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Dural Arteriovenous Malformation—With Special Consideration to Its Nature and Treatment

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Abstract

Seven cases of dural arteriovenous malformation are reported with analysis of the symptoms, hemodynamics, treatment and follow-up results.

The present disease is thought to be progressive and should be definitely distinguished from the intracerebral AVM and traumatic arteriovenous fistula since the hemodynamics of arteriovenous shunt showed extensive and dynamic changes with the lapse of time as observed from the results of long-term follow-up studies.

The best treatment is to primarily block arteriovenous shunt at the dura and tent as much as possible.

Key words: Arteriovenous malformation, dura mater

Introduction

Dural arteriovenous malformation[7,8] should be definitely distinguished from other intracranial arteriovenous malformations from the viewpoint of symptoms and hymodynamics of arteriovenous shunt. In view of the fact that the site of arteriovenous shunt. In view of the fact that the site of arteriovenous shunt is not inside the brain but in the supra or subtentorial dura, the falx cerebri, or the tent, the above name seems to best express the pathological condition of this disease. The authors have experienced 7 cases of this disease up to date and, a report with discussion of its nature and treatment is hereby presented.

Materials (Table 1)

The 7 cases which were experienced in the Department of Neurosurgery, Tohoku University by the end of July, 1976 should be definitely diagnosed as dural arteriovenous malformation (abbreviated as dural AVM hereafter). These seven cases represent 4.6% of the total 150 cases of intracranial AVM.

Representative case report

Case 1. This 51-year-old housewife gradually developed rotary vertigo and a machinery tinnitus since 1961, at the age of 47. In July 1965 when she was first admitted to our department, thrill and bruit over the occipital artery bilaterally, mild exophthalmos and bruit in the right ocular region, and bilateral mild optic atrophy were noted. On carotid arteriograms taken in 1965, both the left and right occipital arteries were abnormally enlarged with tortuosity, showing a shunt to the transverse sinus but no arteriovenous fistula was observed in the area of the internal carotid arteries (Fig. 1).

The vertebral arteriography revealed an abnormally enlarged and torturous vertebral artery which was shunted to the transverse sinus.

Ligation of the bilateral external carotid arteries was performed in August 1965. Headache and tinnitus improved, and she returned to her housekeeping for about 2 years. However, she was readmitted on March 19, 1968 with complaints of right pulsating exophthalmos and progressive dementia. On bilateral carotid arteriograms, the right side demonstrated an abnormally enlarged tentorial artery. On the left the periphery of the posterior temporal artery was also shunted to the transverse sinus (Fig. 2). Thus, the vascular image was greatly changed from the previous one. On subclavicular arteriograms the ascending cervical artery of the thyreocervical trunk showed a shunt to the transverse sinus bilaterally. Since it was

Table 1 Summary of

Case	Name	Age	Sex	Initial symptoms	Symptoms	Recent head injury	Neurological findings	Ophthal-logical findings
1	M. T.	51	Female	Vertigo Tinnitus	Tinnitus Headache Vomiting Visual dis-turbance Rt. pulsating exophthalmus Progressive dementia	—	Progressive dementia	Rt. exophthalmus Blt. optic atrophy
2	Y. M.	44	Female	Headache	Tinnitus	—	N.P.	N.P.
3	S. F.	63	Male	Visual disturbance	Tinnitus Nuchalache Visual disturbance	—	N.P.	Blt. choked disc Rt. Coat's disease
4	Y. Y.	41	Male	Headache	Tinnitus Headache Impaired vision Convulsion	—	N.P.	Blt. choked disc
5	F. K.	75	Male	Tinnitus Occipitalgia	Tinnitus Occipitalgia	—	N.P.	N.P.
6	T. E.	67	Female	Headache	Severe headache Lt. hemiconvulsion Vomiting caused by the ruptured aneurysm	—	Lt. hemiparesis and hemi-hypesthesia caused by the ruptured aneurysm	N.P.
7	T. M.		Male	Headache Lt. hemi-paresis	Lt. hemiparesis Tinnitus Headache Motor aphasia	—	Lt. hemi-paresis Lt. hemi-hypesthesia	Blt. optic atrophy

the cases (Dural AVM)

