Med & Health Jul 2025; 20(2): 805-810 https://doi.org/10.17576/MH.2025.2002.27

CASE REPORT

Unilateral Pulsatile Tinnitus in Young Female Adults: A Report of Two Cases

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Received: 19 June 2024 / Accepted: 01 August 2024

ABSTRAK

Tinnitus berdenyut adalah suatu fenomena bunyi menyerupai degupan jantung yang didengari oleh seseorang meskipun tiada sebarang stimulus yang nyata. Keadaan ini kebiasaannya berkaitan dengan faktor vaskular. Kami menghuraikan dua kes tinnitus berdenyut unilateral yang menarik di mana satu kes disebabkan oleh liku arteri karotid dalaman (ICA) ekstrakranial dan satu lagi berpunca daripada hakisan dinding sinus sigmoid. Kedua-dua pesakit mempamerkan ciri yang sama di mana tekanan jari di kawasan jugulo-karotid ipsilateral melegakan tinnitus mereka. Pengimejan diagnostik, termasuk 'High Resolution Computed Tomography' tulang temporal dan 'Computed Tomography Angiography' kepala dan leher, menjurus ke arah diagnosa yang tepat dalam kedua-dua keadaan. Majoriti kes tinnitus berdenyut mempunyai punca yang boleh dikenal pasti dan modaliti pengimejan memainkan peranan penting dalam pembentukan diagnosa. Pilihan rawatan merangkumi sekadar pemerhatian dan juga intervensi perubatan termasuk pembedahan.
Kata kunci: *Arteri; tinnitus berdenyut; vena*

ABSTRACT

Pulsatile tinnitus (PT) manifests as an auditory sensation synchronised with the heartbeat, occurring without any external auditory stimuli. While its etiology is diverse, vascular factors predominate. We present two intriguing cases of unilateral PT with unremarkable otorhinolaryngological examinations: one attributed to tortuosity of the extracranial internal carotid artery (ICA) and the other to dehiscence of the sigmoid sinus wall. Both patients exhibited a common characteristic whereby compression of the ipsilateral jugulo-carotid region relieved their tinnitus. Diagnostic imaging, including High Resolution Computed Tomography (HRCT) of the temporal bone and Computed Tomography Angiography (CTA) of the head and neck, facilitated accurate diagnosis in both instances. The majority of PT cases have identifiable causes, with imaging modalities playing

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a pivotal role in diagnosis. Treatment options range from observation with reassurance to medical intervention or surgical management.
Keywords: Arterial; pulsatile tinnitus; venous

INTRODUCTION

Tinnitus is the perception of sound in the head or ears in the absence of an auditory stimulus. It is a symptom of an underlying condition, and approximately 14% of the world's population has experienced tinnitus (Jarach et al. 2022). It is generally divided into two types: pulsatile and non-pulsatile tinnitus, with the former being the less common type. Pulsatile tinnitus (PT) comprises about 4% of tinnitus patients. PT is synchronous with the heartbeat and predominantly vascular in origin. Vascular causes can be further subcategorised into arterial or venous origin. The less common non-vascular etiologies include conditions with increased cardiac output such as anemia and thyrotoxicosis. Generally, PT arises as a result of local turbulent blood flow or changes in the density of the temporal bone, which results in increased sound conduction of the normal blood flow in the vessels. Here, we present two cases of unilateral PT in young female adults with characteristic histories, albeit with different vascular etiologies.

