

*Case Report*

Occult Pulsatile Tinnitus in Association with a Prominent Vein of Hypoglossal Canal: Two Case Reports

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The pathway of the vein of hypoglossal canal has not been mentioned in venous origin pulsatile tinnitus. We wished to clarify the possible complications related to this venous variant. We described 2 rare cases of troublesome pulsatile tinnitus associated with prominent vein of hypoglossal canal, which communicates with the jugular bulb and the marginal sinus. Both cases were successfully treated by positioning a stent across the vein of hypoglossal canal and jugular bulb. The 2 present clinical cases represent the first report of such a condition. The therapeutic decision-making is discussed in relation to the persistent pulsatile tinnitus and the etiopathologic hypothesis put forward.

KEYWORDS: Vein of hypoglossal canal, Jugular bulb, Pulsatile tinnitus**■ INTRODUCTION**

Pulsatile tinnitus of vascular origin results from turbulent blood flow that is caused by increased flow volume or lumen irregularity (4,9,19). Arterial causes of pulsatile tinnitus can be differentiated from venous causes by gently compressing the internal jugular vein ipsilateral to the tinnitus (7). This maneuver results in immediate resolution of tinnitus of venous origin but amplifies that of arterial origin. The venous causes of pulsatile tinnitus include diverticulum or aneurysm of the transverse-sigmoid sinus (7,11,12,18), prominent or stenotic sigmoid sinus (5,8,13,14,16), superficial lateral course of sigmoid sinus (10,17), jugular bulb diverticulum (5,14), high dehiscent jugular bulb (2), abnormal mastoid emissary vein and abnormal posterior condylar emissary vein (3,6). Although pathologic causes for the symptoms are always given strong consideration, normal variations should also be included in a possible differential diagnosis. We describe 2 cases of pulsatile tinnitus associated with a vein of hypoglossal canal shown by cerebral angiography.

■ CASE REPORTS**Case 1**

A 35-year-old woman, with no other medical history, presented with a 3-year history of a persistent right-sided pulsatile roaring tinnitus. The bruit interfered with her sleeping and daily function. Otoscopic and neurological examinations were normal. Compression of the right internal jugular vein produced subjective resolution of her symptoms. Magnetic resonance venogram (MRV) examination demonstrated a hypoplasia of the left transverse and sigmoid sinuses, prominent occipital sinus and vein of hypoglossal canal and jugular bulb. Digital subtraction angiography demonstrated the left dilated vein of hypoglossal canal (Figure 1A). The left transverse sigmoid sinus was hypoplastic.

Antiplatelet therapy consisting of orally administered aspirin (100 mg) and clopidogrel (75 mg) was initiated 3 days before the procedure. Endovascular procedure was performed under general anesthesia and systemic heparinization. During the procedure, heparin was given by intravenous infusion to maintain the activated clotting time at >2 times baseline,

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with a target of at least 300 seconds. Percutaneous access was gained through the femoral vein. Guiding catheters (8F) were advanced into the right internal jugular vein. Predilation with 4.0×20mm angioplasty balloon (Aviator, Cordis, USA) was performed at 8 atm. A 4.5mm-30mm Wingspan stent (Boston Scientific, USA) was introduced into the hub of the Vasco catheter (Figures 1B, C). Under road-mapping, the stent was then pushed through the microcatheter and aligned directly across vein of hypoglossal canal. After treatment, postoperative examination showed no neurological deficits and immediate resolution of her symptoms. She was maintained on aspirin and clopidogrel for 1 month and aspirin (100 mg per day) for 6 months. There was no recurrence of the pulsatile tinnitus after 6 months.

Case 2

A 22-year-old woman had a 2-year history of progressive right-sided pulsatile tinnitus. The patient had been in otherwise

good health with no similar previous episodes and no history of trauma. Physical examination revealed a normal tympanic membrane with no evidence of a vascular mass. No bruit was audible. No hearing deficit could be detected. Gentle pressure applied to the upper neck could stop the tinnitus. The pulsatile sound was not audible with a stethoscope. MRV showed an unusual drainage pathway of the torcular sinus, which drained into the occipital sinus, the marginal sinus and the vein of hypoglossal canal, and into the jugular bulb. Bilateral transverse sigmoid sinuses were hypoplastic. The cerebral venous drainage through the right vein of hypoglossal canal was excessively developed. Cerebral angiography confirmed these findings (Figure 2A).

