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Therapeutic Embolization of the Dural Arteriovenous Malformation Involving the Jugular Bulb

Pulsatile tinnitus is a rarely occurring symptom of vascular origin. Most frequently, the symptoms are due to an arteriovenous malformation, to a tumor of the jugular glomus or to a local arterial stenosis. A 39-yr-old Korean male suffering from pulsatile tinnitus of the left ear was diagnosed to have dural arteriovenous malformation of the jugular bulb. Magnetic resonance imaging and angiography revealed a high-velocity vascular lesion encroaching the internal jugular vein and sigmoid sinuses. Digital subtraction angiography demonstrated a dural arteriovenous malformation involving the jugular bulb. The arterial supply was from the neuromeningeal branch of the left ascending pharyngeal artery and inferior tympanic artery. Stenosis of the left jugular vein caused retrograde venous drainage through the contralateral transverse sinus. Superselective embolization of these feeding arteries was successfully performed using 25% mixture of N-butylcyanoacrylate and lipiodol. In postembolization period, his complaints of pulsatile tinnitus and buzzing noise behind his left ear disappeared.

Key Words : *Tinnitus; Arteriovenous Malformations; Embolization,Therapeutic*

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INTRODUCTION

Dural arteriovenous malformations (dAVMs) consist of arteriovenous shunts confined within dural leaflets and account for 10-15% of all intracranial arteriovenous malformations (1). The transverse-sigmoid sinus area is the most common location of dAVMs in the literature (2, 3), but the cavernous sinus area is much common in Korean series (4). However, dAVM involving the jugular bulb is extremely rare. Symptoms and signs of the dural arteriovenous malformations can include pulse-synchronous tinnitus, bruit, headache, papilledema, hemorrhage, proptosis, visual decline, altered mental status, and transient or permanent neurological deficits (1, 5).

The cause and pathogenesis of dAVMs remain unclear. A few cases reported in children suggest that these dAVMs are congenital (6, 7), but dAVM is generally thought to be an acquired lesion (5, 8-10). The association between dAVMs and sinus thrombosis is well recognized (10-12), but a clear cause and effect has not been proven.

Direct surgical resection of dAVM has become an accepted treatment, but significant hemorrhage can result (13). Advances in microcatheter and guidewire technology have made superselective embolization of feeding arteries technically easier.

The authors report a case of dAVM in the jugular bulb with pulsatile tinnitus as the presenting symptom, which was treated successfully with therapeutic embolization.

CASE REPORT

A 39-yr-old man presented with a 5-yr history of left ear tinnitus. There was no history of head trauma. A neurological examination at the time of admission revealed pulsatile bruit on the left retromastoid area, but not demonstrated lower cranial nerve symptoms.

Examination

Magnetic resonance imaging (MRI) revealed a jugular bulb that had signal void dots on T2-weighted images (Fig. 1A), which was homogeneously enhanced with gadolinium contrast media (Fig. 1B). Magnetic resonance angiography (MRA) demonstrated a vascular lesion of the sigmoid sinus-internal jugular vein junction area (Fig. 1C). MRA source images showed that the high-velocity vascular lesion was located on the jugular bulb (Fig. 1D). Left external carotid angiography revealed an early visualization of sigmoid sinus and jugular vein by dAVM supplied especially from inferior tympanic artery and neuromeningeal branches of the ascending pharyngeal artery of the left external carotid artery (Fig. 2A, B). Vertebral angiography showed no definite feeding vessel to dAVM (Fig. 2C, D). The internal jugular