CASE REPORT

Case 1

A 23-year-old female with no underlying comorbidities presented with right-sided tinnitus for 3 months. The tinnitus was described as pulsatile and synchronous with her heartbeat. She denied any associated hearing loss, vertigo, disequilibrium, or prior trauma. She characteristically mentioned that the tinnitus disappears with digital compression of the right jugulo-carotid region. There were no complaints of headache, vomiting, or visual symptoms. She was normotensive, and her BMI was 25.3 kg/m^2. Otoscopic examination, along with the rest of the ear, nose & throat (ENT) and neurological examinations, was unremarkable. No neck bruits or objective PT were identified. Blood tests excluded anemia and hyperthyroidism. Pure-tone audiometry (PTA) revealed normal hearing bilaterally. Computed Tomography Angiography (CTA) of the head and neck was performed and revealed tortuosity of the right extracranial internal carotid artery (ICA) at level C2 (Figure 1). The patient was referred to the Neurosurgical team for further intervention, upon which she opted for conservative treatment. Her perception of tinnitus eventually improved following the explanation of her condition and reassurance.

Case 2

A 32-year-old female with no underlying comorbidities presented with daily symptom of intermittent right-sided PT for the past year. Symptom lasted for lasting 5-10 minutes each episode. She denied any associated hearing loss, vertigo, aural fullness, or prior trauma. The tinnitus is more pronounced after exertion and is relieved with head turning to the right. There were no complaints of headache, vomiting, or visual symptoms. Her body mass index (BMI) was 21.5 kg/m^2. Otologic examination was unremarkable with no evidence of middle ear fluid or mass. There was no neck bruit or objective PT on auscultation. Notably, the tinnitus disappeared upon compression of

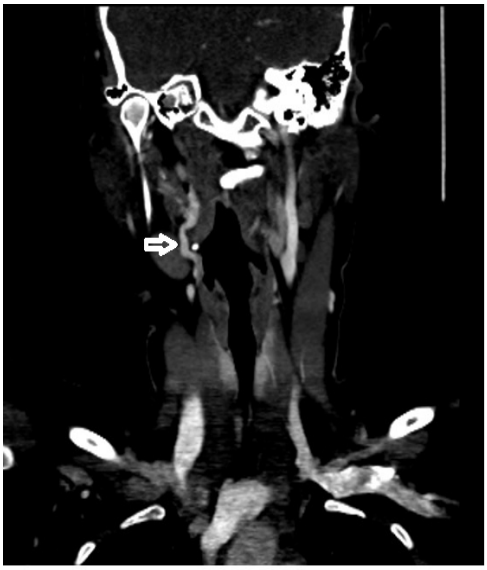

FIGURE 1: CTA showing tortuosity of right extracranial ICA at level C2 (white arrow)

the right jugulo-carotid region. She was also normotensive, not anemic, and euthyroid. Pure-tone audiometry (PTA) revealed normal hearing bilaterally with normal tympanometry. High Resolution Computed Tomography (HRCT) temporal, along with CTA of the head and neck, was performed, which revealed dehiscence of the sigmoid sinus plate (Figure 2). Otherwise, no vascular anomalies were found, and the dural venous sinuses were patent with no evidence of stenosis. The findings were explained to the patient along with the treatment option of right cortical mastoidectomy and sigmoid sinus resurfacing. However, the patient chose a conservative approach in view of the benign pathology and its non-disabling nature currently.

DISCUSSION

Arterial PT mostly occurs due to atherosclerotic carotid disease, which causes turbulent flow as a result of stenosis (Terzi et al. 2015). Other arterial causes include arterio-arterial anastomosis, aberrant carotid artery, and fibromuscular dysplasia. Infrequently, morphological abnormalities of the ICA may lead to PT as well. The Weibel-Fields classification system categorises ICA abnormalities into three types: tortuosity, coiling, and kinking (Weibel et al. 1965). The incidence of ICA tortuosity is 18-34%, which is not uncommon. However, about 80% of cases are asymptomatic and often identified incidentally (Kim et al. 2018). Such incidental discovery may occur in otorhinolaryngology clinics, in which patients may present with lateral pharyngeal wall bulge,

