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Pulsatile Tinnitus Associated With a Rare Congenital Vascular Anomaly: A Case Report

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Abstract

Pulsatile tinnitus (PT) is an uncommon form of tinnitus. Unlike non-pulsatile tinnitus, PT is often unilateral and may have a vascular origin, warranting imaging evaluation to identify its underlying cause. Dehiscence of the bony wall of the internal carotid artery (ICA) canal, with protrusion of the artery into the middle ear, is a rare cause of a pulsatile retrotympanic mass and may be the underlying cause of PT. Because it can mimic other middle-ear pathologies, accurate imaging is essential before any surgical intervention is considered. Congenital absence of the external carotid artery (ECA) is a rare anatomical variant. We report the case of a 39-year-old man with progressive bilateral hearing loss and left PT, in whom a pulsatile retrotympanic mass was found on otoscopy. Computed tomography (CT) of the temporal bone, CT angiography, and time-resolved magnetic resonance angiography revealed a dehiscent left ICA canal, together with an incidental congenital absence of the left ECA and stenosis of the proximal C2 segment of the left ICA. This case highlights the role of imaging in the evaluation of PT and the importance of recognizing rare carotid variants and their potential clinical implications.

Categories: Radiology, Otolaryngology
Keywords: computed tomography angiography, congenital absence of external carotid artery, dehiscent internal carotid artery, magnetic resonance angiography, non-bifurcating carotid artery, pulsatile tinnitus

Introduction

Tinnitus is the perception of sound in the absence of an external acoustic stimulus [1]. Pulsatile tinnitus (PT), in which the sound is synchronous with the heartbeat, accounts for less than 10% of all tinnitus cases [2,3]. It is typically unilateral and is generally associated with vascular conditions, often resulting from turbulent blood flow within the internal carotid artery (ICA) [2,3]. These include vascular tumors, atherosclerotic changes, and vascular malformations [2-5]. Among the structural causes, dehiscence of the ICA canal in the middle ear is a rare condition that presents as a pulsatile retrotympanic mass [2-5]. In addition, anomalies of the carotid circulation may be found incidentally on imaging, with congenital absence of the external carotid artery (ECA) being an extremely rare example [6,7].

This report describes a patient with PT caused by a dehiscent ICA canal, in whom imaging also revealed an incidental congenital absence of the left ECA with stenosis of the proximal C2 segment of the left ICA. We discuss the imaging findings and clinical implications of these two entities.

Case Presentation

A 39-year-old man presented to the ear, nose, and throat (ENT) outpatient clinic with progressive bilateral hearing loss (Figure *1*), more pronounced on the left side, accompanied by left PT (synchronous with the pulse) for several years. The tinnitus was not influenced by the Valsalva maneuver or carotid compression, and no bruit was detected on auscultation. His relevant history included frequent exposure to intense noise in his occupational setting (subway worker). Otoscopic examination revealed a pulsatile retrotympanic mass in the left ear, which warranted further investigation, as well as a posterosuperior atrophic flaccid tympanic membrane and a small perforation (Video *1*). A computed tomography (CT) scan of the temporal bone and a cranio-cerebral CT angiography (CTA) were requested for diagnostic evaluation.

