OBSTRUCTIVE INTERNAL HYDROCEPHALUS FOLLOWING
OPERATIVE REMOVAL OF CHRONIC SUBDURAL
HEMATOMA IN INFANTS

REPORT OF TWO CASES*

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(Received for publication December 23, 1959)

Two patients with this unusual syndrome are presented. One has done well in the 6 years that have elapsed since eventual operative relief of the intracranial pathology via a Torkildsen procedure. The other case terminated fatally, post-mortem studies having been carried out.

The essential sequence of events in this syndrome is that of an infant having unilateral or bilateral chronic subdural hematoma removed, whose head, however, continues to enlarge and on subsequent studies is found to have an internal hydrocephalus, usually of the noncommunicating type, which may be relieved by a Torkildsen procedure, or more recently by the Spitz-Holter or Pudenz valve.

CASE REPORTS

Case 1. A white male infant, aged 3 weeks, was first admitted on June 24, 1953. The child had been born 1 month prematurely. The pregnancy and labor of the mother were uneventful. The head was somewhat large at birth.

At the age of 3 weeks, the circumference of the head was 43 cm., normal being 36.5 cm. The anterior fontanelle was bulging.

Bilateral subdural taps were done through the anterior fontanelle on June 25. Twenty cc. of slightly yellow subdural fluid were removed from each side. Equal amounts of air were injected and films showed air extracerebrally, in the subdural space, bilaterally. Right and left subdural taps were continued in the next few weeks in the ward, with removal of yellow fluid on each occasion. On July 17 a large subdural effusion was evacuated from the left side through a burr hole. At this time the cerebral cortex was fully 1½ inches below the dura mater.

The patient was discharged on July 30.

2nd Admission, Aug. 21, 1953, at age of 11 weeks. There was marked increase in tension of the anterior fontanelle and generalized increase in size of the head, which measured 46.5 cm. in circumference. Both anterior and posterior fontanelles were quite large and the sagittal suture was widely separated.

On August 24, a left parietal osteoplastic bone flap was turned down and subdural fluid and membranes were evacuated and stripped from the cerebral cortex. The brain was depressed considerably beneath the dura mater.

Microscopic diagnosis of the surgical specimen was “organizing subdural hematoma membranes.” It was composed of a loose fibrous stroma, with fibroblasts, lymphocytes, plasma cells, and histiocytes. The outer membrane was much thicker than the inner membrane.

Postoperatively the patient did well, except for the 1st day when there were moderate clonic jerks of the right arm and leg. In the succeeding days, the left subdural space was tapped, progressively smaller amounts of fluid being removed.

The child left the hospital on September 13. It was thought that the chief problem thereafter would be a somewhat slow or delayed expansion of the left cerebral hemisphere to take up the “dead space” incident to the evacuation of the subdural fluid.

3rd Admission, Oct. 14, 1953, at age of 4+ months. On October 20, a large collection of straw-colored fluid was evacuated from the right subdural space through a burr hole. A #12 French catheter was left in the space for several days. This time there was no collection of fluid in the left subdural space. A week later the fontanelle again was quite tense. The patient was discharged on October 30.

4th Admission, Nov. 22, 1953. Ventriculography via the anterior fontanelle showed very large lateral ventricles and subdural air as well (Fig. 1).
On December 17, this operation was carried out on the right side, connecting the enormous right lateral ventricle to the cisterna magna, using a #10 French catheter.

Six days postoperatively the fontanelle was flat and on Jan. 4, 1954, the child left the hospital with the anterior fontanelle entirely collapsed and concave. He was last seen on July 23, 1959, his age then being 6 years, and he was in excellent condition (Fig. 2). He had had no headaches, convulsions or any neurological deficit so far as the parents could determine. The Torkildsen tube was still in place and it had caused no difficulty in the intervening 5½ years since it had been inserted originally. The child seemed to have high degree of intelligence.

Case 2. A white male infant, aged 7 weeks, was admitted on Dec. 19, 1958. He came to the emergency room with a history of loss of appetite, listlessness and vomiting for several hours. His parents stated that the infant had dropped off a bed the night before, seemingly without losing consciousness. It was learned that he had been in another hospital 1 week prior to this admission because of a feeding problem and difficulty in gaining weight. In fact, the child weighed only as much as he had weighed at birth.

He was a thin infant, and slightly lethargic. Circumference of the head was 40° cm., and circumference of the chest was 36 cm. Both anterior and posterior fontanelles were open, but showed no increased tension on palpation, except when the infant cried. Evidence of recent trauma to the head was demonstrated by the presence of ecchymosis and hematomas of the scalp. Roentgenograms of the skull showed an extensive stellate fracture of the left posterior parietal region, extending both anteriorly and posteriorly, with separation of the fragments up to 1 cm., but no actual depression (Fig. 3).