Oto-laryngological findings	Feeding arteries visualized on the angiogram	Operation	Result and the period of follow-up study
Thrill Bruit	Blt. occipital arteries Blt. posterior branches of middle meningeal arteries Blt. marginal tentorial arteries Blt. ascending cervical arteries Rt. dorsal meningeal artery Lt. ascending pharyngeal artery Blt. posterior meningeal branches of the vertebral artery	Ligation of blt. external carotid arteries Blt. occipito-suboccipital craniectomy Clipping of feeding arteries at blt. dura mater and tentorium Ligation of blt. thyreo-cervical trunks	Died 6 months after discharge by progressive dementia
Thrill Bruit Blt. conductive deafness	Blt. occipital arteries Blt. posterior branch of middle meningeal arteries Blt. marginal tentorial arteries Blt. lateral tentorial arteries Blt. ascending cervical arteries Blt. dorsal meningeal arteries Blt. ascending pharyngeal arteries Rt. posterior meningeal branch of the vertebral artery	Ligation of blt. external carotid arteries Blt. occipito-suboccipital craniectomy Clipping of feeding arteries at blt. dura mater and tentorium Dural plasty at the upper and under portion of transverse sinus by artificial dura mater	Improved 3 years and 6 months after discharge
Thrill Bruit Blt. perceptive deafness	Blt. occipital arteries Blt. posterior branch of middle meningeal arteries Rt. marginal tentorial artery Rt. lateral tentorial artery Rt. ascending cervical artery Rt. ascending pharyngeal artery	Ligation of blt. external carotid arteries Separation of periosteum from occipital bone	Decreased in visual acuity 2 years after discharge
Bruit	Rt. occipital artery Rt. marginal tentorial artery Blt. ascending cervical arteries Rt. ascending pharyngeal artery Blt. posterior meningeal branches of the vertebral arteries	Ligation of rt. external carotid artery Separation of periosteum from suboccipital bone Rt. suboccipital craniectomy Ligation of rt. thyreo-cervical trunk Ventriculo-peritoneal shunt	Died due to deterioration after convulsion 3 years after discharge
Thrill Bruit	Blt. occipital arteries Blt. posterior branches of middle meningeal arteries Blt. marginal tentorial arteries Lt. dorsal meningeal artery Lt. ascending cervical artery Lt. posterior meningeal branch of the vertebral artery	Ligation of blt. external carotid arteries Separation of periosteum from occipital bone	Unchanged 3 years and 9 months after discharge
N.P.	Rt. occipital artery Rt. marginal tentorial artery	(Clipping and wrapping of aneurysmal neck of the rt. IC·PC junction)	Unchanged 2 years after discharge
Thrill Bruit	Blt. superficial temporal arteries Lt. occipital artery Blt. posterior branches of middle meningeal arteries Blt. anterior meningeal arteries Blt. middle meningeal arteries Lt. marginal tentorial artery	Excision of anteromedial part of dura mater and falx cerebri, including superior and inferior sagittal sinus Ventriclo-peritoneal shunt	Unchanged 6 months after discharge

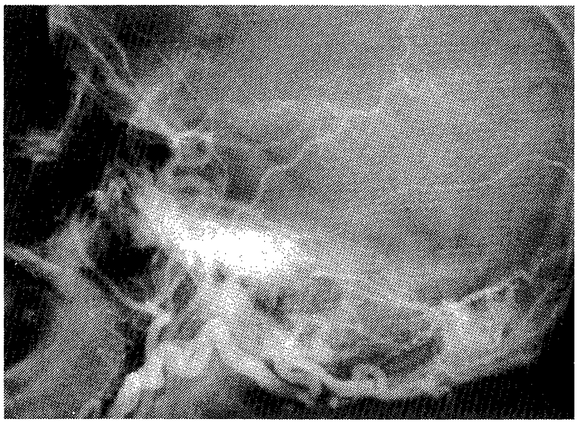

Fig. 1 Case 1. 51 y female
Left carotid angiogram
Shunt from the occipital artery and the posterior branch of middle meningeal artery into the transverse sinus can be seen.

