CHOROID PLEXUS PAPILLOMA CAUSING SPONTANEOUS SUBARACHNOID HEMORRHAGE

REPORT OF CASE AND REVIEW OF LITERATURE

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Among the non-aneurysmal causes of bleeding into the subarachnoid spaces are vascular malformations and vascular tumors. Of the latter, papillomas of the choroid plexus have only recently been implicated as a cause of blood appearing in the cerebrospinal fluid. The following case represents such an unusual lesion which was recognized and treated during life, though terminating fatally because of complications.

REPORT OF CASE

J.G., a 15-year-old school girl, was admitted to the Ohio State University Hospital on June 28, 1955. She had been well until 8 days prior to this when there developed sudden severe frontal headache associated with vomiting, slight nuchal rigidity, mental fogginess and drowsiness. Examination disclosed bloody (cherry red) cerebrospinal fluid under 280 mm. of pressure. By the morning of admission, she was stuporous with paroxysms of severe headache and vomiting.

Examination. There was bilateral early papilledema with unequal pupils, the left being slightly larger than the right. A positive left Babinski sign was present. On lumbar puncture the cerebrospinal fluid was found to be under 300 mm. of pressure and contained 29,000 red blood cells and 65 white blood cells per c.mm., with a total protein content of 66 mg. per cent. Bilateral carotid angiograms revealed a questionable vascular anomaly in the right posterior superior temporal area.

Course. Bilateral papilledema increased rapidly to 3 D. with associated retinal hemorrhages. On June 30, 1955, a vertebral angiogram (Fig. 1) disclosed the vascular anomaly in the right posterior superior area. On July 2, her condition worsened with increase in papilledema, a slow pulse and increasingly bloody cerebrospinal fluid containing a total protein content of 1,280 mg. per cent. Bilateral posterior ventriculostomy disclosed blood clots in the right lateral ventricle. Ventriculograms revealed moderately dilated lateral and third ventricles without displacement and a filling defect in the right lateral ventricle at the trigonal area, considered to be a mass lesion—either the remains of a blood clot or a neoplasm.

Operation. On July 4, 1955, through a parieto-occipital transcortical approach, a tumor, about 3×2×2 cm. in size, was found within the trigonal area of the right lateral ventricle, extending anteriorly proximal to the foramen of Monro and for a short distance into the temporal horn. Its arterial supply was multiple, with at least four vessels from the floor and medial wall of the body of the lateral ventricle and others from the floor and wall of the temporal horn. One large vein was seen to drain the blood, probably into the internal cerebral veins. As these vessels were clipped the size of the tumor decreased most remarkably so that upon its removal it was about one-third its original mass.

Course. Her immediate postoperative course was unsatisfactory the next 2 days. Re-
exploration was necessary on July 18 (14th postoperative day) and disclosed severe cerebral edema and blockage of the foramen of Monro with blood clot. A Torkildsen ventriculocisternostomy was established as a precautionary measure. In the following days there developed troublesome accumulations of subgaleal fluid, projectile vomiting and very severe headaches. On July 26 (22nd postoperative day), re-exploration disclosed an epidural blood clot of moderate size with considerable cerebrospinal fluid locked in this area, having come out alongside of the Torkildsen tube, and the latter was blocked with blood clots. She appeared to improve for about 36 hours, then left hemiplegia developed, and she lapsed into coma and died on July 31 (27th postoperative day).

Postmortem Examination. The brain was edematous with severe swelling in the posterior half of the right cerebral hemisphere. Infarction of the brain in the area supplied by the

right posterior cerebral artery was evident. Severe uncal herniation through the tentorial incisura with consequent pressure against the posterior cerebral artery was considered to be the cause of the encephalomalacia. Retrograde thrombosis from the extensive vascular supply to the site of the tumor could not be ruled out as a contributory or a primary cause.

Histopathology of Tumor. The tumor (Fig. 2) presented the characteristic appearance of a papilloma of the choroid plexus. The villi-like structures were covered with cuboidal cells whose cytoplasm was granular and contained minute vacuoles and a central nucleus. The central portion of the villi was made up of loose and somewhat scanty connective tissue, but was rich in blood vessels. The absence of glial fibers and blepharoplasts differentiated it from a papillary ependymoma.

Comment. The unfortunate loss of the patient in this case does not deter from its
clinical significance and interest. The presence of blood in the right lateral ventricle with angiographic evidence of a probable vascular anomaly in the ventricle and a mass defect in the ventricle completed the picture that is diagnostic of the possibility of a papilloma or other mass in the ventricle.