On December 22, the patient’s condition became worse. There was pronounced nuchal rigidity with opisthotonos, and his fontanelles were tense and bulging. A left subdural tap was done through the anterior fontanelle and yielded 100 cc. of grossly bloody fluid. The infant improved considerably.

On December 24, he began to vomit and another 100 cc. of subdural bloody fluid were obtained from the same area. An attempt to tap both lateral ventricles failed. On December 26, bilateral temporal burr holes were made, under general anesthesia, and chronic subdural hematomas were evacuated from both the right and left sides of the cerebral convexities. Well organized outer and inner membranes were found on both sides. On the left, the encapsulated collection of fluid was much larger than on the right. No drains were left in place. The immediate postoperative course was satisfactory.
Fig. 3. Case 2. Note egg-shell type of fracture (arrow) in posterior part of parietal bone on left side of skull with separation of the fragments. Note also the enormously dilated lateral ventricles and air within subdural space. The lateral ventricles could not be tapped on admission 1 month previously (see text).

On Jan. 3, 1959, however, the patient again became irritable, presenting a very slow pulse and opisthotonos. A left subdural tap was done through the trephine opening and deeply xanthochromic fluid was recovered. Following the tap there was clinical improvement, except that the opisthotonos did not disappear. Repeated subdural taps were done, but fluid reaccumulated.

On January 12, under general endotracheal anesthesia, a left frontotemporal craniotomy was carried out, with evacuation of a large accumulation of bloody subdural fluid, and removal of a large portion of both outer and inner membranes. After evacuation of the hematoma, the brain began to expand until it reached the dural surface. The left lateral ventricle then was cannulated at a depth of 5 mm. and a large amount of xanthochromic fluid under markedly increased pressure was removed. It is to be remembered that, a month previously, the ventricles could not be tapped (see above). After injection of air, it was thought that the child also had an internal obstructive hydrocephalus, which would eventually necessitate a shunting procedure.

By the 2nd postoperative day, the size of the head had increased considerably. Ventricular taps were done on January 14 and 15 in an attempt to control the increased intracranial pressure.

On January 23, the patient was operated upon again under general endotracheal anesthesia in an effort to perform a ventriculo-upper cervical shunt by the Torkildsen technique. The short arms of a soft rubber "T" tube were inserted easily into the enlarged right and left occipital horns of the lateral ventricles. Upon opening the dura mater of the upper cervical region, however, it was found that the subarachnoid space was completely obliterated or had never developed. The dura mater and pia arachnoid were extremely adherent, leaving no subarachnoid space for the insertion of the lower end of the tube. No cerebrospinal fluid was found at this level. While the dissection was being carried out, a cardiac arrest developed, which necessitated prompt opening of the chest and cardiac massage. A feeble, irregular cardiac contraction resumed. The chest was closed and the shunting procedure was discontinued.

The patient died 3½ hours later in the recovery room. Autopsy was obtained.

The fixed brain was sectioned on February 12. The ventricles were rather large and the foramen of Magendie was large and open, but the foramina of Luschka were closed completely, bilaterally.

Microscopic description of the surgically removed subdural membranes, and the brain at autopsy is as follows:

1. Typical thick outer membranes of chronic subdural hematoma removed surgically show a loose fibrous stroma with fibroblasts, histiocytes and a few lymphocytes and plasma cells.

2. Section of the cerebral cortex (obtained post mortem) shows disintegration of white matter and reactive gliosis (giant plump astrocytes), indicating that the most severe trauma in this child had occurred probably at birth.

3. Normal arachnoid in some areas (Fig. 4).

4. Moderate thickening and hypercellularity of the arachnoid with practically obliterated subarachnoid spaces in other areas (Fig. 5), associated with: (a) pronounced thickening with increase of arachnoid fibroblasts (Figs. 6 and 7), and (b) extensive proliferation of the subependymal glial layer around the lateral ventricles (Fig. 8).