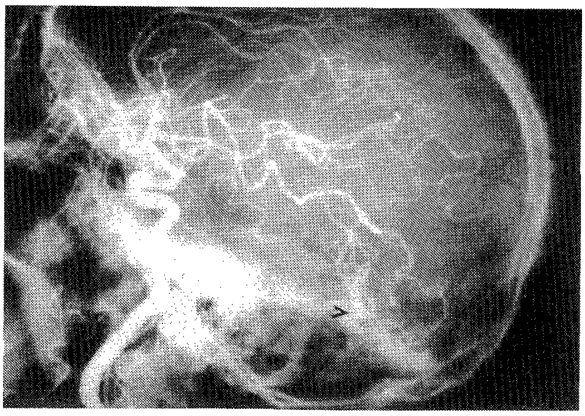

Fig. 2 Case 1. 51 y female
Left carotid angiogram after ligation of left external carotid artery. Shunt from the posterior temporal artery into a transverse sinus was found. (half arrow)

thought impossible to block and remove such a complicated arteriovenous shunt in a single procedure, its gradual decrease was proposed. The thyreocervical trunk was ligated first on the right side and then on the left, followed by a transverse incision of the neck for extirpation of abnormal vessels which branched out from the nuchal muscle branches of the vertebral artery and the ascending cervical artery. In addition, craniectomy of the left occipito-suboccipital region was performed, followed by that of the right occipito-suboccipital region to clip the tentorial arteries and the posterior branches of the middle meningeal artery on both sides as much as possible. These 7 operations resulted in recovery of spontaneous speech and in improvement of the right pulsating exophthalmos and vascular murmurs, and she was discharged, However, 6 months after discharge, she died of general debility in another hospital.

Case 2. This 44-year-old housewife was admitted to our hospital on December 18, 1972 with a complaint of tinnitus on the right of 4 months duration. Before her admission, on October 16, 1972 ligation of the right external carotid artery was performed in another hospital. On bilateral carotid angiograms, the tentorial arteries were shown bilaterally, and the occipital artery from the left external carotid artery was shunted to the transverse sinus (Fig. 3). The ascending cervical artery of the right thyreocervical trunk was well developed to flow into the transverse sinus (Fig. 4). On February 2, 1973 craniotomy of the bilateral occipito-suboccipital region was performed, and the arteriovenous shunts in the tent bilaterally were blocked by clipping. Furthermore, the tent was cauterized and partially removed adjacent to the transverse sinus to completely block the shunt flow. The supra- and subtentorial dura were replaced by an artificial dura and ligation of the left external carotid artery was simultaneously performed (Fig. 5). She was discharged on March 20, 1973 (Fig. 6). Follow-up studies are being performed every 6 months, and at present, 42 months after discharge, the vascular murmurs can no longer be heard. On angiogram, the tentorial arteries was reduced bilaterally, while the bilateral ascending cervical arteries were dilated again in the nuchal muscle.

Case 7. This 34-year-old male was admitted to

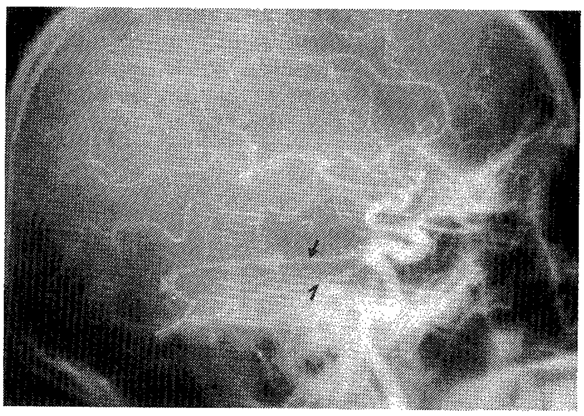

Fig. 3 Case 2. 44 y female
Right carotid angiogram after ligation of right external carotid artery. Lateral and marginal tentorial artery could be seen.
Dorsal meningeal artery was also found, marginal tentorial artery (arrow) lateral tentorial artery. (half arrow)