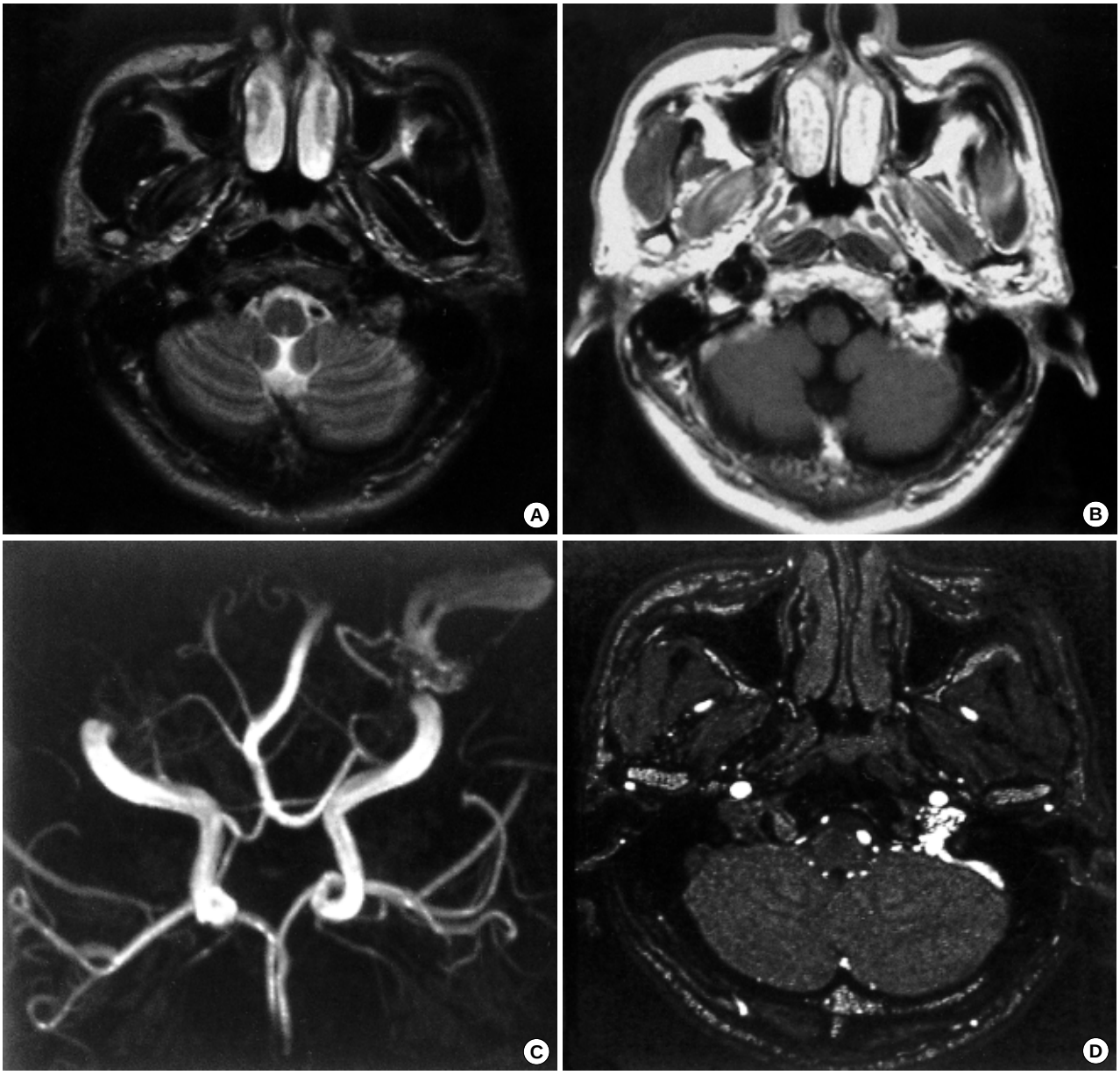

Fig. 1. Magnetic resonance imaging reveals a jugular bulb that had signal void dots on T2-weighted image (A), which was homogeneously enhanced with gadolinium contrast media (B). Magnetic resonance angiography (MRA) demonstrates a vascular lesion of the sigmoid sinus-internal jugular vein junction area (C). MRA source images shows that the high-velocity vascular lesion was located on the jugular bulb (D).

vein and sigmoid sinus were partially occluded on venous phase angiogram and left venous drainage was diverted to the contralateral sinuses. From a transarterial approach, 0.010-inch microcatheters (Prowler, Cordis, FL, U.S.A.) were navigated into the above feeding arteries from the external carotid artery, which were then embolized with 1.8 mL 25% mixture of N-butylcyanoacrylate (NBCA, Braun, Melsungen, Suisse) and lipiodol (Fig. 3A-E). The embolized glue was well located at the intranidal portion (Fig. 3F, G). Follow-up external carotid angiogram demonstrated no dAVM and no visualization of the early venous drainage (Fig. 3H). The patient was free from tinnitus after embolization and exhibited no neurological deficits including lower cranial nerve signs.