A combination of 100 mg of aspirin and 75 mg of clopidogrel started 3 days before stent placement. The procedure was done under general anesthesia with monitoring by anesthesiologists. Transfemoral venous approach was performed and



Figure 1: **A)** Sinus phase of the left carotid artery injection demonstrating a hypoplasia of the left transverse and sigmoid sinuses with a prominent drainage pathway of occipital sinus, the marginal sinus and the vein of hypoglossal canal, and into the jugular bulb. **B)** X-ray film showing the stent was deployed. **C)** Sinus phase of the right carotid artery injection after stent placement showing a more laminar blood flow through the hypoglossal canal vein.



Figure 2: **A)** Sinus phase of the left carotid artery injection showing bilateral hypoplastic transverse and sigmoid sinuses. The cerebral venous drainage through the right occipital sinus, marginal sinus and vein of hypoglossal canal was excessively developed. **B)** Fluoroscopic image showing that a 4-mm×20-mm angioplasty balloon in the vein of hypoglossal canal was predilated. **C)** Sinus phase of the left carotid artery injection after deployment of a 6-mm×30-mm Solitaire stent showing more blood was shunted into the vein of hypoglossal canal.

an 8F guiding catheter was introduced into the internal jugular vein. A heparin bolus was given immediately before the interventional part of the procedure to increase the activated clotting time (ACT) to a minimum of 300 seconds. The vein of hypoglossal canal was predilated with a 4-mm×20-mm angioplasty balloon (Aviator, Cordis, USA) (Figure 2B). A 6-mm×30-mm Solitaire stent (Covidien, USA) was deployed across the vein of hypoglossal canal (Figure 2C). She was maintained on aspirin and clopidogrel for 1 month and aspirin (100 mg per day) for 6 months. There was no recurrence of the pulsatile tinnitus after 2 months.

■ DISCUSSION

The development of cerebral venous drainage pathways has been well depicted in human anatomic studies. The pathway of the vein of hypoglossal canal, however, has not been mentioned in venous origin pulsatile tinnitus. We wished to clarify the possible complications related to this venous variant. The vein of hypoglossal canal is cranial emissary vein, which communicates with the jugular bulb and the marginal sinus (15). The diameter of the vein of hypoglossal canal is usually as small as that of a silk thread. It occasionally may be as large as 2 to 4 mm, as shown by our series. The vein of hypoglossal canal can be so well developed that it can give a loud “venous” pulsatile bruit and a thrill.

One entity that has been described many times in the otolaryngologic literature but often forgotten is the essential type of tinnitus or cervical venous hum (9,13). Essential tinnitus is nearly always pulse synchronous in nature because of the distinct pulsation in the intracranial venous system, as well as in the adjacent extracranial venous system (13). Essential tinnitus is thought to be most likely related to the distortion of laminar flow within the internal jugular vein after compression of the vein by the adjacent transverse process of the atlas and sternocleidomastoid muscle when the head is turned, usually away from the side of the tinnitus (3,6,9). In the present case, the abnormal MRV findings and the lack of familiarity with the entity along with uncertainty as to whether this truly was the cause of the tinnitus prompted further investigation with cerebral angiography. The exact cause of the tinnitus and its pulsatile nature are difficult to understand. It is most likely related to turbulent blood flow within the tortuous venous sinus, as has been suggested in the case of a jugular bulb deformity (2,5,14) and similar to the turbulent flow expected within the abnormal emissary vein in the case of venous hum (3,6).

Therapeutic options in such a situation are limited. Occlusion of the sigmoid sinus or ligation of the jugular vein has been suggested if an expanding vascular mass has been excluded and adequate contralateral venous drainage is seen to be present (3,6). In this case, MRV and cerebral angiography demonstrated no significant flow through the transverse and sigmoid sinuses. Therefore, stentplasty of the vein of hypoglossal canal was offered to the 2 patients. This technique based on our much experiences in stenting treatment of venous sinus stenosis causing pulsatile tinnitus (1,8). We

postulate that by changing flow characteristics through the hypoglossal canal emissary vein accounting for the beneficial result. We must assume that in some manner the flow became more laminar as more blood was shunted into the emissary vein.

■ CONCLUSION

Prominent vein of hypoglossal canal represents a rare cause of pulsatile tinnitus and can be treated by stent deployment. MRV and cerebral angiography can be helpful in excluding a vascular anomaly, such as venous variations.

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