CONCLUSION

A case of granuloma of the brain due to Schistosomiasis Japonicum is presented. It is to be pointed out that while this represents a rare situation, further cases will likely appear in the future presenting themselves as intracranial lesions.

REFERENCES

1. Ask, J. E. Personal communication.

ARTERIO-VENOUS ANGIOMA (HAMARTOMA) OF THE BRAIN WITH INTRACEREBRAL HEMORRHAGE

REPORT OF A CASE WITH OPERATIVE REMOVAL OF THE HEMATOMA AND RECOVERY

OSCAR A. TURNER, M.D.*
Department of Surgery, Youngstown Hospital Association, Youngstown, Ohio
(Received for publication August 26, 1946)

The arterio-venous angioma of the brain is a hamartomatous lesion composed of a tangled mass of interlacing vessels which resemble both arteries and veins.11 These lesions have been described under a variety of names (Table 1) but in general have a rather characteristic morphology. When seen at the operating table they appear to involve little more than the surface of the brain, but at autopsy are seen as a wedge-shaped mass which extends to the ventricles. On the surface most of the vessels appear to lie in close proximity but deeper in the

* 226 North Phelps Street, Youngstown, Ohio.
ARTERIO-VENOUS ANGIOI~A OF BRAIN

cerebral substance they are frequently separated by gliotic brain tissue. Inasmuch as some portion of the angioma is in contact with the ventricle, rupture into the ventricular system during a convulsion is a common cause of death in patients with these lesions. In the following case, however, the hemorrhage resulted in the formation of an intracerebral hematoma which was removed surgically.

The patient was a 38-year-old, obese male admitted to the North Unit of the Youngstown Hospital on Mar. 7, 1946. For about 2 years he had experienced intermittent headache associated with nausea and vomiting. There had been occasional twitching of the left side of the face and of the left upper extremity for about 1 year but these had not been constant nor had they been severe enough to alarm the patient.

The day prior to admission the patient had a severe headache with nausea and vomiting, and this was followed by what was described as a "fainting spell" with subsequent development of a spastic left hemiplegia. After this episode he became quite drowsy but continued to complain of severe right hemicranial headache. There had been no symptoms referable to the visual system and no evidence of a speech disorder. There was no history of tinnitus or symptoms referable to the auditory system. The past history, aside from that given above, was non-contributory. Blood Kahn and Kline tests were negative; no abnormalities were found in the usual laboratory studies. The patient was right-handed.

CASE REPORT

TABLE 1

<table>
<thead>
<tr>
<th>Terminology</th>
<th>Author</th>
</tr>
</thead>
<tbody>
<tr>
<td>Angioma arteriole</td>
<td>Cushing and Bailey(^8)</td>
</tr>
<tr>
<td>Cirsoid aneurysm</td>
<td>Uiberal(^9)</td>
</tr>
<tr>
<td>Arteriovenous angioma</td>
<td>Wolf and Brock(^10)</td>
</tr>
<tr>
<td>Arterio-venous aneurysm</td>
<td>Bergstr~and, et al.(^1)</td>
</tr>
<tr>
<td>Aneurysmal angioma</td>
<td>Craig(^2)</td>
</tr>
<tr>
<td>Arterio-venous hamartoma</td>
<td>Turner and Kernohan(^22)</td>
</tr>
</tbody>
</table>

symptoms referable to the auditory system. The past history, aside from that given above, was non-contributory. Blood Kahn and Kline tests were negative; no abnormalities were found in the usual laboratory studies. The patient was right-handed.

Examination. He appeared to be acutely ill and quite lethargic but was nevertheless able to answer questions coherently. Blood pressure was 126/82; pulse 68; respirations 24; and temperature 99.2. There was a left spastic hemiplegia with practically total paralysis of the upper and lower extremities. Facial paralysis of central origin was present on the left and was severe enough to cause marked slurring of speech. However, there was no evidence of an aphasic disorder. The deep reflexes on the left were hyperactive and the Babinski sign was strongly positive on this side. The abdominal reflexes were absent on the left but normal on the right and there was sustained ankle clonus on the paralyzed side.