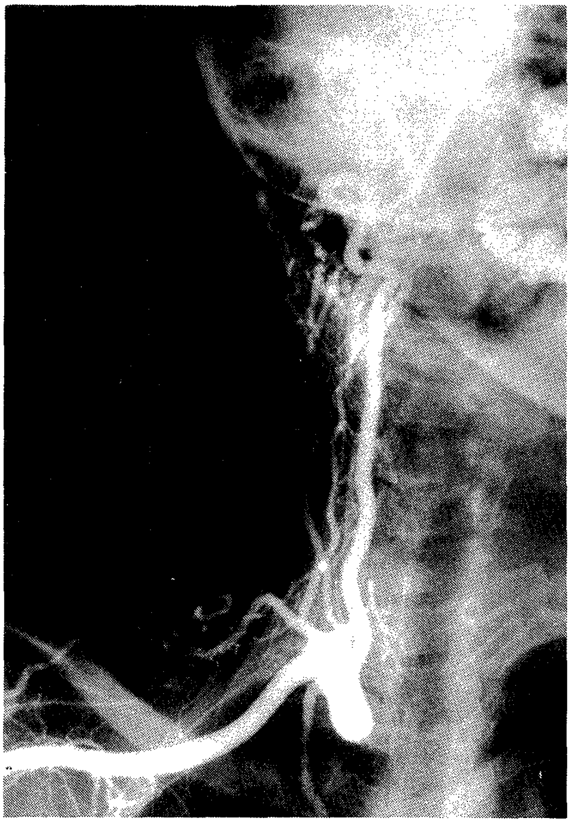

Fig. 4 Case 2. 44 y female
Right subclavial angiogram
Shunts from ascending cervical artery into the transverse sinus was found.

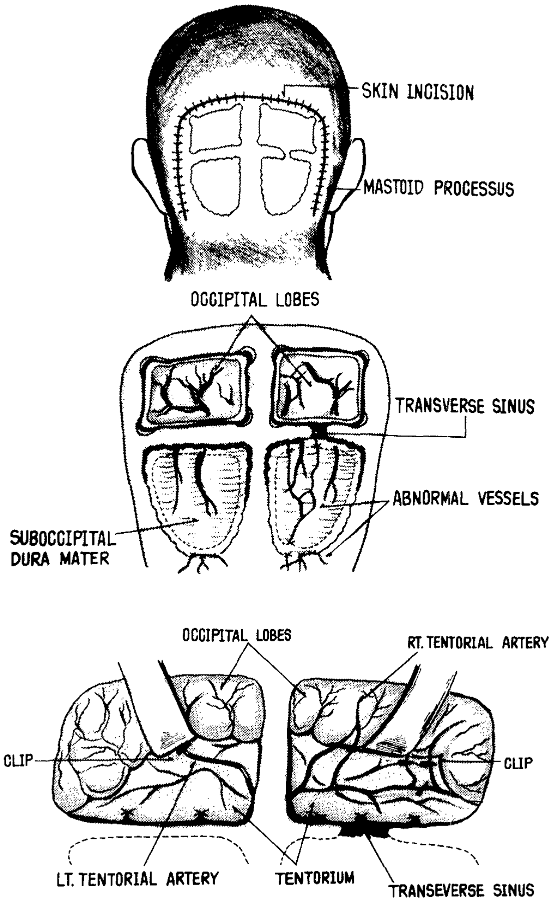

Fig. 5 Illustration of technique for clipping of feeding arteries at bilateral dura mater and tentorium.

our hospital on March 22, 1976, with progressive headache, left hemiparesis and motor aphasia. Besides these complaints, he had a 15-year history of bilateral dilated scalp arteries.

Upon admission, his mental function deteriorated and bruit was audible on the forehead bilaterally. Mild optic atrophy was observed in the bilateral ocular fundi. On bilateral carotid angiograms, superficial temporal arteries, anterior and middle meningeal arteries were shunted to the anterior part of the superior and inferior sagittal sinuses bilaterally. Left marginal tentorial artery and occipital artery were also visualized as additional feeders.

On the venous phase, reverse venous flow to the frontal cortical veins were demonstrated remarkably bilaterally (Fig. 7). On May 28, 1976 bilateral frontal craniotomy was performed. At operation, a group of dilated veins were noted in the bilateral frontal cortex and the anterior falx.

The feeding arteries, which came from the bilateral anterior ethomoidal arteries through the falx into the malformation were identified. The anteromedial part of dura mater and falx cerebri were excised including the superior and inferior sagittal sinuses to completely block the shunt flow.

