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CASE REPORT

Clinical Efficacy of Internal Jugular Vein Ligation in Treatment of Vascular Tinnitus

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Pulsatile tinnitus of vascular origin is one of the most common forms of this condition. Surgical management of the problematic lesion can be effective in vascular tinnitus. Here, we present three cases of pulsatile tinnitus of venous origin. Despite general tinnitus therapy and tinnitus rehabilitation therapy, tinnitus was persistent in these cases. Therefore, the patients were treated with ligation of the internal jugular vein (IJV). During angiography, the site for venous ligation was confirmed by compression of the internal jugular vein with the probe of a Doppler sonograph. The results in these cases are presented along with a review of the relevant literature. IJV ligation is thought to be an appropriate treatment modality in selected cases of venous tinnitus and Doppler sonography is a very effective and non-invasive method for determining the appropriate ligation level.

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Venous vascular pulsatile tinnitus is synchronous with the human pulse and may be due to turbulence in the blood flow, which is generated by increasing blood flow or stenosis of the vessel lumen. [1] If appropriate, surgical treatment can result in more rapid improvement of venous vascular pulsatile tinnitus than sensory neuronal tinnitus.[2, 3]

Here, we present the results in three cases of venous pulsatile tinnitus treated with ligation of IJV along with a discussion of the treatment of pulsatile tinnitus of vascular origin.

Case 1

A 37-year-old woman was referred to our tinnitus clinic with a 13-year history of a low-pitched sound in her left ear. At the time of referral, she was 7 months pregnant. The sound was pulsatile, described as "thump, thump," and was aggravated during exercise or in spring. She had no history of hearing loss, dizziness, earfullness, head or neck disease, drugs causing ototoxicity, or any other systemic disease. Japanese translations of the THI and visual analog scale of annoyance caused by tinnitus (VAS) were administered to evaluate the nature of the tinnitus. Her answers indicated that she suffered from tinnitus for half the day, except at bedtime, and the loudness score reached 8, the annoyance score 8, and the daily life impact score of 8 on the VAS (range: 0-10). No objective tinnitus around the neck, ear, or orbit could be heard using an electronic stethoscope ausculscope. The intensity of her tinnitus changed with head rotation or in the Valsalva maneuver. Gentle pressure on the left lower neck caused it to disappear almost completely. Some hypotympanic bluish shadow behind the normal tympanic membrane was seen through the otoscopic endoscope.

The results of blood tests, serologic tests, and urinalysis were normal. Some improvement of lower frequency air conduction hearing loss of the left ear in pure tone audiometry was observed after her tinnitus disappeared during compression of the left internal jugular vein (Figure 1). In tympanometry, both ears

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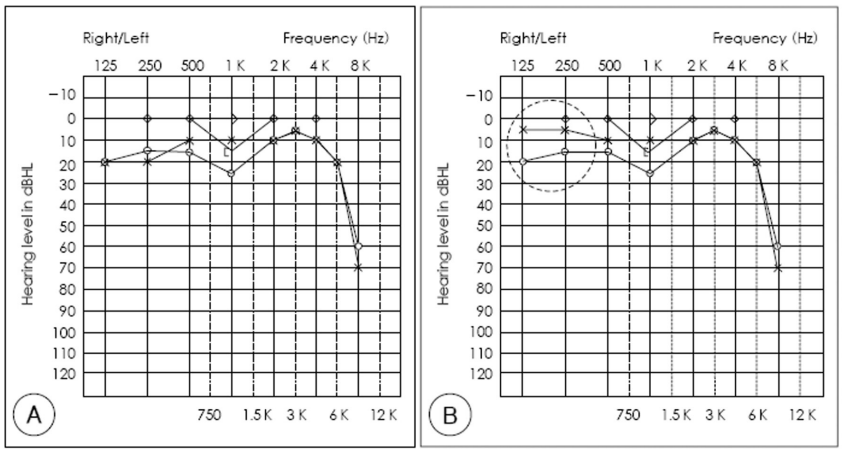