No sensory defect could be elicited on examination. Confrontation fields disclosed a left homonymous hemianopsia with sparing of the macular area as far as could be determined. The margins of the optic discs were blurred but there was no measurable elevation. The retinal vessels appeared hyperemic but otherwise normal. The left pupil was fixed and measured 9 mm. as compared to 7 mm. on the right. All other cranial nerves were normal. There was no bruit on auscultation of the skull. Roentgenogram of the skull disclosed no diagnostic abnormality.

Despite the absence of significant changes in the x-rays of the skull, the long history with few signs indicative of any marked increase in intracranial pressure over a 2-year period suggested the presence of a slowly growing tumor. However, the clinical evidence of extensive involvement, sufficient to include the leg, arm, and face area, as well as enough of the temporal lobe to involve the optic tracts, was rather difficult to visualize unless the lesion were situated deeply enough to include the internal capsule. Accordingly, ventriculography was carried out. On the left side the ventricle was reached at about 5 cm. from the surface and the
fluid, although under increased pressure, was normal in appearance. On the right, the ventricle could not be reached and no unusual resistance was encountered.

Films taken after the introduction of about 15 cc. of air into the left ventricle showed the ventricular system to be shifted to the left (Fig. 2). On the left side the entire ventricle could be outlined but on the right, only a small amount of air had entered the system, visualizing the posterior portion of the body, the occipital, and the temporal horns. There appeared to be a massive lesion occupying the region comparable to the anterior two-thirds of the body of the right ventricle.

Operation. Craniotomy was carried out under novocaine anesthesia and a large osteoplastic flap was reflected to include the posterior portion of the frontal as well as the upper temporal regions. The bone flap was centered over the motor cortex and the base of the flap was placed moderately low in the temporal region. The exposed dura was under moderate tension and in the central portion there could be seen a very large dark vessel which elevated this portion of the dura above the surrounding surface.

The dural flap was reflected with the base toward the superior longitudinal sinus. Upon opening the dura the brain bulged considerably and presented a remarkable appearance. The entire central portion of the exposure was covered by a mass of tortuous and coiled vessels which varied in diameter from 1 mm. or less to the largest, the exposed surface of which measured about 1 cm. in width (Fig. 3). The latter vessel corresponded to the position of the central vein and was partially buried in the cerebral substance. This vessel had a reddish-blue color, did not pulsate visibly, and followed the course of the central fissure to disappear into the substance of the brain just short of the upper edge of the exposure. Proximally it disappeared into the lip of the Sylvian fissure. The individual vessels varied in appearance between that of arteries and veins but about two-thirds of those composing the mass were of a peculiar reddish-blue color. A few of the vessels pulsed noticeably but as a whole the lesion did not show the degree of pulsation that is usually present in the typical arterial type of angioma. In the central portion of the lesion the cortex was completely covered by the mass of vessels but at the periphery the vessels were slightly smaller, somewhat less closely applied, and the adjacent cortex had a fine capillary network on the surface.

Just anterior to the lip of the Sylvian fissure was an area measuring approximately 2 cm.
ARTERIO-VENOUS ANGIOMA OF BRAIN

in diameter which was the site of vascular thrombosis and scarring. The pia-arachnoid was thickened and had a gray-white color with considerable yellow and brown pigment present. This area was moderately firm to palpation and had the appearance of a lesion that had been present for some time. A similar area was present just anterior to the upper third of the

![Image of reconstructed drawing of lesion found at operation. Note the dilated central channel, the variable character of the vessels and the two areas of thrombosis. Dotted line indicates site of cortical incision. Inserts show site of bone flap and method of exposing hematoma.](image)

enlarged central vein and involved the vessels of the varix at the anterior edge of the tumor mass.

The brain itself appeared to be moderately soft and palpation disclosed no firm tumor, nor did the introduction of a ventricular needle at the periphery of the lesion locate an area of abnormal resistance. Because the degree of herniation of brain appeared to be greater than could be accounted for by the varix itself, a small incision was made in the cortex at the anterior and superior edge of the varix. At a depth of about 3–3 cm. a large currant-jelly type of clot was encountered; about 50–60 cc. of this material were removed. The remaining cavity
appeared to underlie the upper two-thirds of the motor cortex and biopsy of the wall disclosed no evidence of tumor. Following removal of the clot there was considerable hemorrhage from an area just under the mid portion of the varix and this required the use of a considerable amount of fibrin foam to control. Nothing was done to the varix itself and the wound was closed allowing a large decompression in the temporal region.