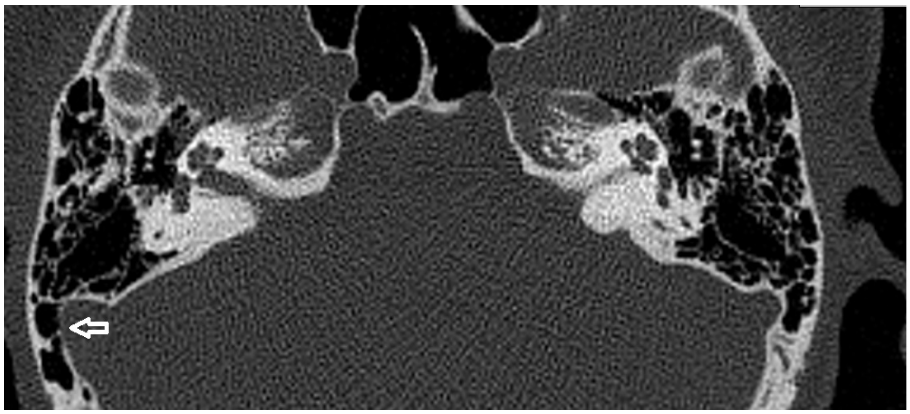

FIGURE 2: HRCT temporal showing right sigmoid sinus wall dehiscence (white arrow)

causing an abnormal sensation in the throat, dysphagia, or cough. There are also cases incidentally discovered during oropharyngeal surgeries such as tonsillectomy, in which a pulsating lateral wall mass was encountered (Gürbüzler et al. 2013).

Pertinently, PT is also a common presenting symptom of ICA tortuosity. The main causes of tortuosity are atherosclerosis, hypertension, connective tissue disease, and congenital deformities. In our patient, it is likely due to a congenital origin. The ICA is derived embryologically from the third branchial arch artery and the distal segment of the dorsal aorta. As the fetus matures and elongates, the heart descends into the thorax and straightens the vessels. An aberration in this process contributes to the redundancy of the ICA and thus, tortuosity of the vessel (Park et al. 2012). It is worth noting that not all patients with tortuous ICA experience PT. Apart from the tortuosity itself, the presence of other associated conditions of hyperdynamic circulation may cause turbulent blood flow and thus, manifesting in PT (Sismanis et al. 2008).

There are several treatment options for extracranial ICA tortuosity causing PT. These include end-to-end anastomosis following resection of the tortuous part, endovascular stenting to straighten the affected part, and external straightening technique. De Ridder et al. (2013) reported the first case of successful external arterial remodeling using Teflon as an external material to straighten the tortuous segment. There is also potential for anxiolytic and beta-blocker as non-surgical interventions. Beta-blockers reduce heart rate and consequently, the arterial flow while anxiolytics provide myorelaxing effect (Albertino et al. 2005). Spontaneous resolutions have also been reported before, which was explained by the possible evolving morphology of ICA resulting in cessation of flow turbulence (Sismanis et al. 2008).

On the other hand, sigmoid sinus wall dehiscence (SSWD) or sigmoid sinus diverticulum (SSD) are the two sigmoid sinus anomalies that can cause PT of venous origin. SSWD refers to a condition where there is bony dehiscence of the sigmoid sinus wall, while SSD involves focal outpouching of the sigmoid sinus into the mastoid air cells through the dehiscence (Wang et al. 2014). The enhanced conduction of blood flow sound through the mastoid air cells may explain the occurrence of PT in SSWD.

SSWD exhibits a female preponderance and is often associated with idiopathic intracranial hypertension (IIH), which also tends to affect females, typically in the obese, middle-aged group. However, in our patient, she had a normal BMI without any symptoms or signs of IIH. The pathogenesis of SSWD remains contentious. Since the majority of cases tend to present at a later age, the likely postulation was altered hemodynamics leading to increased turbulence or velocity, causing heightened impact forces on the sigmoid wall. Eventually, this results in dehiscence of the focal wall of the sigmoid sinus over time. Potential factors contributing to altered hemodynamics include age, dyslipidemia, intracranial hypertension, and dominance of the venous systems (Wang et al. 2014). SSWD predominantly occurs on the right side in tandem with the dominance of right-sided venous drainage in most individuals. Meanwhile, Liu et al. (2015) posited that SSWD may be a congenital condition, as the radiological prevalence of SSWD was similar across different age groups. Contrary to expectations for an acquired condition, their study did not find a higher prevalence among older individuals (Liu et al. 2015). This finding also contradicted the hypothesis suggesting SSWD as an early indication of osteoporosis.