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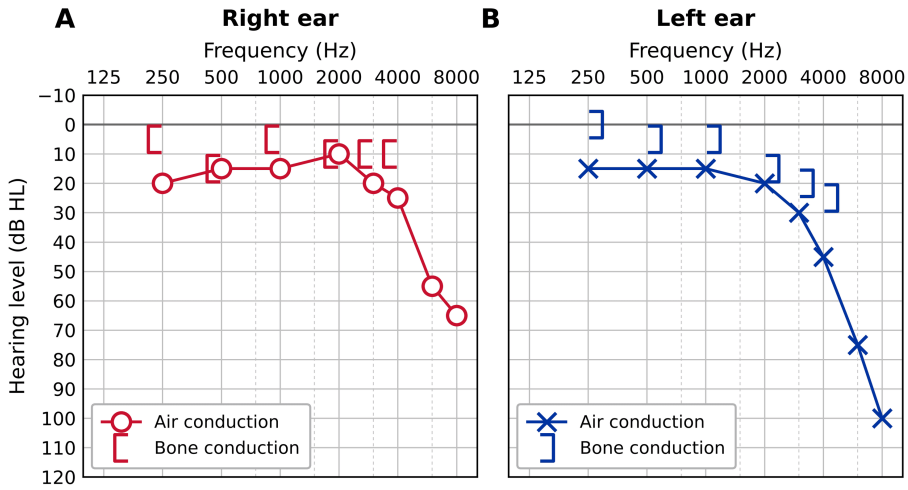

FIGURE 1: Pure-tone audiometry results

Pure-tone audiogram. (A) Right ear; (B) left ear. Hearing thresholds in dB HL from 250 to 8000 Hz. Right ear: hearing is within normal limits from 250 to 4000 Hz, followed by a steeply sloping high-frequency loss, moderate at 6000 Hz (55 dB HL) and 8000 Hz (65 dB HL). Left ear: hearing is within normal limits from 250 to 2000 Hz, followed by a progressively sloping high-frequency loss: mild at 3000 Hz, moderate at 4000 Hz, severe at 6000 Hz and profound at 8000 Hz (100 dB HL). Bone-conduction thresholds largely match air-conduction thresholds, consistent with a predominantly sensorineural hearing loss.

VIDEO 1: Otoendoscopy demonstrating a pulsatile tympanic mass

Otoendoscopy revealing a pulsatile retrotympanic mass in the left ear, as well as a posterosuperior atrophic flaccid tympanic membrane and small perforation visible with the Valsalva maneuver.

View video here: https://vimeo.com/1232327613?share=copy&fl=sv&fe=ci

CT of the temporal bone, performed without intravenous contrast, revealed a hypotympanic dehiscence of the left carotid canal, with arterial protrusion over the floor of the tympanic cavity and bulging of the left tympanic membrane (Figure *2A*), together with a focal reduction of the functional caliber of the left ICA at this level (Figure *2B*). The right ear showed normal bony covering of the ICA.

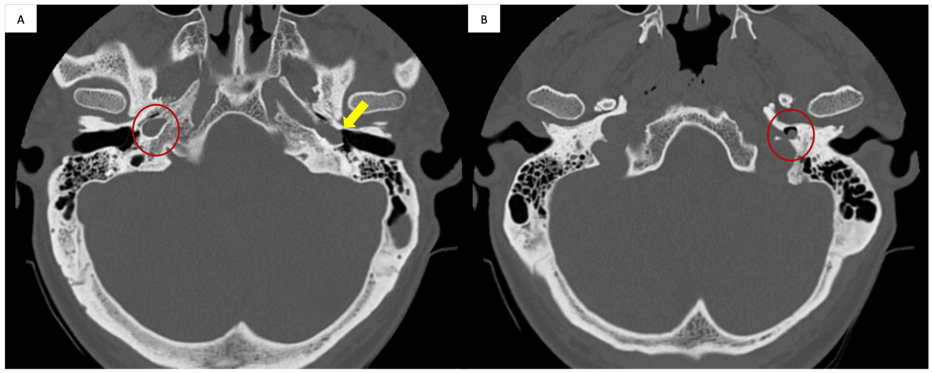

FIGURE 2: Computed tomography of the temporal bone showing dehiscence of the left internal carotid artery canal

Axial computed tomography (CT) images without intravenous contrast administration. (A) Defect in the bony covering of the left internal carotid artery (ICA) (yellow arrow), with arterial protrusion over the floor of the tympanic cavity. The right ear shows the normal bony covering of the ICA (red circle). (B) Same examination, showing focal reduction of the functional caliber of the left ICA at this level (red circle).

