Arteriovenous Fistula at Scalp: Rapid Progression After Embolization of Contralateral Facial Arteriovenous Malformation

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Multiple craniofacial arteriovenous malformations (AVM) are rare, and they are hemodynamically related to each other by complex and intercommunicating vascular networks. This report presents a case with two craniofacial AVMs located in anatomically different regions. One AVM changed in flow amount and venous drainage route after embolization of the other one, which may suggest a new guideline for treatment of multiple craniofacial AVMs.

Key Words : Multiple; Arteriovenous malformation; Arteriovenous fistula; Craniofacial; Embolization

An arteriovenous malformation (AVM) of the face or scalp is an abnormal connection between the feeding arteries and draining veins, without the normal capillary bed in the subcutaneous layer of the scalp. The draining veins may be grossly dilated and tortuous, and may show variceal dilatation (1-5). It is generally thought that AVM arises from multiple developmental defects causing the primitive capillary bed to fail to persist (6, 7). It is also thought that increased arterial pressure and flows lead to opening and dilatation of normal, latent AV shunts. Local ischemia and minor trauma also plays a role in enlargement of arteriovenous fistulas (AVF). Multiple craniofacial AVMs occurring in one patient are extremely rare, and only a few cases with multiple systemic malformations such as hereditary hemorrhagic telangiectasia or Rendu-Osler-Weber disease have been reported (8). Multiple

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Tel. 82-2-2290-9153 Fax. 82-2-2293-2111 E-mail: yjleeee@hanyang.ac.kr Neurointervention 2010;5:36-39 craniofacial AVM may be hemodynamically related to each other by complex and intercommunicating vascular networks.

This report describes a case with two craniofacial AVMs located at anatomically different regions. Interestingly, we found rapid progression of a scalp AVM after embolization of the other AVM on the contralateral side of the face. This may reflect a hemodynamic relationship between the AVMs located at anatomically different regions, and may suggest a new guideline in treatment of multiple craniofacial AVMs.

CASE REPORT

A 55-year-old man presented with a 4-month history of tinnitus in the left ear and pulsatile headache. He had been treated for diabetes and hypertension for 4 years; otherwise he had no underlying disease. MR imaging of the neck showed a highly vascular mass in the left preauricular region. The initial digital subtraction angiogram revealed two AVMs located at the left preauricular region and right frontal scalp, respectively. The left one had a complex nidus fed mainly by the left superficial temporal artery (STA), and was larger in flow amount (Fig. 1A). The right one appeared to be a fistulous type fed by the right middle meningeal artery

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Arteriovenous Fistula at Scalp

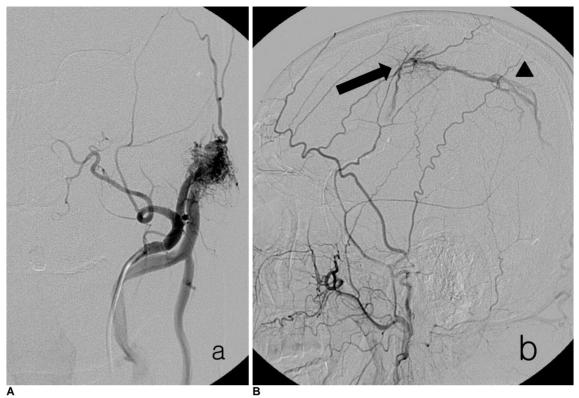


Fig. 1. Left ECA(External carotid artery) angiogram (**A**) showing high-flow AVM fed by STA at the left preauricular region. Right ECA angiogram (**B**) showing small AVF (arrow) fed by MMA and STA at the right frontoparietal scalp. Note that the main venous drainage (arrow head) is to the parietal region and secondarily to the temporal region.

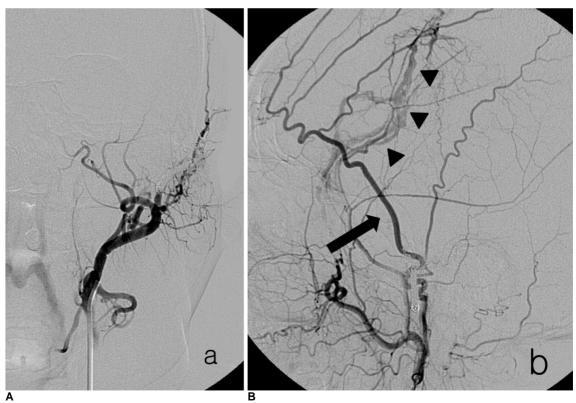


Fig. 2. Left ECA angiogram (**A**) after embolization with glue and coils, showing significantly decreased shunt flow. Follow-up right ECA angiogram (**B**) showing a significant increase in the amount of shunt flow compared to the initial angiogram (Fig.1B). Note that STA size (arrow) and venous drainage routes (arrow heads) are also changed.