DISCUSSION

Dural arteriovenous malformations (dAVMs) are unique vascular abnormalities comprising numerous tiny connec-

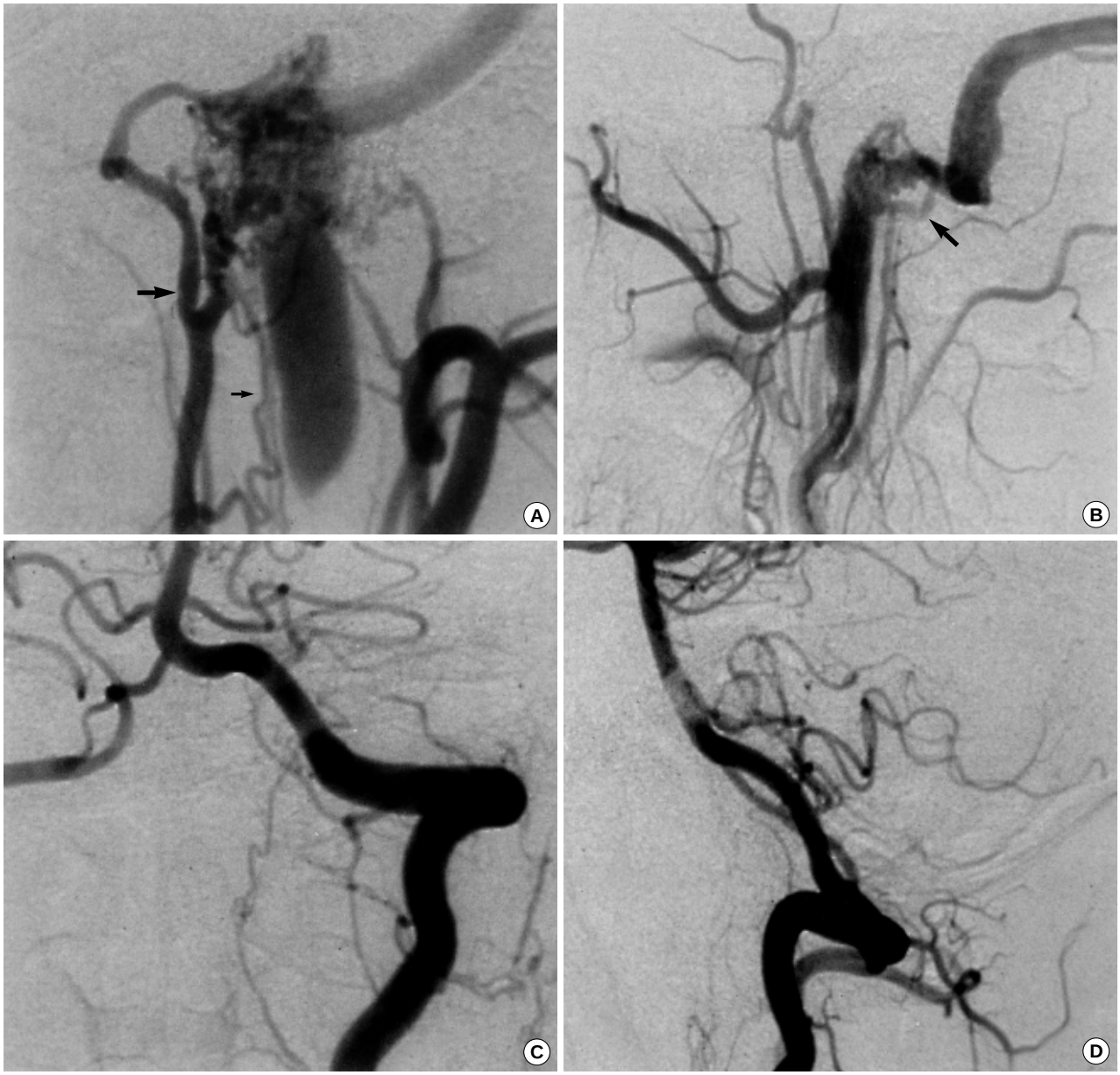

Fig. 2. Left external carotid angiograms (A: anteroposterior, B: lateral view) reveal an early visualization of sigmoid sinus and jugular vein by dural arteriovenous malformation supplied from inferior tympanic artery (short arrow) and neuromeningeal branches (long arrow) of the ascending pharyngeal artery of the left external carotid artery. Vertebral angiograms (C: anteroposterior, D: lateral view) reveal no definite feeding vessel to dAVM.