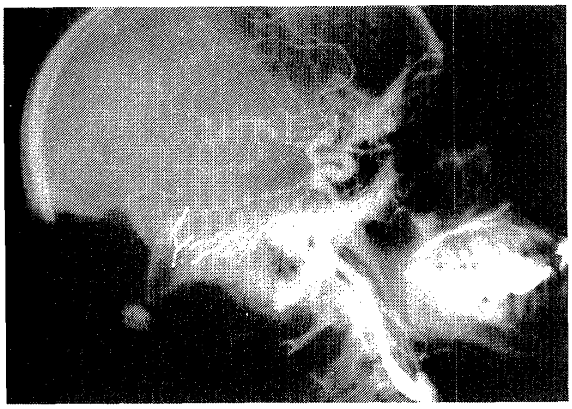

Fig. 6 Case 2. 44 y female
Right carotid angiogram after clipping of tentorial arteries.
Collateral pathways into the transverse sinus were slightly visualized.

The dura mater were replaced by Lyo. dura (human dura mater) (Fig. 8). Thirteen days after the radical operation, ventriculo-peritoneal shunt was carried out because of ventricular reflux in

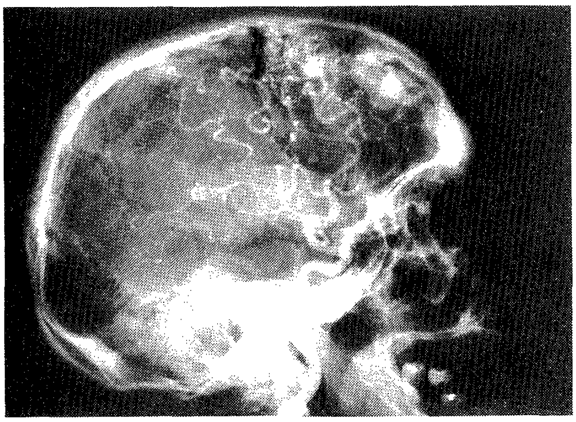

Fig. 7 Case 7. 34 y male
Right carotid angiogram
Reverse venous flow to the frontal cortical veins can be seen.

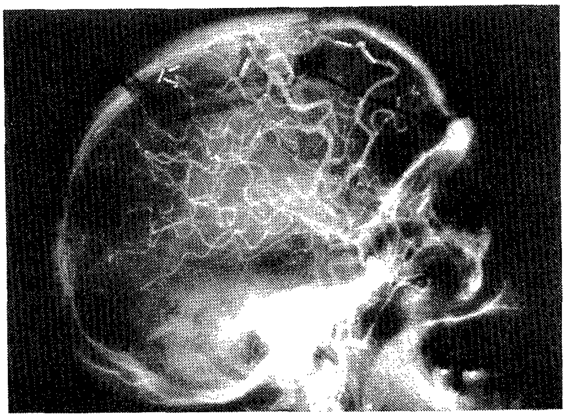

Fig. 8 Case 8. 34 y male
Right carotid angiogram after excision of anteromedial part of dura mater and falx cerebri, including of superior and inferior sagittal sinus.
Colateral pathways into the sagittal sinus cannot be seen.

cisternographic findings. He was discharged on July 19, 1976, and went to another hospital for rehabilitation. His mental functions remained unchanged.

Summary of the cases (Table 1)

