CASE REPORT

Endovascular treatment of jugular bulb diverticula causing debilitating pulsatile tinnitus

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SUMMARY

We describe the case of a patient who presented with debilitating pulsatile tinnitus in association with two jugular bulb diverticula. The diverticula were treated with stenting of the jugular bulb and coil embolization of the diverticula over two procedures. This resulted in successful resolution of symptoms and at 10 months follow-up the patient is asymptomatic. The technique is discussed with regard to similar published cases and surrogate measures of safety taken from the literature pertaining to idiopathic intracranial hypertension.

BACKGROUND

Jugular bulb abnormalities may be identified in association with a number of symptoms including pulsatile tinnitus and vertigo.^{1 2} These symptoms can be debilitating and have a significant impact on quality of life.³ Treatment options range from conservative management to surgery,⁴ but patients may not be able to tolerate symptoms with the former and the latter procedure may be associated with significant morbidity. Dural sinus stenting is increasingly used for treatment of patients with idiopathic intracranial hypertension (IIH) with relatively low morbidity.⁵ We describe the use of a stent-assisted coil approach to treat a patient with two jugular bulb diverticula (JBD) and severe pulsatile tinnitus.

CASE PRESENTATION

A previously healthy 58-year-old woman presented with a 10-month history of debilitating right-sided pulsatile tinnitus that was maximal when recumbent and was depriving the patient of sleep. The patient's quality of life and ability to perform normal activities of daily living were suffering as a result.

INVESTIGATIONS

The patient was investigated with carotid Doppler ultrasound, which was normal, and subsequently underwent a MR scan including high resolution T2 imaging of the cerebello-pontine angle and contrast-enhanced MR venography. This demonstrated two right-sided wide-necked JBD protruding superiorly and laterally (figure 1A). A digital subtraction cerebral angiogram was then undertaken which excluded a dural arteriovenous fistula and confirmed the jugular bulb abnormality (figure 1B).

TREATMENT

The patient was commenced on prophylactic aspirin (100mg daily) 1 week prior to the initial treatment, to remain on this long-term, and was anticoagulated during each procedure with heparin. Via a right

femoral venous approach, a 6F Neuron MAX 088 long sheath was navigated into the right internal jugular vein. A 12×60 mm self-expandable Luminexx stent (Bard, Georgia, USA) was exchanged over an 035 inch guidewire and deployed across the jugular bulb from the sigmoid sinus to the internal jugular vein (figure 2). The superior diverticulum was then catheterized through the indices of the stent using a PX400 microcatheter (Penumbra, California, USA) and the diverticulum was coiled using six detachable Penumbra 400 coils. Several coil loops herniated into the lateral diverticulum and, although it remained only partially coiled, it was hoped that this would be sufficient to alleviate the patient's symptoms. After the procedure the patient's symptoms diminished but did not completely settle and a second procedure to recoil the lateral diverticulum was undertaken 5 months later. On this occasion, angiography confirmed patency of the stent, no alteration in the venous drainage pattern, but complete occlusion of the superior diverticulum and residual filling of the lateral diverticulum. A 6F Envoy MPD guide catheter (Cordis, California, USA) was navigated into the right internal jugular vein and an SL10 microcatheter (Stryker, Michigan, USA) was used to catheterize the lateral diverticulum. A series of DELTAPAQ coils (Codman Neuro, Massachusetts, USA) were deployed to occlude the diverticulum (figure 3).

OUTCOME AND FOLLOW-UP

Following the second procedure the patient was asymptomatic and remains so at 10 months clinical follow-up.

DISCUSSION

JBD have been demonstrated in 1.2–7.9% of CT imaging series.^{6 7} These are probably acquired lesions; the jugular bulb forms after 2 years, increasing in size to plateau in adulthood.⁸ The prevalence of jugular bulb abnormalities increases during the first four decades of life and stabilizes thereafter.⁹ While it is not possible accurately to identify what percentage of diverticula are symptomatic (and it is recognized that many will be asymptomatic⁶), they have been described in association with pulsatile tinnitus and other symptoms such as vertigo and hearing loss in a number of case series.^{1 2 6 10–12} Patients tend to be >50 years of age at presentation.^{10 11}

Pulsatile tinnitus is associated with insomnia and may impact on quality of life.³ One series identified diverticula as a minority cause behind high riding or dehiscent jugular bulbs, internal carotid artery

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Figure 1 (A) Coronal contrast-enhanced magnetic resonance venogram demonstrating right-sided jugular bulb diverticula (white arrowheads). (B) Frontal projection venous phase of a right internal carotid artery injection digital subtraction angiogram confirming the presence of the diverticula at the right jugular bulb.

disease, or dural arteriovenous fistula.¹² Diverticula may cause pulsatile tinnitus through turbulent flow and/or expansion into adjacent structures. Friedmann *et al*¹³ suggested an association between erosion into the vestibular aqueduct and pulsatile tinnitus as well as hearing loss. Bilgen *et al*⁶ classified diverticula as either posterior- or anterior-extending. The posterior-extending diverticula were more often associated with sensorineural hearing loss and tinnitus with vertigo, and the anterior-extending diverticula were more often associated with sensorineural hearing loss and tinnitus without vertigo.