Cranio-cerebral CTA confirmed the ICA canal dehiscence at the hypotympanic level, with protrusion into the tympanic cavity. It also showed stenosis of the proximal C2 segment (Bouthillier classification) of the left

ICA, which translates into narrowing of the effective arterial lumen (Figure 3), with a minimum diameter of approximately 3 mm, without evidence of an intra-
or extraluminal obstructive cause, likely representing congenital hypoplasia or narrowing.

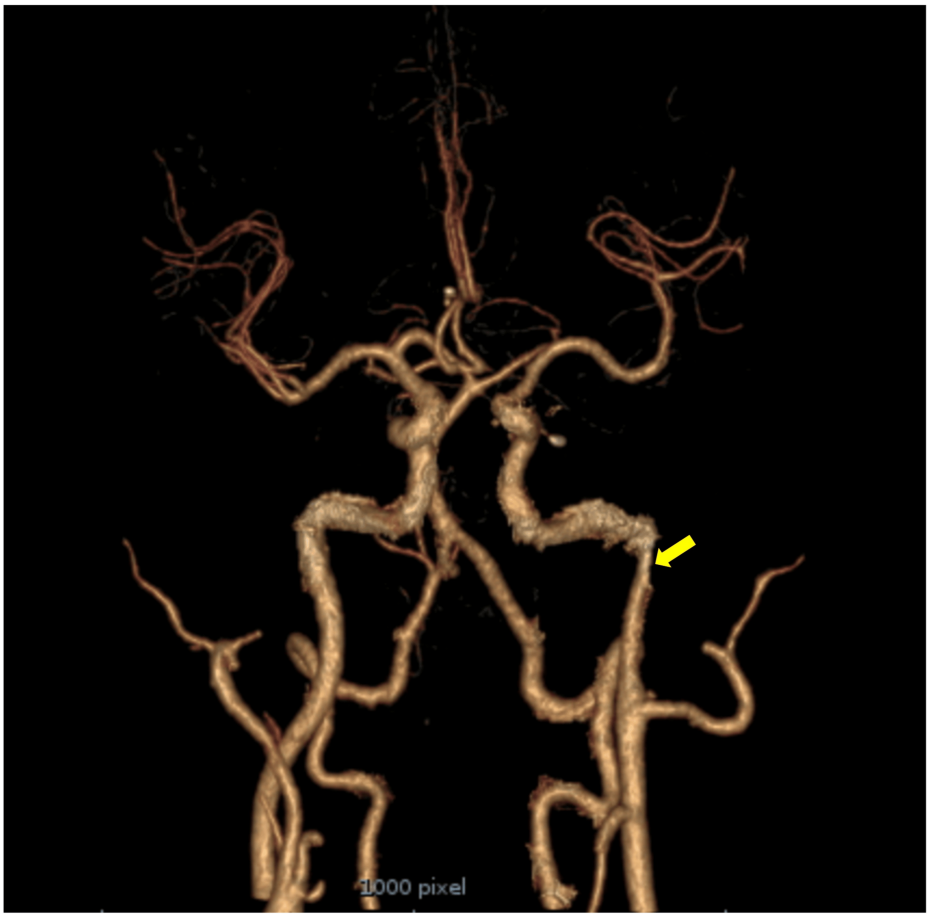

FIGURE 3: Computed tomography angiography (3D reconstruction) of the head and neck

Computed tomography angiography of the head and neck (3D reconstruction) showing normal extracranial vasculature on the right side with normal bifurcation of the common carotid artery and, on the left side, absence of the external carotid artery (ECA), a rare congenital anomaly, with branches that are typically derived from ECA arising from the internal carotid artery (ICA). Also notice stenosis of the proximal C2 segment of the left ICA (yellow arrow), resulting in narrowing of the effective arterial lumen with a minimum diameter of approximately 3 mm.