tions between branches of dural arteries and a venous sinus with the nidus. Essential defining features are the nidus of the malformations and the early appearance of venous structures during the arterial phase of angiography (2). The most common location of dAVMs is in the transverse-sigmoid venous sinuses and the cavernous sinus (2-4). However, dAVM involving the jugular bulb that is the internal jugular vein/sigmoid sinus junction area has not been previously documented. Its clinical presentation is similar to that of the tumor of jugular glomus. Harris et al. (14) have reported a dAVM or a jugular glomus tumor as a possible cause of pulsatile tinnitus. The audible sound may result from turbulence in the blood stream, secondary to a change in the blood velocity as it passes through an arteriovenous fistula or a highly vascularized tumor of the jugular glomus. In the present case of a dAVM, the intraluminal stenosis itself might be responsible for the turbulence.

The cause and the pathogenesis of dAVMs are controversial, but are classified into two major categories, congenital and acquired. dAVMs have a very high incidence of abnor-

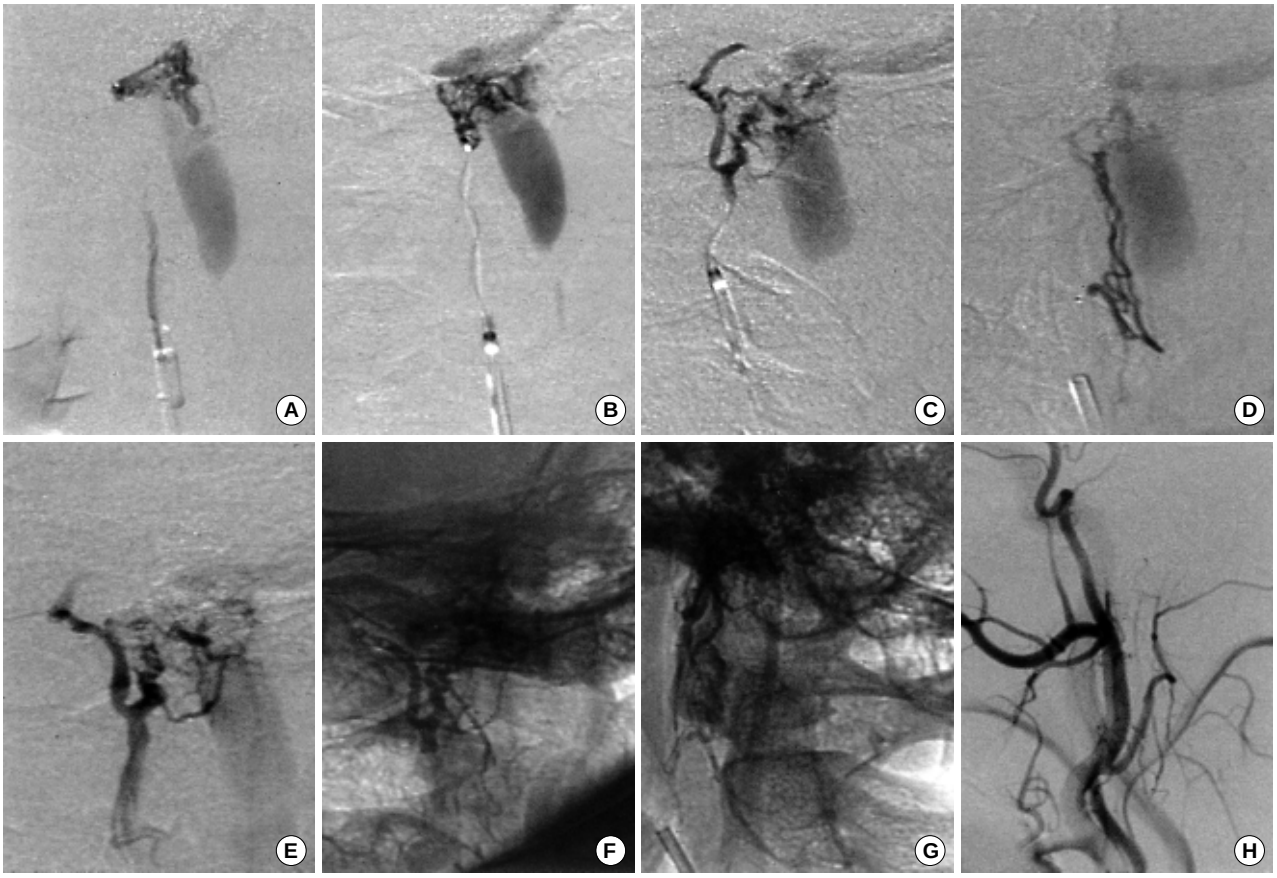

Fig. 3. From a transarterial approach, 0.010-inch microcatheters were navigated into the neuromeningeal branches (A, B, D, E) and inferior tympanic branch (C) from the external carotid artery, which were then embolized with 1.8 mL 25% mixture of N-butylcyanoacrylate and lipiodol (A-E). The embolized glue was well located at the intranidal portion (F, G). Follow-up external carotid angiograms (H: lateral view) demonstrate no dural arteriovenous malformation and no visualization of the early venous drainage.

malities of venous channels. These abnormalities are described as irregularity and rigidity of the wall, stenosis and septation with filling defects in the lumen, and retrograde flow with partial or complete occlusion of the draining venous sinus or vein. Such changes are believed to be due to venous thrombosis, which is thought to be responsible for the occasional disappearance of these lesions (15, 16). Thrombosis or thrombophlebitis of the draining vein or sinus appears to be a common feature of these lesions and may be related to their origin (5). Awad et al. (2) classified the progression of dAVMs into three stages. They speculated that sinus thrombosis and the opening of embryonic arteriovenous communication played an important role in the onset of this disease. This hypothesis was supported by sinus thrombosis with tumor (17, 18), trauma (19), and craniotomy (20). Nishijima et al. (10) proposed that the mechanisms of the progression of dAVM are as follows: 1) a single arteriovenous fistula forms in the dura near a venous