**Postoperative Course.** This was uneventful. Eight days after operation the patient had slight voluntary movement in the upper extremity and this progressed slowly. He was discharged from the hospital on the 15th postoperative day.

When seen approximately 6 months later the area of decompression was soft and pulsed quite markedly. For the first time a loud systolic bruit could be heard on auscultation of the operative site as well as in the right carotid region. The optic discs showed no elevation but the margins were moderately blurred. The retinal vessels appeared to be slightly distended but there was no evidence of a vascular tumor of the retina. There was a moderate degree of voluntary movement in the right lower extremity, but the improvement in the function of the right upper extremity had not progressed very far.

**DISCUSSION**

The complete clinical syndrome of the arterio-venous angioma as described by Cushing and Bailey consists of (1) intracranial bruit; (2) increased extracranial vascularity (vascular scalp, carotid hypertrophy, and cardiac hypertrophy); (3) papilledema; and (4) convulsive seizures often with focal symptoms and frequently with long periods of remission. However, as Noran has pointed out, intracranial bruit and increased intracranial pressure are not common findings. It must be remembered that Cushing and Bailey emphasized this variable feature of the bruit.

Inasmuch as the vessels forming the lesion are closely related to or partially composed of those vessels that normally supply the brain, interference with many of these vessels may lead to infarction of additional cortex. Also, indiscriminate closure of venous channels draining the mass of vessels may lead to a considerable retrograde distension of the deeper veins with subsequent rupture of the overlying cortex or further impairment of circulation to the brain substance.

In addition to the symptoms listed above, these lesions may cause acute manifestations by several means. Subarachnoid hemorrhage, such as described by Noran, may occur in association with or possibly as the prodromal phase of an intracerebral or intraventricular hemorrhage. Intrinsic thrombosis with subsequent further impairment of the peripheral circulation may result in the occurrence of focal symptoms while, as has been noted above, massive hemorrhage may precipitate acute intracranial pressure. In the case described above, the relation of the thrombosis of portions of the lesion to the history of focal twitchings involving the left side of the face and left upper extremity is not clear.

It has been pointed out that massive hemorrhage into the ventricular system is the usual cause of death in a patient suffering from this type of lesion, but the case described here illustrates the fact that extensive hemorrhage is not necessarily followed by fatality and that rupture may occur into the substance of the brain rather than into the ventricular system. According to Craig and Adson, Cushing was probably the first to actually remove an intracerebral clot. Since his report in 1903 a considerable literature has grown with many reports of successful evacuation of an intracerebral hematoma. A perusal of the literature, however, reveals no instance in which the hematoma was secondary to an angiomatic lesion. In Cushing's case the hemorrhage was secondary to hypertension and although the patient survived the operation he died 2 weeks later from pneumonia. Russell and Sargent reported a case in 1909 and here again the hemorrhage was apoplectiform. Most of the intracerebral hematomas described in the literature can be placed into one of three etiological groups as follows: (1) post-traumatic, immediate or delayed; (2) secondary to hypertension and/or arteriosclerotic cardiovascular disease; and (3) of unknown etiology. Naffziger and Jones reported 3 cases in 1928, all being of the delayed post-traumatic type. Penfield in 1933 reported 2 cases, one of a young boy in whom the hemorrhage occurred in relation to excessive...
vomiting following smoking. In the second patient, the hemorrhage was apparently due to hypertension.

Pilcher\(^9\) has recently reported 8 cases of hematoma, 3 of which were post-traumatic and 5 of unknown etiology. Of the latter, one patient was known to have arteriosclerotic heart disease. Craig and Adson\(^3\) in 1936 reported 9 cases of operative evacuation of an intracerebral hematoma. Of these, 2 patients had a definite history of trauma, 1 developed symptoms following a week-end of strenuous activity, and a 4th, with symptoms of 4 months duration, had an injury 12 years previous to operation. In 4 other cases the cause of the hemorrhage was unknown, while in the other the hemorrhage was probably related to rheumatic fever and malignant endocarditis. Wilkins\(^14\) two cases were both of traumatic origin.