Incidentally, absence of the left ECA was suspected on CTA, although the level of carotid bifurcation was only partially depicted. Axial contrast-enhanced images showed a normal bifurcation of the right common carotid artery (CCA) and absence of the left ECA, with branches that are typically derived from the ECA arising from the ICA (Figure 4). At a superior level, the left facial artery, a branch of the ECA, was seen originating from the left ICA (Figure 4B), and at a lower level, its descending course encircling the submandibular gland could be followed (Figure 4C). The origin of the internal maxillary artery, also a branch of the ECA, was likewise identified arising from the left ICA (Figure 4D).

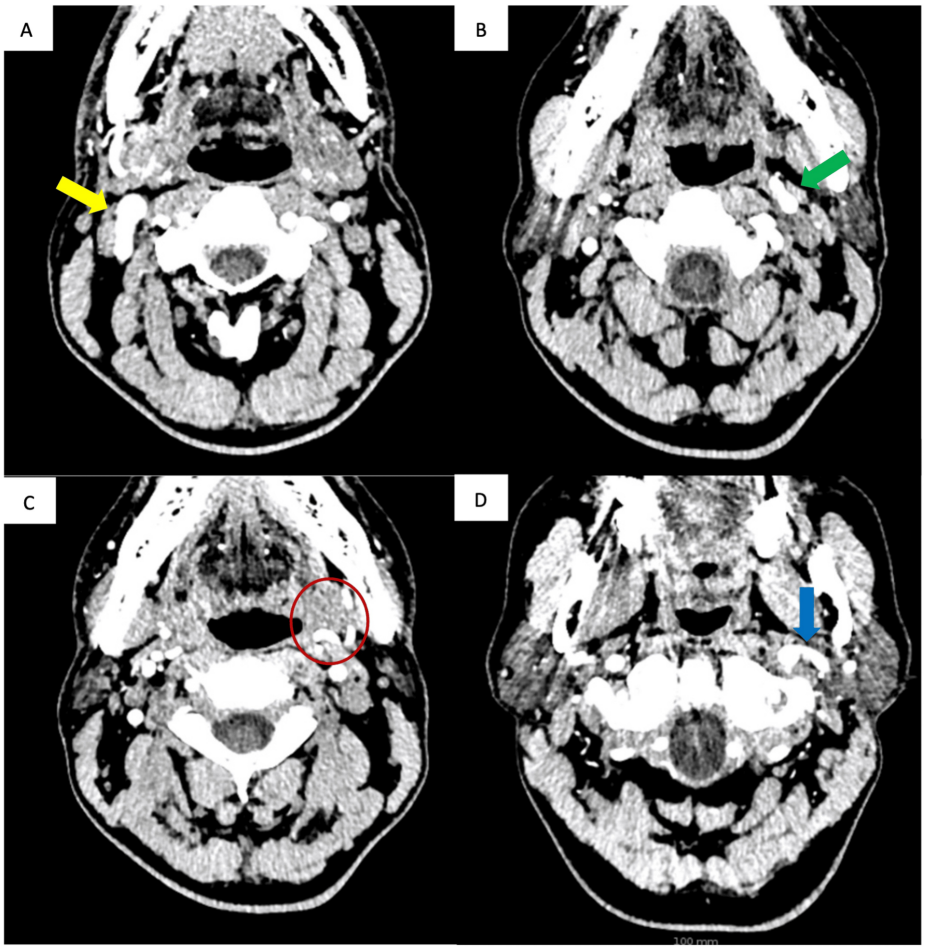

FIGURE 4: Computed tomography angiography showing absence of the left carotid bifurcation

Computed tomography angiography, axial slices, depicting the absence of the left external carotid artery (ECA) and branches that typically originate from the ECA arising from the internal carotid artery (ICA). (A) Cervical sections at the level of the bifurcation of the common carotid artery demonstrating a normal bifurcation on the right side (yellow arrow) and absence of the ECA origin on the left side, at the same level. (B) At a superior level, we can observe the left facial artery (a branch of the ECA) originating from the left ICA (green arrow) in this patient with congenital absence of the left ECA. (C) At a lower level, we can observe the descending course of the left facial artery after originating from the left ICA, encircling the submandibular gland (red circle). (D) At this level, we can observe the origin of the internal maxillary artery (a branch of the ECA) from the left ICA (blue arrow).