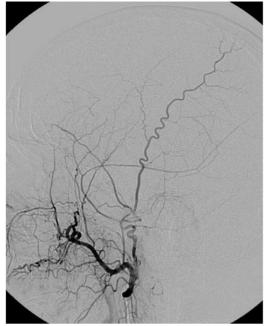


Fig. 3. 2-month follow-up angiograms of right ECA (after glue embolization) showing no residual or recurring AVM.

(MMA) and STA (Fig. 1B). Initially, transarterial embolization was performed at the left STA with nbutyl cyanoacrylate (NBCA) and coils to reduce shunt flow, because the left one was symptomatic and had much higher flow on angiogram. However, though the pulsatile tinnitus was much improved, complete occlusion of AVM could not be achieved by endovascular treatment alone (Fig. 2A). The asymptomatic right AVM was scheduled to be treated at the next session. The follow-up angiogram after 5 weeks showed significant changes of the right scalp AVM, including remarkably increased shunt flow and a change in venous drainage course (Fig. 2B). This AVM could be completely treated by transarterial embolization with NBCA (Fig. 3). The left facial AVM was surgically removed 2 months later, because pulsatile tinnitus recurred.

DISCUSSION

Unlike intracranial AVM, craniofacial AVM is rare. Treatment is difficult for several reasons. For example, craniofacial AVM has high shunt flow with complicated vascular anatomic connection, and also involves cosmetic problems. Thus there has been no consensus on the treatment of craniofacial AVM. In the past, the treatment of choice for AVM in the scalp and face was surgical excision or ligation of the feeding arteries for a long time (9). However, with progress in endovascular surgical technique, embolization has become an integral part of the treatment of these malformations. Cure of these lesions may be attained by embolization alone in some patients, or by embolization followed by surgical removal (10).

In the cases of multiple AVMs, the most symptomatic and hemodynamically active one should be the first one to be treated (8). But in our case, a small and asymptomatic AVM turned into a larger and symptomatic one after treatment of the other AVM. The hemodynamics of multiple craniofacial AVMs is unknown; however, they may be closely related to each other by complex and intercommunicating vascular networks. We know of no past studies that describe a change in flow amount and venous drainages route of a craniofacial AVM after manipulation of another craniofacial AVM. We postulated a mechanism that caused the change in the hemodynamic environment or its balance in our case. First, after embolization of the major feeder of one AVM, the other AVM's shunt flow and feeding artery was progressively enlarged. Like other vascular systems, the scalp has many collateral pathways. After occlusion of the proximal portion of the left STA, which was the main feeder, the right STA might have increased its flow to compensate for the reduced flow in the region formerly fed by the left STA. Increase in flow of the right STA might eventually have caused the increase in shunt flow. Second, we found changes in the venous drainage route of the right AVM after embolization of the left one. The reduction in flow of the AV shunt of the left AVM after embolization may have caused an ipsilateral venous pressure decrease. This may have influenced the venous drainage route of the contralateral scalp, although it unclear how.

The case presented here could provide a new guideline for establishing a treatment strategy for multiple craniofacial AVMs. Because of their close hemodynamic relationships, even small AVMs could change quite rapidly after treatment of another AVM. Thus the size and symptoms should not be the only guideline in treatment of multiple craniofacial AVMs, and all of them should be carefully considered for treatment.

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Arteriovenous Fistula at Scalp

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두피 동정맥루 : 반대측 두경부 동정맥루 색전술 이후 발생한 혈류역학적 변화

한양대학교 의과대학 영상의학교실

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다발성 두경부 동정맥루는 드문 혈관 질환이며, 서로 복합적으로 연결된 혈관 조직으로 혈역학적인 연관관계를 가지고 있다. 저자들은 해부학적으로 다른 위치에 발생한 두개의 두경부 동정맥루의 치료 과정 중 하나의 동정맥 루에 대한 색전술 이후 나머지 다른 하나의 동정맥루의 혈류의 변화를 확인할 수 있었던 한 증례를 보고하고자 한 다.

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