Intracerebral Arteriovenous Malformation Supplied by Ethmoidal Arteries

—Case Report—

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Abstract
An intracerebral arteriovenous malformation supplied by bilateral ethmoidal arteries in a 51-year-old male is described. Operation revealed a vascular conglomerate on the cerebral surface which formed an arteriovenous shunt. The feeding artery and draining vein were clipped and incised, and the malformation was removed en bloc. An intracerebral or extracerebral vascular anomaly supplied by the ethmoidal artery is thought to be derived from a similar developmental fault, in which an arteriovenous shunt becomes trapped in an extradural location, the dura mater, the subdural space, or the cerebral parenchyma, during later fetal development.

Key words: ethmoidal artery, dural arteriovenous malformation, intracerebral arteriovenous malformation, anterior cranial fossa

Introduction
Dural arteriovenous malformations (AVMs) in the anterior cranial fossa are a classification of a vascular anomaly located in the anterior cranial fossa, and supplied by the ethmoidal arteries and drained by intradural veins. The incidence of this anomaly is far less than that of dural vascular anomalies occurring in the cavernous sinus and transverse sigmoid sinus. More than 30 cases have been reported, three with nidi on the cortical surface or in the cerebral parenchyma. We report a new case of a cerebral AVM supplied by the ethmoidal arteries and discuss the pathogenesis of these cerebral vascular anomalies compared to the extracerebral lesions.

Case Report
A 51-year-old male suddenly experienced severe headache while playing golf. Computed tomography performed in a nearby hospital disclosed an intracerebral hematoma in the right frontal lobe. He was referred to our hospital for further evaluation and treatment.

He had no significant past medical history. There were no neurological deficits on admission. Headache was the only symptom. Right internal carotid angiograms revealed a vascular malformation in the anterior cranial fossa, supplied by the ethmoidal artery via a dilated ophthalmic artery. The cortical draining vein ran upward along the cerebral convexity and flowed into a dural vein, which ran away from the midline laterally. The left ethmoidal artery also supplied the vascular malformation through the interethmoidal anastomosis (Fig. 1). There was no contribution from the external carotid arteries.

Opening the dura mater via a bifrontal craniotomy disclosed the dilated erythematous cortical vein. Following this vein proximally led to a vascular conglomerate on the frontopolar cerebral surface. An artery originating in the frontal base flowed into the nidus (Fig. 2). This feeding artery was clipped and incised. The nidus was then dissected from the brain. Small pial arteries, probably peripheral branches of the anterior cerebral artery, flowed into the nidus. Finally, the draining vein was clipped and incised. The vascular malformation was removed en bloc.

The postoperative course was uneventful. Angiography demonstrated no vascular anomaly postoperatively. Histological examination showed...
that the malformation consisted of small arteries and thickened dilated veins.

**Discussion**

Dural AVM in the anterior cranial fossa typically occurs in males in the sixth decade, about 10 years older than dural AVMs occurring in other areas. Cranial bruits are seldom heard. Most patients present with intracranial hemorrhage, except where found incidentally. Excellent operative results have been achieved with only rare severe neurological deficits.

Our patient had a vascular conglomerate on the cerebral surface which formed an arteriovenous shunt. The term cerebral AVM is thus more appropriate. Three other cases with similar operative findings have been reported. The nidi were generally embedded in the cerebral parenchyma with only a very small proportion exposed on the cortical surface. Tanaka et al. reported a case with the nidus on the surface. A branch of the anterior cerebral artery supplied the lesion in addition to the ethmoidal arteries. Tiyaworabun et al. also found a nidus supplied by small branches of the anterior cerebral artery at operation, which was not demonstrated by angiography.

Streeter first described the differentiation and development of the vascular system in human embryo, citing five distinct stages. The primitive vascular plexus differentiates into primitive arteries, capillaries, and veins during the second stage. The development of the three layers of vessels, superficial, dural, and pial, occurs in the third stage. Cerebral AVM is generally accepted as a congenital condition, derived from faulty development of the cerebral vascular system during the second stage. However, the pathogenesis of dural AVMs is controversial. Some authors have suggested that these lesions are acquired, especially in the cavernous sinus and transverse sigmoid sinus. Recently, the term dural arteriovenous fistula rather than malformation has been applied to this entity.

We considered the possibility that cerebral AVM and dural AVM supplied by ethmoidal arteries may have a different pathogenesis, the former congenital and the latter acquired. These two entities are indistinguishable clinically and radiologically, except when a contribution from the anterior cerebral artery is shown angiographically. We examined the relationship between the nidus and the dura mater in previously reported dural AVMs in the anterior
cranial fossa with detailed operative findings.\textsuperscript{1-5,7-12,19,23,26} Ishimitsu \textit{et al.}\textsuperscript{7} resected an erythematous draining vein with aneurysmal dilatation in the extradural space, suggesting that the arteriovenous shunt was located extradurally. This was the only case in which the operation used the extradural route, all others were treated by intradural procedures. In four patients, abnormal vascular conglomerates were attached to the internal surface of the dura mater.\textsuperscript{2,3,12,24} In another two patients,\textsuperscript{5,21} a group of abnormal vessels lay between the dura mater and the cerebral surface. Abumiya \textit{et al.}\textsuperscript{1} found an abnormal vascular nest invaginated into the frontal lobe at the base of the skull, which was similar to those of Terada \textit{et al.} \textsuperscript{21} and Tiyaworabun \textit{et al.}\textsuperscript{22} Katayama \textit{et al.}\textsuperscript{9} and Takeda \textit{et al.}\textsuperscript{19} did not refer to the nidus, but operative findings suggested that the vascular anomalies might be simple arteriovenous fistulas with a direct shunt between the ethmoidal artery and the intradural vein. Other authors\textsuperscript{8,10} found that the dura mater was involved in the nidus and resected the basal dura, but no histological examination of the resected dura was reported. These previous reports suggest differences between dural AVM in the anterior cranial fossa and in the cavernous sinus or transverse sigmoid sinus. In the latter, the dura mater itself forms an arteriovenous shunt, containing abnormal vasculature. In contrast, the nidus in the former lies outside the dura mater, although attached in many cases.

Olivecrona and Ladenheim\textsuperscript{17} suggested that the relationship of the malformation to the dura mater is determined during the third stage of development of the cerebral vascular system. The location of the nidus is variable in the dural AVMs supplied by the ethmoidal arteries in the anterior cranial fossa, and cerebral AVMs fed by the ethmoidal arteries are also observed. Therefore, an intra- or extracerebral vascular anomaly supplied by the ethmoidal artery is derived from a similar developmental fault, in which an arteriovenous shunt becomes trapped in an extradural location, the dura mater, the subdural space, or the cerebral parenchyma, during later fetal development. Anatomical descriptions of more cases are required to better define this unusual entity.

\textbf{References}


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