Figure 1. (A) Pure tone audiogram of patient 1 showing lower tone conductive hearing loss. (B) Pure tone audiogram with left internal jugular vein compression showing recovery of lower tone hearing loss (dotted circle).

were type A, and there was no wave. The results of auditory brainstem response test were normal. Her tinnitus was matched with the sound of 28 dB HL at 125 Hz in the tinnitus matching test. In temporal bone CT, a high jugular bulb with some bony dehiscence on the left side was identified (Figure 2A). Temporal MRI and brain MRA examination to exclude other diseases that could be responsible for the pulsatile tinnitus revealed no abnormalities except the high jugular bulb. Despite previous general tinnitus therapy and tinnitus rehabilitation therapy, her tinnitus was persistent. Therefore, we took into consideration the surgical method of ligation of ipsilateral IJV.

The results confirmed that the contralateral venous flow was normal on angiography when the left internal jugular vein was compressed, so we could minimize the risk of side effects such as increased intracranial pressure after ligation of the internal jugular vein. In addition, we identified the lowest level of the left internal jugular vein at which the tinnitus disappeared when venous flow was blocked by compression with the Doppler probe. The day after angiography, the left IJV was ligated at the fixed level under general anesthesia. After surgery, she still felt some tinnitus although the intensity was reduced compared to the preoperative level. Her tinnitus disappeared two weeks after surgery, and she reported neither annoyance nor discomfort on the VAS. She has been symptom-free for 28 months.

Case 2

A 21-year-old man complained of tinnitus in the right ear lasting for 3 years that was described as a “thump, thump” sound with a pulsatile pattern. He had no other audiological symptoms and no systemic disease. The same questionnaire as that described above indicated that he had experienced tinnitus for 40% of the day, except when sleeping, with the loudness score of 7, the annoyance score of 7, and the daily life impact score of 8 on the VAS (range: 0-10). The continuous bothersome sound prevented him from sleeping soundly. No objective tinnitus around the neck, ear, or orbit could be heard using an electronic stethoscope ausculscope. The tinnitus was enhanced with left-side head rotation and disappeared with right-side head rotation or in the Valsalva maneuver. Gentle pressure on the right lower neck caused it to disappear almost completely. Some hypotympanic bluish shadow behind the normal tympanic membrane was seen through the otoendoscopic examination.

The results of blood tests, serologic tests, and urinalysis were normal. All audiological examinations except the tinnitus matching test were normal. His tinnitus was matched with the sound of 36 dBHL at 125Hz. A high jugular bulb with some bony dehiscence on the right side was observed on temporal bone CT (Figure 2B). Temporal MRI and brain MRA examination revealed no abnormalities except the high

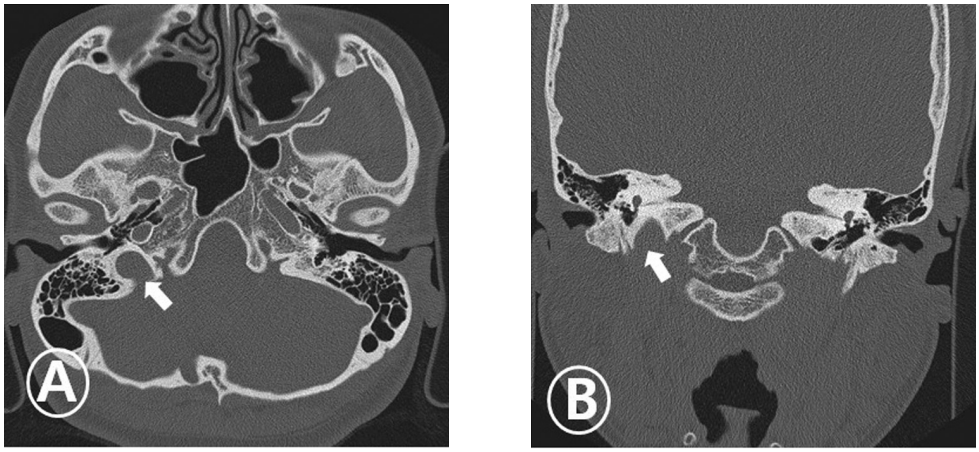

Figure 2. (A) Axial image of temporal bone CT shows bulging of the left jugular bulb into the tympanic cavity, consistent with high jugular bulb (arrow). (B) Axial image of temporal bone CT showing bulging of the right jugular bulb into the tympanic cavity, consistent with high jugular bulb (arrow).

jugular bulb.Despite tinnitus rehabilitation therapy for 1 year, his tinnitus was persistent. Therefore, we planned surgical ligation of the ipsilateral IJV.