Dural AVM of our 7 cases was located in the posterior cranial fossa in 6 cases and in the frontal falx in 1 case. Five of the 7 cases were males. The symptoms of dural AVM developed in and after middle age in 6 cases. Age upon admission ranged from 34 to 75 with a mean of 53. Two major symptoms at the onset were headache and tinnitus. One of the patients (Case 6) was hospitalized because of a headache attributed to subarachnoid hemorrhage from an aneurysmal rupture at the right internal carotid-posterior communicating junction, but showed no symptoms due to dural AVM itself. Tinnitus was observed in 6 of the 7 cases. Thrill was often palpable in the region of the extracranial feeders, and vascular murmurs were audible by auscultation in 6 of the 7 cases. Headache in the frontal and occipital regions was a frequent complaint but it was mild in degree. Visual disturbance was observed in 4 of the 7 cases. Funduscopic findings showed papilledema in 2 cases, optic atrophy in 2 and pulsating exophthalmos in 1 (Case 1). A noteworthy symptom was that patients fell gradually into dementia in the course of several years due to cerebral ischemia ensuing the growth of the arteriovenous shunt, as noted in Case 1, Case 4, and Case 7. Neuroradiologically, the plain x-ray of the skull demonstrated enlargement of the arterial markings of the feeders was observed in 3 cases. Examination of the hemodynamics of arteriovenous shunt in 6 cases of posterior cranial fossa showed that the main feeding arteries were as follows: 1) The tentorial branch of the right internal carotid artery was only the feeder in 1 case (Case 6); 2) In addition to the bilateral external and internal carotid arterial systems, the left thyreocervical trunk was the feeder in 1 case (Case 5); (3) In addition to the right external and internal carotid arterial systems, the ascending cervical artery of the right thyreocervical trunk was the feeder in 1 case (Case 4); 4) In addition to the bilateral external carotid arteries, the right internal carotid artery and thyreocervical trunk were the feeding arteries in 1 case (Case 3); and (5) The feeders were all main arteries above the neck, namely, the bilateral external carotid arteries, the nuchal muscle branches of the vertebral artery, and the thyreocervical trunk in 2 cases (Case 1 and 2). On the other hand, bilateral superficial temporal arteries, bilateral anterior and middle meningeal arteries and right marginal tentorial artery were the feeders in the case of frontal falx (Case 7).

The draining vein was the transverse sinus or the sigmoid sinus in 6 cases of posterior cranial fossa. However, the sinuses such as the straight sinus and superior sagittal sinus and cerebral and cerebellar veins were visualized as reverse flow in 2 (Cases 1 and 4) of 6 cases. On the other hand, superior and inferior sagittal sinuses were the drainers in the case of frontal falx.

Operation was performed on the 6 cases (Case 6 underwent operation for aneurysm alone) by various methods: 1) Ligation of the external carotid artery; 2) Blocking of arteriovenous shunts by detachment of the skin and periosteum of the posterior fossa; 3) Blocking of arteriovenous shunt by craniectomy of the posterior fossa; 4) Blocking of the tentorial artery and the middle meningeal artery in the dura and tent; 5) Blocking of the thyreocervical trunk; 6) Blocking and extirpation of the muscle branch and dural branch of the vertebral artery and the ascending cervical artery of the thyreocervical trunk in the nuchal muscles; 7) Excision of dural venous sinuses including dura mater and falx (Case 7); and 8) Shunt operation for cerebrospinal fluid. These procedures were performed independently or in combination.

In the 6 operated cases, the hemodynamics of arteriovenous shunt and preoperative symptoms were varied in degree. Postoperatively all of them showed transient improvement of tinnitus, visual acuity, and headache. However, 2 of the 6 cases died due to aggravation resulting from general debility in 1 case and due to deterioration 3 years after discharge in 1 case. The other 4 cases are being observed in follow-up studies—24 to 42 months at present. Two of them (Cases 3 and 5) showed recurrence of symptoms and re-opening of the shunt tracts by angiography in follow-up studies. In histological findings of the dura and abnormal vessels sampled in 4 cases during operation the intradural arteries showed abnormal dilatation, or a large artery with sparse elastica interna due to abnormal thickening of the intima, suggesting the growth process of AVM where a normal artery was once dilated due to an abnormal blood pressure, leading to secondary degeneration.

Discussion

The 7 cases present showed no past history of trauma and they are definitely thought to be dural arteriovenous malformation as described by Newton.[9,7]

Most of our cases had onset in and after the middle age, in close agreement with reports by Houser,[3] Nicola,[8] and Newton.[7]

Characteristic symptoms were tinnitus and vascular murmurs. As another feature, perfusion disorder in the cerebral blood circulation due to an elevated venous pressure, provoked papilledema and optic atrophy. It is interesting that in the case with a remarkable volume of shunt (Case 1), pulsating exophthalmos occurred. It is noteworthy that the hemodynamics of arteriovenous shunt tends to show extensive and dynamic changes with the lapse of time, accompanied by a progressive dementia.