sinus; 2) arterial blood begins to flow into the sinus via communicating dural veins, leading to the formation of thrombosis; 3) pressure in the venous sinus increases, and this increase in internal pressure along with the organization of thrombi causes fibrous thickening of the sinus intima and interruption or proliferation of the elastic lamina of the sinus; 4) stenosis or occlusion of the sinus lumen occurs. In our case, there was no thrombosis of the sinus, but stenosis of the internal jugular vein and sigmoid sinus was found. Our case can be explained with Nishijima's hypothesis. However, it does not prove that there is a cause and an effect.

The therapeutic strategy for dAVMs includes transarterial embolization (21), transvenous embolization (22), and surgical resection of the involved sinus (13, 23). Placement of embolic materials within the nidus of the malformation can result in cure. Halbach et al. (21) reported that only 59% of patients treated was completely cured by transarterial embolization. Although transvascular embolization of these pathways is technically possible, the risk of embolic reflux and stroke makes these less desirable. If embolic material flows through the nidus and occludes venous drainage, aggravation of symptoms may result with diversion of venous drainage into cortical pathways. Although surgical excision of the involved sinus is possible, massive intraoper-

ative hemorrhage can occur because of the rich vascularity of the surrounding structures (13). We used a transarterial approach with the placement of embolic materials, liquid adhesives, within the nidus. Because of the high arterial pressure within the involved sinus, there is often retrograde drainage of blood away from the sinus into cortical veins. Disconnection is important, if liquid adhesive embolic agents alone are contemplated, to prevent the flow of embolic agents into these cortical veins. When these connections have been interrupted and the sinus isolated by occluding its outflow, complete stasis is often observed after the injection of contrast material or embolic agents. To prevent this potentially devastating complication and eliminate the need for surgical interruption of draining veins metal coils was often placed into the affected sinus with transvenous approach. Our case is not a proper candidate for coil embolization with transvenous approach because venous channels are already stenotic and venous hypertension is aggravated.

In conclusion, we report a case of dAVM involving the jugular bulb with mild stenosis of the internal jugular vein and sigmoid sinus. The dAVM completely disappeared after transarterial embolization with liquid adhesive.

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