres. As to the conducting fibres of the corona radiata, they were, no doubt, atrophied by pressure to some extent, but enough remained to carry on the conduction. The general involvement of the whole left side of the body did not depend so much on a cortical destruction as on that of the conducting fibres. The apex of the tumor struck them at a point of convergence, where they unite to form the internal capsule.

It is on this ground, too, that the irritative symptom—the tremor—has to be explained. This tremor partook very much of the nature of the kind met with in cerebro-spinal sclerosis. The anatomico-pathological substratum of this affection is the sclerotic patches in the pyramidal tract of the brain (centrum ovale) and spinal cord. The interruption,

[1] I am well aware that this substitution theory is still a mooted point, but clinical evidence speaks in its favor.

or, at all events, the impairment, of conduction in the converging motor fibres must, in this case, too, be looked upon as the cause of the tremor. Like that observed in sclerosis, it was an intention tremor, increasing in proportion with the efforts at movement.

It may not be amiss to state here that the case reminded one somewhat of chorea electrica.

Eye symptoms. Besides the choked disc, which was a general pressure symptom, due, no doubt, to the dropsical condition of the ventricles, there was the localizing one of left homonymous hemianopsia. From the nature of the lesion it is clear that this was basal in origin. In its gradual growth from the convexity toward the ventricle the cyst could hardly fail, considering its size, to exert pressure downward, beyond the basal ganglia, and impinge upon the optic tract. The point of pressure must have been behind the corpora geniculata, for the promptness with which the pupils reacted to light and accommodation showed that the latter (being the reflex centres of the iris) were not involved by the pressure and remained intact. The tract behind the corpora geniculata, however, between these and the cortical visual centres, was entirely interrupted, for the hemianopsia was complete. Central vision, however, was good.

Headache. That a tumor of very considerable size may exist in the cranial cavity without giving rise to headache has time and again been observed in cases of this class. But here we are confronted with the apparently paradoxical phenomenon that, with increasing size of the growth causing the pain, the latter ultimately disappeared. In spite of the very pronounced "descending neuritis" giving evidence of general brain pressure, there was in the latter months of the development of the tumor not a trace of headache. This feature of the case might be explained by assuming that in the beginning, when the pressure of the developing echinococcus was brought to bear directly upon the sensitive nerves of the dura mater, pain was the result of irritating the living nerves of that membrane; that these latter in the course of time became necrosed by the ever-increasing pressure, and that thus the pain ceased. This is, of course, not to be regarded as an explanation, but only as an attempt at such, of the difficult and obscure problem of the genesis and nature of the headache in some cases of intra-cranial growths.

Reflexes. Of the superficial reflexes only the plantar was tested. It was found to be present.

The knee-jerk could be elicited on the unaffected side with difficulty, and was slightly exaggerated on the left.

Examination of the mass removed. Besides the common sac, which was torn and broken up in shreds by the scraping process resorted to in the operation, there were a number of larger and smaller vesicles, the total mass constituting about a handful of material. Under the micro-

scope the characteristic scolices of echinococcus could be shown. They were abundant, and were demonstrated in the living state at a meeting of the St. Louis Medical Society, March 21, 1891.

It is difficult to make even a rough estimate of the number of heads contained in the whole mass, but, judging from the abundance of brooding capsules found, almost all of which contained living parasites, the specimen must have contained many thousands of such heads. Many of these were, no doubt, set free by the rupture of the capsules produced during the operation. Their failure to re-develop is possibly to be accounted for by the not inconsiderable discharge of cerebro-spinal fluid due to the puncture of the lateral ventricle. The mass of the fluid was probably sufficient to wash them away and carry them outside of the cranial cavity.

Time and manner of infection. The time could not, of course, be established with any degree of certainty. It probably does not date back much over a year previous to the operation. The handling of the diseased (and undoubtedly infected) dog, which could be demonstrated as the more than probable cause of the infection, shows once more the great danger to which children are exposed when allowed to fondle dogs.

The fact that the echinococcus occurs most frequently in communities (*e. g.*, Iceland) where people live in close contact with these animals should be heeded more than is done at present. Owing to the scarcity of post-mortems in our country the number of echinococcus infections, whether of the brain or other organs, is impossible to estimate. But I believe it is not overstating the case when I say that only a minimal percentage of echinococcus infections in children are diagnosticated or even suspected, and that more obscure affections are due to the invasion of the organism by this parasite than is commonly believed. I have no doubt that with legitimate brain surgery becoming more widely disseminated and understood by the generality of surgeons, more cases will come under observation within the next few years than one would, at first thought, be inclined to believe. The cases of operative interference for echinococcus of the brain have all occurred within the last few years. It is, perhaps, needless to call special attention to the fact that in all of them the patients were young people.

Thermal abnormalities. There was an elevated temperature before and after the operation. According to the researches of Ott,[1] Hughlings-Jackson, Sawadowski,[2] W. Hale White,[3] and others, the chief heat-producing centres are in the corpus striatum and in the cortex around

[1] Journal of Nervous and Mental Diseases, 1888, xiii. p. 85; January, 1889.

[2] "Zur Träge über die Localization der Wärme-regulirenden Centren," etc., Centralbl. d. med. Wiss., 1888, Nos. 8–10.

[3] "On the Parallelism Between the Three Thermic Mechanisms and Dr. Hughlings-Jackson's Three Levels," Brit. Med. Journ., 1890, p. 949.

the Rolandic fissure. Both of these heat centres were implicated in this case. The temperature was highest (105.4°) when their disturbance was greatest—*i. e.*, when the hernia cerebri had reached its greatest dimensions. The rapid fall and return to the normal so promptly after the pad had been applied is very instructive, and amounts almost to the precision of a physiological experiment. No attempt was made to ascertain the existence of any difference of temperature between the right and left sides.

It is, perhaps, of some interest to call attention to a case mentioned by Sawadowski,[1] and reported by Bagojawlensky, in which a high temperature in a patient could be explained post-mortem only by the existence of echinococcus cyst in both the corpora striata.

The Prognostic Features of the Case.

Instances of spontaneous discharge and cure of cerebral echinococcus through the cranial bones—the convex walls as well as the base of the skull—through nose and mouth, have been reported. Cases of cerebral echinococci discharging into the ventricles and thus causing death are also on record. In this case, judging from its clinical course and the condition revealed by the operation, death of the child would have been only a matter of time. The sac had been constantly enlarging, and instead of making any further inroad by corrosion upon the skull-bone it delved down into the brain substance, and, as stated above, only a translucent membrane—possibly the ependyma—separated it from the ventricle.

Fortunately, the hydrops of the ventricle was only beginning, the descending neuritis only moderate, central vision still good; so that, in this respect, the termination was a happier one than in the only other successful case extant—that of Graham and Grubbe, whose patient remained blind.

It was, likewise, a favorable feature in our case that no epileptic distubances had as yet manifested themselves. However successful an operation may have been, the epileptic change in the brain substance is ever to be dreaded as soon as a well-pronounced epileptic seizure has once taken place. Nothing of the sort, not even unconsciousness, had ever been noticed.

That no mental impairment took place is, as a matter of course, due to the location of the lesion in the right hemisphere. A like defect in the opposite one would doubtless have left a considerable degree of mental failure behind.

[1] Mentioned in the article sub. 2.

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