Although Houser[3] stated that there is no such large volume of shunt as causing cerebral ischemia in this disease, the progressive dementia in our Cases 1, 4 and 7 was attributed to nothing but cerebral ischemia due to arteriovenous shunt or cerebral venous congestion. Therefore, we feel that it is necessary to follow-up the prognosis of the present disease for at least several years after discharge.

In the examination of the pathogenesis, angiography, and operative methods of this disease, it is important to understand the arteries feeding the dura mater.[3]

As for 6 cases of the dural AVM of posterior cranial fossa, the meningohypophyseal trunk, the dorsal meningeal artery, the lateral tentorial artery and the marginal tentorial artery were characteristically visualized as the feeders of dural AVM in 11 of Newton's 16 cases[7] and in all of our 6 cases. On the other hand, the dural branch from the occipital artery and the ascending pharyngeal artery are highly significant as the feeder. The shunt from the occipital artery occurs in most of the present disease, as it was observed in 13 of Newton's 16 cases and in all of our 6 cases. Ascending pharyngeal artery was visualized in 3 (4 sides) of our 6 cases and in 2 of Houser's 12 cases.[3] It is generally believed that the dural branch from the extracranial vertebral artery feeds the falx cerebelli and subtentorial dura. In most of our cases, the nuchal muscle branch of the vertebral artery developed abnormally and showed net-like anastomosis with the occipital and ascending cervical arteries as indicated by Storrs[10]. Another important feeder is the posterior branch of the middle meningeal artery.

In summary, every dural and tentorial branch in the posterior cranial fossa can be the feeders in this disease. Therefore, it is not as simple as can be interpreted under the term[1,2,6,9,11] extracranial artery-intracranial venous sinus fistula, but it is an extensive disease where all main arteries above the neck can be feeder via every route in typical cases.

On the other hand, in cases of dural AVM of frontal falx, every dural branch of the anterior half of cranial cavity can also be the feeders.

It is evident in view of the characteristics of the feeders that surgical treatment of this disease has many problems. However, there are few reports on the detailed method of operations. Based on our cases and in other reports, the surgical methods for the present disease are roughly divided into: 1) to block the feeder at a rather remote site proximal to the arteriovenous shunt such as the external carotid artery and thyreocervical trunk; 2) to block the feeder at a relatively near site to the shunt by craniotomy or ablation of the skin and periosteum alone; (3) to clip. all of the tentorial and or dural branches of the internal carotid artery at a nearest site of the shunt[2,5] After the craniotomy; 4) to excise dural venous sinuses such as the sigmoid sinus, the transverse sinus and the superior sagittal sinus.[4,13]

The first is a simple method. However, as in Case 1, the longer the distance to the site of shunt, the easier the collateral pathways are formed. Although the second method completely blocks the dural branches from the extracranial feeders, the tentorial branches of the internal carotid artery and dural branches develop postoperatively as feeders, as in Case 3. The third and fourth methods are the most ideal radical operations as illustrated in Cases 2 and 7. By the third method, however, it is not always easy to block all of the tentorial and dural arteries in the dura, tent and falx. The fourth method should be restricted to cases affected unilaterally in case of posterior cranial fossa. On surgically treating our 7 cases, we repeated trial and error several times at the beginning of our study. At present, we perform craniotomy in relatively young cases, as in Cases 2 and 7, in an attempt to block all feeders. In cases with poor general conditions and senile cases, such as Cases 3 and 5, we performed only detachment of the skin and periosteum of the occipital region in addition to ligation of the external carotid artery. Since this relatively simple method reduced the volume of shunt to some extent, though temporarily, we think that it should be tried as the first choice when age is a major factor.

Shunt operation of cerebrospinal fluid is attemped to decrease intracranial pressure. However, in view of the fact that the symptom of increased intracranial pressure in this disease is mainly due to perfusion disorder of the venous blood, shunting procedure should strictly be a supplementary means.

Now, what should be discussed is the nature of this disease. Since there are cases in which as long as the disease persists, the hemodynamics of arteriovenous shunt tends to enlarge and progress, accompanied by gradual aggravation of symptoms. Even tumorous changes are possible. Histologic findings of abnormal vessels in the dura were abnormal enlargement of arteries and thickening of the internal wall, as in other reports,[12] and tumorous components were not detected in our investigations of 4 cases. Further study on this subject is necessary.

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