5. No evidence of neoplasm was seen in the brain on thorough gross and microscopic study.

6. Patency of the aqueduct of Sylvius was demonstrated.

Comment. These 2 cases are examples of complicated pathology in infants, which began apparently as subdural collections of fluid overlying the cerebral hemispheres on one or both sides and ended in a form of internal hydrocephalus, usually noncommunicating or obstructive, which may be relieved by some type of shunt operation, either the Torkildsen procedure or the Spitz-Holter valve, as the individual case requires. The interesting problem centers around the pathogenesis of these cases. Perhaps the most logical explanation was found in a French journal in which Thibaut stated that the skull will increase in volume in cases of chronic subdural collections. He cited...
Davidoff and Epstein on this point, and also referred to Elvidge and Jackson as stating that such an increase in the size of the head may be caused by an associated internal hydrocephalus in these cases. Elvidge and Jackson studied the pathology of 55 cases of subdural hematoma in children. If the hematoma is large, it will compress the underlying hemispherical surface over a large area, and will thereby obliterate the subarachnoid spaces over the cerebral convexity, thus causing a profound circulatory disturbance of the cerebrospinal fluid. This will result in an internal hydrocephalus of the communicating type with an increase in the size of the head, even after the subdural clot has been removed. Thibaut stated that internal hydrocephalus may be of the non-communicating (obstructive) type when the subdural hematoma in children is associated with a temporal lobe herniation with compression and distortion of the midbrain and the
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Fig. 6. Case 2. Section from base of brain showing thickening of arachnoid, without evidence of inflammation, as well as obliteration of subarachnoid spaces. This alteration at level of the basal cisternae explains how non-communicating hydrocephalus may be produced when such pathology is encountered at level of the cisterna magna and at the foramina of Luschka and Magendie.

Fig. 7. Case 2. Another section of the arachnoid at base of brain showing proliferation of arachnoidal fibroblasts as well as formation of new capillaries. This means that the process has a certain "age" and was not caused by very recent hemorrhage.

...aqueduct of Sylvius. Pneumoencephalographic demonstration of the latter type of internal hydrocephalus was accomplished by Robertson in a 9-month-old child who was born after a difficult labor and who, at the age of 8 months, presented a progressive increase in size of the skull. The air was shown to stop at the aqueduct of Sylvius and examination of the basilar cisternae suggested a temporal-lobe herniation on the same side as the hematoma or hydroma. Craniotomy revealed a large subdural hydroma over the...
whole extent of the cerebral hemisphere on the same side as the temporal-lobe herniation.

Whether noncommunicating or communicating, Thibaut\textsuperscript{10} stated that internal hydrocephalus occasionally can stabilize itself, resulting in normal or nearly normal spinal fluid circulation. The skull then may become too large for its contents and, with time, there will be thickening of the internal table of the skull and hypertrophy of the frontal and other bony paranasal sinuses in order to compensate for the empty spaces. This is called "hyperostotic hydrocephalic skull," as described by Schüller. When the subdural hematoma is bilateral, there will be a total symmetrical increase in volume of the skull, whereas in the case of a unilateral collection of subdural blood, the increase in size or volume of the skull may be only on the same side. When the collection of subdural fluid is bilateral, there will be what usually is designated as a bilateral external hydrocephalus, which is what occurred for a considerable period of time in our Case 1. In the event of bilateral external hydrocephalus, if no block along the base of the cerebral convexities develops, the lateral ventricles will remain small. This occurs often, of course, after ordinary evacuation of subdural clot, with prompt recovery of the patient, in cases of children or of adults.

In our 2 cases, how did the internal hydrocephalus develop? It is a well known fact that red blood cells in the subarachnoid space will set up a reactive sterile arachnoiditis. This has been demonstrated by several authors. The senior author\textsuperscript{7} of this paper, in 1941, demonstrated in the experimental laboratory that, to a large extent, blood spontaneously becomes agglutinated in the subarachnoid spaces over the cerebral convexities and in the basilar cisternae. Subdural hematoma, especially in infants, is often associated with some degree of subarachnoid bleeding as well.

Adams\textsuperscript{1} conducted a similar study using intact tagged red cells injected into the subarachnoid space (into the cisterna magna of dogs). He demonstrated that approximately 25 per cent of the blood is absorbed directly into the blood stream in an intact state. Only insignificant hemolysis was thought to occur. The remaining 75 per cent of the injected red cells became enmeshed and fixed in the arachnoid. Adams (referring also to Sprong's\textsuperscript{9} earlier work) stated in his summary that this experimental finding may
explain the occurrence of communicating hydrocephalus following subarachnoid hemorrhage by obstruction of the normal absorptive subarachnoid mechanism somewhere along its course. We would like to add that the same explanation might be given to obliteration of the outlets from the 4th ventricle and, therefore, a form of obstructive hydrocephalus might occur also in cases of massive subdural hematomas, often associated with definite or even massive subarachnoid bleeding. Furthermore, distortion and overlying pressure on the brain stem and the aqueduct of Sylvius contribute to a partial or complete basilar obstruction to the normal flow of cerebrospinal fluid, and its out-flow via the iter and the 4th ventricle to the subarachnoid space.

Mabon stated that it usually is not necessary to carry out ventriculography in cases of suspected internal hydrocephalus after removal of subdural clot in infants, unless there is evidence of increased intracranial pressure. The pressure of a large subdural effusion on one or both sides may compress the normal subarachnoid spaces over the cerebral hemisphere to such an extent that there is interference with the normal absorptive mechanism and thus a communicating internal hydrocephalus would result. We believe that the combination of all these factors may well play a part in the production of internal hydrocephalus in patients such as are cited in this paper.