Contralateral venous flow was normal on angiography after compression of the right internal jugular vein. In addition, we identified the lowest level of the right internal jugular vein at which the tinnitus disappeared when venous flow was blocked by compression with the Doppler probe (Figure 3). The day after angiography, the right IJV was ligated at the fixed level under general anesthesia (Figure 4). Complete resolution of the tinnitus occurred immediately and the patient has been symptom-free for 35 months.

Case 3

A 33-year-old woman complained of right ear tinnitus lasting for 7 months, which was reported to sound like "Drrr, Drrr" with a pulsatile pattern. She had no other audiological symptoms and no systemic disease. The same questionnaire as that described above indicated that she experienced tinnitus for 70% of the day except when sleeping, with the loudness score of [7], the annoyance score of [8], and the daily life impact score of [9] on the VAS (range: 0-10). She was unable to sleep due to the constant sound and suffered depression. No objective tinnitus around the neck, ear, or orbit could be heard using an electronic stethoscope ausculscope.

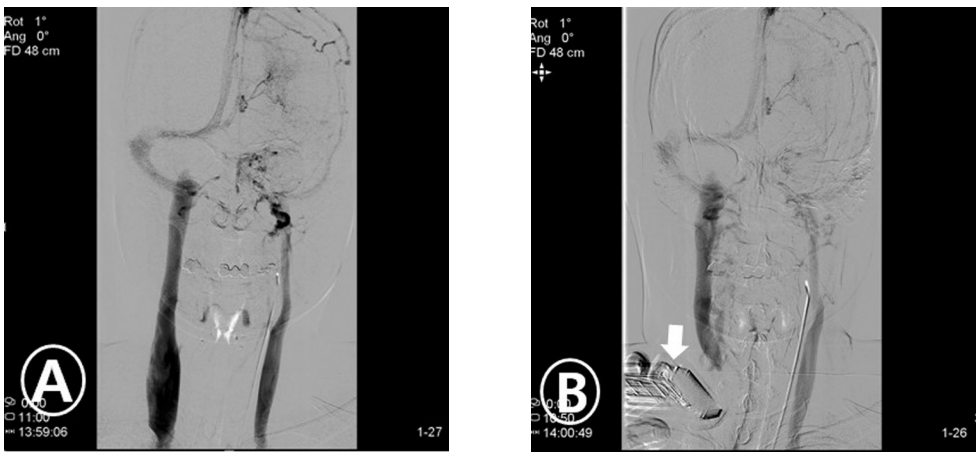

Figure 3. Dominant venous drainage of the right IJV was seen in four-vessel angiography (A). Preoperative surgical ligation site, the lowest portion that showed complete cessation of venous drainage and subjective pulsatile tinnitus by compression with the Doppler sonograph probe was selected during angiography (B: arrow).

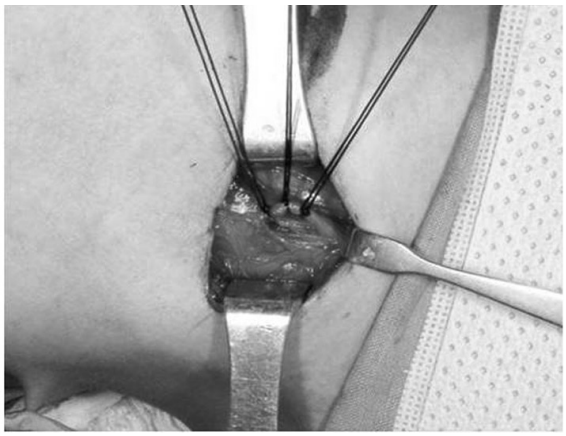

Figure 4. Right IJV ligation was performed with four 0-0 black silk sutures.