Diagnosis can be made with MR venography, but the optimal imaging tool is probably a combination of CT venography and skull base CT, allowing identification of the relationship to adjacent structures.^{2, 10} The surgical treatment of JBD involves mastoidectomy and impaction of bone wax.⁴ Endovascular treatment offers a relatively minimally invasive approach since the surgical option offers the theoretical risk of disruption of the adjacent facial nerve, middle ear, and dura. Several authors have described using endovascular techniques to treat transverse-sigmoid sinus aneurysms or stenoses and diverticula presenting with pulsatile tinnitus^{14, 15} using coil embolization or stenting. Dural venous sinus stenting is a growing treatment option for selected patients with IIH, but is also used to treat

dural sinus thrombosis or stenoses relating to dural arteriovenous fistula.

Endovascular techniques for treatment of JBD have previously been reported in the context of acute postoperative hemorrhage¹⁶ and for the treatment of vertigo.¹⁷ To our knowledge, this is the first time that stent-assisted coiling has been employed for the treatment of jugular diverticula associated with pulsatile tinnitus. Hitier *et al*¹⁷ described the treatment of three patients with jugular bulb abnormalities and vertigo (including two patients with diverticula) using similar techniques to those described here, with clinical success. These authors employed carotid Wallstents (Stryker) delivered via a femoral approach using an 8F guiding system. In our opinion the choice of stent is heavily dependent on trackability, and stent positioning may necessitate a jugular venous puncture or use of a robust 035 inch wire in order to achieve position.

Critical to offering a treatment such as this are the technical success rate and procedural complication rate. A surrogate situation from which data can be obtained is that of stenting for IIH. In a systematic review of the literature,⁵ 207 patients with IIH were treated. The technical success rate was high at 95%, with three major complications directly related to stent placement equating to a risk of 1.5%: one case of vein perforation

Figure 2 Lateral digital subtraction venogram via right internal jugular vein injection showing stent position.

Figure 3 Frontal digital subtraction venogram via right internal jugular vein injection showing coil embolization of the two diverticula.

leading to subdural hematoma, one case of retroperitoneal hematoma that did not require treatment, and one transient contrast extravasation. There were two reported cases of in-stent thrombosis (a rate of 1.6%), but the follow-up period ranged widely from 2 to 108 months and post-stent imaging was variable with both invasive and non-invasive techniques used. A recent study of long-term patency of mean 22 months using catheter venography demonstrated an in-stent stenosis rate of 17% (4/23 patients), but none were flow-limiting.¹⁸

These results should be extrapolated to the JBD population with caution since IIH is a disease of young women whereas most reports of tinnitus in association with diverticula are in older patients. Could, for example, older patients be more at risk of in-stent stenosis or thrombosis? The latter can be treated with thrombolysis, but the consequences including venous infarction and intracranial hemorrhage (particularly if stenting a dominant sinus) are potentially devastating and need to be weighed against the severity of the presenting symptoms. For prophylaxis of thrombotic complications we preloaded and maintained our patient with aspirin and anticoagulated her during the procedures. However, it is evident that, in the setting of IIH, most operators employ dual antiplatelets (aspirin and clopidogrel) for 3–6 months to allow endothelialization with heparin during treatment followed by aspirin long-term or a short period of anticoagulation followed by single antiplatelet therapy.⁵ We followed this patient with repeat catheter angiography/venography as we were intent on treating the residual diverticulum. The presence of the coils within the petrous temporal bone may obscure vascular detail, as depicted by follow-up CT, and perhaps the most appropriate non-invasive follow-up imaging may be contrast-enhanced MR venography. We planned to follow this patient clinically after the second procedure since the stent showed no evidence of stenosis at 5 months and she was asymptomatic following the second coiling. One could argue that further follow-up imaging is necessary since late in-stent stenosis is a recognized complication.¹⁸

Learning points

- ▶ Jugular bulb diverticula may be seen in association with pulsatile tinnitus.
- ▶ In some cases this can impact on quality of life and be debilitating.
- ▶ Stenting in combination with coil embolization can be used successfully to treat pulsatile tinnitus associated with jugular bulb diverticula in extreme cases.

CONCLUSION

We have demonstrated that pulsatile tinnitus associated with two JBD can be successfully treated using an endovascular approach employing a sigmoid sinus to internal jugular vein stent with coil embolization of the diverticula. The endovascular approach may therefore be a useful option in extreme cases.

Contributors All authors contributed to the manuscript.

Competing interests None.

Patient consent Obtained.

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