The microscopic study of the brain in our fatal case (Case 2) demonstrated that in certain cases there is obliteration of the subarachnoid spaces not only in the basilar cisternae and around the brain stem, but also occasionally over the cerebral convexities.

Theoretical ideas, therefore, concerning the production of internal hydrocephalus in patients with, or following removal of chronic subdural hematoma, particularly children, include:

1. Obliteration of subarachnoid spaces over the convexity of the brain, first by the overlying subdural mass of fluid or blood, and secondarily by fibrosis and adhesions (Figs. 4 and 5).

2. Obliteration of subarachnoid spaces at the level of the basilar cisternae by (a) organization of the subarachnoid blood incident to the hemorrhagic lesion and (b) pressure exerted by herniating parts of the brain (Figs. 6 and 7).

3. Narrowing or occlusion of the aqueduct of Sylvius, secondary to the herniated uncus and also to pressure exerted on the brain stem by distortion and pressure from above when the patient had a subdural hematoma or hematomas over the cerebral hemispheres (Fig. 8).

Concerning surgery for correction of the subsequent internal hydrocephalus, it would seem advisable now to perform a ventriculo-caval shunt, rather than a Torkildsen procedure, as, in attempting the latter operation, one might find an extremely adherent or even obliterated subarachnoid space or cisterna magna, as in our Case 2, making it impossible to insert the lower end of the catheter properly. In future cases, we expect to carry out a ventriculocaval shunt in all patients of this type.

Of course, it could be conjectured that the internal hydrocephalus present in these 2 cases was either of a congenital type, present even before the formation of the hematoma, or concomitant with, but not necessarily related to, the clot over the cerebral surface.

However, even though these possibilities may be true, we believe that this is not the case. We base this assumption on several facts: (1) The development of internal hydrocephalus following subdural hematoma over the cerebral convexity, be it communicating or noncommunicating, has been demonstrated to our satisfaction by the cases reported by Thibaut, Robertson, and Elvidge and Jackson. (2) We have histological proof in our Case 2 that the arachnoidal thickening of the base of the brain had been present for certainly more than a few days, as demonstrated by the fibrosis and new capillaries formed, and by the evidence of a relatively old cerebral injury, as shown by the gliosis in the cortex. The old cerebral injury probably produced some subarachnoid bleeding, as well as tearing of some bridging veins, which in turn caused the subdural hematoma. (3) We believe that if
internal hydrocephalus had been present before the formation of the subdural clot, the mere presence of a state of increased intracranial pressure would most likely have minimized or prevented the accumulation of blood in the subdural space. (4) For some time after the evacuation of the subdural hematoma, especially in Case 1, there was no evidence whatever of increased intracranial pressure, the frontanelles being flat, and indeed quite concave.

The relationship between the subarachnoid bleeding and the formation of internal hydrocephalus was first suggested by Bagley in 1928, and the conclusion was drawn then that although some absorption of blood always takes place, there is, nearly always, an associated meningeal thickening, which in turn may create a block within the pathways of absorption of the cerebrospinal fluid and finally result in internal hydrocephalus. Foltz and Ward also suggested the possible role of an adhesive arachnoiditis of the basal leptomeninges, secondary to subarachnoid bleeding, as a factor in the pathogenesis of infantile congenital hydrocephalus.

Ingraham and Matson, in discussing hemorrhage and its relationship to infantile hydrocephalus, made the statement that "There is . . . clinical evidence available that intracranial bleeding at the time of birth or before birth may be followed by fibrosis of the leptomeninges, particularly at the base of the brain. This fibrosis, together with organization of residual blood within the basilar cisternae, may lead to progressive obliteration of the latter spaces." This statement, however, is hardly what we have reference to in the present paper, which describes, rather, the occurrence in a child, in the first few weeks or months of life, of a large chronic subdural hematoma or hydroma on one or both sides and removed successfully by craniotomy or drainage through a burr hole, only to be followed by, or at times be associated with, an enormous noncommunicating or communicating internal hydrocephalus, eventually relieved by some type of shunt operation.

CONCLUSION

Two cases have been presented of what were large unilateral or bilateral subdural hematoma (or hydroma) in infants, of surgical importance (overlying cerebral hemispheres), originally relieved by the usual evacuation through a burr hole or by craniotomy, to be followed, however, by the development of definite internal hydrocephalus, usually of the obstructive type, as shown by ventricular air studies and/or the indigo-carmine test by ventriculospinal puncture. The pathologic process ultimately should be relieved, as in Case 1 herein reported, by some type of shunt operation, either the Torkildsen operation or (preferably) the insertion of the Spitz-Holter valve. The possible pathogenesis and mechanism of development of this interesting course of pathological events are discussed briefly.

REFERENCES