The tinnitus disappeared on right-sided head rotation or in the Valsalva maneuver and was increased on left side head rotation. Gentle pressure on the right lower neck caused it to disappear almost completely. Both tympanic membranes were normal on otoscopy. The results of blood tests, serologic tests, and urinalysis were normal. The results of all audiological examinations except tinnitus matching test were normal. The tinnitus was matched with 39 dBHL at 125Hz. Temporal bone CT, temporal MRI, and brain MRA showed no specific findings.

Previous general tinnitus therapy and tinnitus rehabilitation therapy were ineffective. Contralateral venous flow was normal on angiography after compression of the right internal jugular vein with the Doppler probe. In addition, we identified the lowest level of the right internal jugular vein at which the tinnitus disappeared in the manner indicated earlier. The day after angiography, the right IJV was ligated at the fixed level under general anesthesia. Complete resolution of the tinnitus occurred immediately and the patient has been symptom-free for 38 months.

Discussion

A vascular etiology is suggested in cases of pulsatile tinnitus synchronous with the pulse.[4] Pulsatile tinnitus may be heard around the neck, due to turbulent flow, by increased blood flow or lumen stenosis.[5,6] Pulsatile tinnitus can be classified into arterial type or venous type, according to the vessel of origin, and these two types can be easily differentiated by applying gentle digital pressure over the ipsilateral IJV. This maneuver has no effect on the intensity of the arterial type, whereas it causes the venous type to subside immediately.[1] However, Morrison reported that firm digital pressure over mastoid and suboccipital region reduced the intensity of tinnitus of dural arterivenous malformation.[7] Vascular pulsatile tinnitus can increase in both intensity and rate during exercise, and show changes in the intensity of tinnitus with head rotation.[8] In 1902, Mann demonstrated that head rotation decreased the venous return from the ipsilateral side of the brain and increased the flow in the contralateral jugular vein. The mechanism is a “propping open” of the contralateral jugular vein and includes squeezing of the ipsilateral jugular vein by the respective sternocleidomastoid muscles.[2,4] As vascular tinnitus may ensue from the turbulent flow of blood, ipsilateral head rotation, ipsilateral lateral neck compression, and the Valsalva maneuver will decrease blood volume, and so reduce the tinnitus.

Common causes of vascular pulsatile tinnitus include arteriovenous malformation, venous hum called essential pulsatile tinnitus, jugular bulb abnormality, aneurysm, arteriovenous fistula, vascular tumor, atherosclerotic disease, and pseudotumor cerebri syndrome.[1,4,6] In addition, the tinnitus tends to be aggravated under conditions of increased blood volume, such as anemia, hyperlipidemia, beriberi, valvular heart disease, hyperthyroidism, pregnancy, medication, or increased intracranial pressure.[4,8]

Diagnosis of vascular pulsatile tinnitus requires thorough physical examination, taking into consideration the remarkable features of tinnitus. Physical examinations should include otoscopy, auscultation of the ear canal, periauricular region, orbits, neck, and chest. In addition, neurological examination, including evaluation of the cranial nerve, audiological tests, such as pure tone audiometry, tympanometry, auditory brainstem response tests, and tinnitus matching tests, are required. In auscultation,

electronic stethoscope ausculscope or phonocephalography is more sensitive than a conventional stethoscope.[1,8] If the case is confirmed as objective tinnitus, the next step should be to determine the possible connection with the heartbeat.