In our patient, no evident factors were found to suggest flow turbulence. However, we are mindful of documented cases of tinnitus following COVID-19 vaccine administration. It's conceivable that the vaccine may induce turbulence and consequently PT, particularly in the presence of congenital SSWD. While this might be a plausible hypothesis, further investigation is warranted to validate a potential association.

Studies have shown that the presence of SSWD may not always cause PT. This could be due to hemodynamic changes not being sufficient or prominent enough to cause tinnitus. Additionally, the loudness of tinnitus may not have sufficiently crossed the patients' threshold of perception. The primary treatment option for SSWD is surgical intervention, specifically sigmoid sinus resurfacing. This procedure is advocated as the preferred and highly effective treatment for SSWD. The mechanisms of resolution are attributed to several factors, including the creation of a sound baffle following reinforcement of the bony wall, which reduces sound transmission to the cochlea, and minimised bone conduction of sound through the mastoid air cells by simple mastoidectomy (Kim et al. 2016). Sigmoid sinus resurfacing has generally been proven to be a very safe and effective procedure, with only minor complications reported, such as retroauricular area collapse, persistent periauricular numbness, and ear fullness (Wang et al. 2017). Rarely, there are anecdotal reports of complications such as cerebral venous thrombosis following sigmoid sinus resurfacing. However, these occurrences could not be attributed solely to the surgery, as the reported patients had other risk factors for thrombosis as well (Yun et al. 2021). Nevertheless, care must be taken to avoid compressing or traumatising the sigmoid sinus. Among the materials used for sigmoid sinus resurfacing are bone cement, bone pate, bone wax, temporalis fascia, Surgicel, and Gelfoam. Although the surgical techniques and choice of materials differ according to individual surgeon preferences, a combination of these materials is often used to perform multilayer resurfacing. The choice of material, in terms of source (autologous vs. artificial) and density (hard vs. soft), was found to have no significant association with the resurfacing outcome (Liu et al. 2019).

CONCLUSION

PT is a bothersome condition that can potentially impair patients' quality of life significantly. However, a thorough history and physical examination may provide useful clues to the possible cause, which can then be elucidated with the guidance of imaging modalities. When PT is synchronous with the heartbeat and the patient reports cessation or diminished tinnitus following compression of the ipsilateral jugulo-carotid region, vascular abnormalities are highly suspected. Otoscopic examination and blood tests can exclude conditions such as middle ear neoplasm, jugular bulb anomalies, anemia, and hyperthyroidism. CTA and HRCT temporal are reliable preliminary imaging modalities to ascertain the etiology, particularly carotid tortuosity and SSWD as in our patients above. Patients with vascular tinnitus mostly have treatable underlying etiologies. Nevertheless, at the very least, awareness of the nature of the conditions, appropriate counseling, and tinnitus retraining therapy itself may benefit tremendously for those patients who are not keen for any interventions.

Authors' Contributions: Kuganathan Ramasamy and Wan Nabila Wan Mansor were major contributors in writing and editing the

manuscript. All authors read and approved the final manuscript.

Funding: The authors have no relevant financial or non-financial interests to disclose.

Ethics approval and consent to participate: Ethical approval for this study was waived and informed and verbal consent to participate were obtained from the patients.

Acknowledgement: Not applicable

Consent for publication: Informed consent and written consent to publish the patients' clinical details and clinical images were obtained from the participants in this study.

Conflict of interest: The authors declare no competing interests.

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