In most of the cases reported recovery has been the rule and when death has occurred following operation it has usually been the result of complications or related to the etiological condition rather than the hemorrhage itself. Pilcher\(^9\) reported 2 deaths: one patient was a 62-year-old man who died of unknown cause 6 months after operation; the other was a 56-year-old man with arteriosclerotic heart disease, who had recurrence of symptoms 2 months after operation and died 7 months later. Of the group reported by Craig and Adson\(^3\) one patient died on the 9th postoperative day, possibly from cerebral hemorrhage, and the other died of endocarditis 3 months after operation.

It is to be noted that removal of the hematoma in the case described here was followed immediately by severe bleeding, apparently from the original site of hemorrhage. It seems likely that the pressure developed by the hematoma itself was sufficient to occlude the bleeding vessel, preventing massive hemorrhage with subsequent death. In retrospect, examination of the x-rays of the skull showed an area of bone absorption which corresponded with the upper portion of the large central vessel in the tumor-mass (Fig. 1). However, there were no changes in the skull to suggest the presence of a vascular lesion.

SUMMARY

A case has been described in which an intracerebral hematoma resulting from rupture of a deep vessel of an arterio-venous angioma (hamartoma) was removed surgically.

It has been pointed out that acute symptoms of intracranial pressure in the presence of a vascular lesion may be caused in several ways and that intracerebral hemorrhage may not necessarily prove fatal.

REFERENCES

A NEW METHOD FOR THE CONTROL OF HEMORRHAGE IN CEREBRAL SURGERY

MASON TRUPP, M.D.

Department of Neurological Surgery, Washington University Medical School, and Barnes and St. Louis Children's Hospitals, St. Louis, Missouri

(Received for publication July 11, 1946)

“All surgeons who make for themselves opportunities to observe the manipulative work of their fellows must appreciate the present tendency toward the abandonment of the applauded methods of comparatively few years ago.” With this statement Dr. Harvey Cushing in 1911 introduced a paper on the control of bleeding in operations for brain tumors read before the American Surgical Association.1

In the field of neurological surgery, few if any useful methods have been relegated to the scrap heap and many neurological surgeons, during the course of a single operation, may make use of all possible aids to control hemostasis from the scalp incision until the last suture has been tied.

In no other type of surgery does bleeding so obscure the clear view essential to the safety of delicate manipulations as are required in surgery of the central nervous system, unless it is cardiac surgery. White’s observations have disclosed that the loss of blood in major neurosurgical operations is much larger than had been suspected, and his estimates of the amount lost, from 1 to 2 liters, were confirmed by Webster at the University of Pennsylvania.4

In order to minimize the loss of blood, the neurological surgeon has summoned to his aid numerous useful methods. Among them are hemostasis with various clamps and tourniquets, the application of weights on pendent instruments thus allowing the galea by mere weight to close the bleeding points, the use of novocain fortified with epinephrine, bone wax to stop bleeding from diploic spaces of bone, muscle fragments, cottonoid pledgets and strips, silver and tantalum wire clips, gauze, ligatures, hot irrigating solutions, gelatine sponge, thrombin, electro-coagulation and suction.²

With all these methods ready at hand the neurological surgeon’s ability occasionally remains challenged by an unsatisfactory anaesthesia. Notwithstanding the use of all these devices for the control of hemorrhage, an occasional case may be encountered in which all fail and consequently prolong the operation. To overcome this a new device has been developed which has been tried out by Dr. Sachs and is now being used on the neurosurgical service at Barnes Hospital.

An apparatus of simple construction has been designed to speed up a clear-view exposure of bleeding points so that electro-coagulation can be rapidly performed, and at the same time eliminate many cumbersome, obscuring and often elusive cottonoid strips. The method makes use of a controlled jet of sterile compressed air delivered at the bleeding site, permitting precise exposure of severed vessels for electro-coagulation. The apparatus consists of a series of bottles containing antiseptic solutions to wash the compressed air, and the conventional sucker tip is used for its delivery. The volume of air pressure is controlled by a flow-meter which delivers 12 to 15 liters per minute.