In cases of hearing loss of 20 dB or more at lower frequency, patients with venous pulsatile tinnitus should undergo repeated audiography with gentle digital pressure over the ipsilateral IJV to determine any hearing improvement, as in case 1 in the present study.1 Some improvement of lower frequency air conduction hearing loss of the left ear on pure tone audiometry can result from disappearance of the masking effect of the tinnitus with compression of the left internal jugular vein. Tympanometry should be considered for patients suspected of tensor tympani myoclonus[1], and vascular lesions of the middle ear.[9,10] Its analysis sometimes can give useful information like the vibratory effect of the vascular lesion such as dehiscent jugular vein, which showed a fluctuation of pressure synchronous with pulse within the middle ear.[10]

Ultrasound study, temporal bone CT, temporal MRI, brain MRA and angiography can be helpful for patients in imaging studies.[6] Ultrasound study is non-invasive and very helpful for patients with atherosclerotic carotid or subclavian artery disease, or valvular disease. In particular, patients with carotid bruits should undergo carotid ultrasound study prior to any extensive radiological evaluation. Changes in tinnitus in patients with venous hum or high jugular bulb can be observed with compression of the ipsilateral IJV with an ultrasonic probe. At this point, we can predict the surgical results of IJV ligation; this makes it unnecessary to perform invasive angiographic balloon test, which can block the blood flow and result in loss of tinnitus.[3,11] In this study, the lowest site for venous ligation was confirmed preoperatively by compression of the IJV with a Doppler sonograph probe. We developed this method, which can reduce complications, such as increased intracranial pressure, facial edema, headache, and the probability of development of contralateral tinnitus, which can be caused by contralateral blood flow after ligation.[3]

In vascular pulsatile tinnitus, findings of the tympanic membrane on otoscopy may suggest the next step of imaging study.[1] Initial screening with temporal MRI and brain MRA are strongly recommended for patients with normal results on otoscopy. Temporal bone CT is less sensitive in brain vascular lesions. Carotid angiography should be strongly considered in patients with normal MRI and MRA to exclude dural arteriovenous fistula.[6] The diagnosis of intracranial dural arteriovenous fistula is not excluded with normal results from MRI/MRA examinations.[12] If clinically suspected, digital subtraction angiography will be required to confirm or exclude dural arteriovenous fistula.[12,13] For patients with a retrotympanic mass, temporal bone CT should be the initial diagnostic imaging method used to evaluate glomus tympanicum, aberrant internal carotid artery, or jugular bulb abnormality. Carotid angiography or neck CT can be added, based on the results.[6,11]

The purpose of management of vascular pulsatile tinnitus is to correct the causative factors or remove the problematic lesion surgically. General systematic problems should be corrected before surgical treatment.8 In cases of symptomatic high-dehisced jugular bulb, such as cases 1 and 2, repair using pieces of mastoid cortical bone; septal, conchal, or tragal cartilage; and surgical bone wax has been reported[14], and ligation of the ipsilateral IJV showed good results.[6,14] Most cases of high jugular bulb are asymptomatic, but surgical management is required in cases with pulsatile tinnitus, such as venous hum loud enough to disturb sleeping or hearing.[2] Tinnitus produced by an enlarged jugular bulb can be due to turbulent venous flow between the relatively rigid sigmoid sinus and the irregularly enlarged jugular bulb.[15]

Venous hum or essential pulsatile tinnitus, as in case 3, can be successfully treated with ligation of the ipsilateral IJV.[3,8,16] There are some studies contrary to this suggestion. Rouillard et al reported that ligation of the jugular vein usually did not produce long-term successful results and blood shunting with symptoms in the other ear could be occurred.[10] The results of this

procedure have been very inconsistent, so that some reports have indicated recurrence of pulsatile tinnitus after ligation of the IJV and the development of intracranial hypertension.[1] Therefore, the degree of annoyance and daily life impact should be evaluated before surgical management of venous hum or essential pulsatile tinnitus, and it is necessary to spend sufficient time in observing the patient. Cases with no subsequent systemic problems related to pulsatile tinnitus can be indicated for surgery. In all cases in the present study, tinnitus disappeared completely after surgery and the patients were satisfied with the results over the follow-up period.

In conclusion, vascular pulsatile tinnitus could be improved markedly with surgical management of the lesion. However, in essential pulsatile tinnitus, the patient should be evaluated thoroughly and care should be taken in deciding the surgical indication. Considering the side effects of high level ligation of the IJV, it is useful to find the lowest site for venous ligation, confirmed by compression of the IJV with the Doppler sonograph probe.

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