Criticism of the manner in which this case was handled surgically may well be in order. Difficulties were encountered in controlling the bleeding from the tumor bed at the first operation and undoubtedly this was a primary factor in subsequent complications since blood clot was present in the ventricle at the second operation. In retrospect it may be apparent that certain changes in the surgical approach might have proved efficacious, such as (1) a more radical removal of brain tissue in approaching the ventricle (removal of a core of brain instead of a spreading cortical incision); (2) a little lower approach on the hemisphere (incision over the trigonal area instead of through the wound of the occipitoparietal ventriculostomy); and (3) it might have been far better to have left the dura mater open and the bone flap loose or out entirely, thus allowing for cerebral swelling.

DISCUSSION

While it is true that in most cases of subarachnoid hemorrhage the cause is leaking intracranial aneurysm, it is likewise apparent that in from 20 to 40 per cent
of the cases complete cerebral angiography fails to reveal such a lesion.* In two previous reports, one of us (KHA)\textsuperscript{1,4} has emphasized that intraspinal and intracranial neoplasm may give rise to subarachnoid hemorrhage. Of 41 reported cases of intracranial tumor with associated subarachnoid hemorrhage about 50 per cent were primary gliomas of the brain, 25 per cent were pituitary tumors and nearly 15 per cent were intracranial vascular tumors.

Bleeding from a papilloma of the choroid plexus, to our knowledge, was first described by Graves and Fliess\textsuperscript{a} and subsequently by Friedman and Solomon;\textsuperscript{5} however, in these cases the tumor was found at necropsy. Ernsting\textsuperscript{2} appears to have been the first to report a case in which the diagnosis was made clinically and operation was performed. The patient was a 26-year-old male who had had at least one previous (by 3 years) episode of intraventricular bleeding. The second attack was sudden in onset and characterized by slight mental confusion, slight nominal aphasia, a right upper quadrant homonymous hemianopsia and slight lower facial weakness. Left carotid angiography was done and an "intracerebral vascular lesion, measuring 3 cm. in its antero-posterior diameter and 1.5 cm. in its vertical diameter, was demonstrated in the left posterior temporal region. It was supplied by a vessel arising from the internal carotid artery just proximal to its bifurcation, presumably the left anterior choroidal artery." Pneumoencephalography confirmed the presence of a mass occupying the greater part of the trigonal region of the left lateral ventricle and it "appeared to change its position with alterations in the posture of the head." At the first operation via the temporal lobe, only a biopsy was done, but on the second approach to the tumor, a more adequate temporal exposure allowed complete extirpation with subsequent uneventful recovery.

The diagnosis of an intraventricular mass was made, in the case of Ernsting\textsuperscript{2} and in our case, through both angiography and pneumoencephalography. As more of these lesions are surely to be reported, however, it may well be that these minor vascular changes as shown by angiography in the trigonal region of the lateral ventricle or the temporal horn itself will prove of diagnostic significance and thus delay in their diagnosis and surgical extirpation will be avoided.

**SUMMARY**

It has been emphasized that severe subarachnoid hemorrhage may occur from papillomas of the choroid plexus as well as other neoplasms and from vascular malformations. In this report a papilloma of the choroid plexus in the right lateral ventricle was shown to be the source of severe intraventricular hemorrhage. Cerebral angiography and ventriculography were helpful in establishing the presence of the tumor which was surgically removed. This appears to be the second such case to be diagnosed clinically. Two others from autopsy material have been described previously.

It is suggested that in those cases in which angiography fails to disclose the etiology of subarachnoid hemorrhage, further studies—closer scrutiny of technically better angiograms together with ventriculography—may occasionally prove fruitful

\* These figures are only an approximation gathered from various series in the literature. Hamby\textsuperscript{4} has reported, "In a series of patients surviving subarachnoid hemorrhage, a smaller percentage of aneurysms (41 per cent) was disclosed by angiography than in a series studied at autopsy (93.6 per cent)." The accuracy of any such figures is questionable since it is the consensus that in those cases in which there is no aneurysm, subarachnoid hemorrhage is less likely to be fatal and to recur, hence less likely to appear in autopsy material for comparison.
in establishing the source of the hemorrhage in such a lesion as choroidal papilloma or other tumor.

REFERENCES