Given these findings, magnetic resonance angiography (MRA) with time-resolved sequences (TRICKS) was performed (Figure *5*). It confirmed the absence of the conventional CCA bifurcation on the left side with congenital absence of the ECA, and an anomalous branching pattern of the left ICA, resulting in branches that typically originate from the ECA arising directly from the ICA, in contrast to the normal bifurcation of the right CCA into ICA and ECA. The focal stenosis of the proximal C2 segment of the left ICA was also demonstrated (Figure *5*), with a minimum luminal diameter of approximately 3 mm.

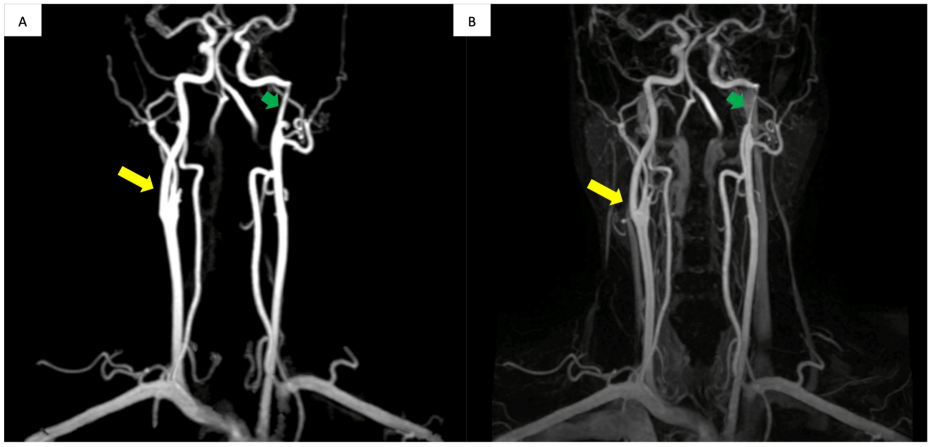

FIGURE 5: Time-resolved magnetic resonance angiography (TRICKS) with coronal 3D reconstructions confirming absence of the left external carotid artery

Coronal 3D reconstructions of time-resolved magnetic resonance angiography (TRICKS) acquired in the arterial phase (A) and in a later arteriovenous phase (B), demonstrating congenital absence of the left external carotid artery (ECA). Note the absence of the conventional carotid bifurcation on the left side, with branches typically arising from the ECA emerging from the left internal carotid artery (ICA). In contrast, the right common carotid artery bifurcation (yellow arrow) shows normal origin of the ICA and ECA. Also noticeable is the stenosis of the proximal C2 segment of the left ICA (green arrow), with a minimum luminal diameter of approximately 3 mm.

Considering the imaging findings, conservative management was chosen, and surgery was not performed, given the associated risks and the low impact of the tinnitus on the patient's quality of life.

Follow-up focusing on cardiovascular risk factors was initiated. CTA was repeated six months after initial diagnosis with no further findings and stability of the described alterations.

Discussion

This case illustrates two distinct findings: a dehiscent ICA canal as the cause of a pulsatile retrotympanic mass and PT, and a rare congenital vascular anomaly identified incidentally, namely unilateral absence of the ECA.

Dehiscence of the ICA canal in the middle ear, characterized by thinning or absence of the bony plate that separates the ICA from the middle ear, is a rare condition and a potential cause of a pulsatile retrotympanic mass resulting in PT [2-5].

Imaging is a useful tool for identifying the underlying cause of PT. CT is particularly valuable in cases of ICA dehiscence, as it allows detailed evaluation of the anatomical relationships between the tympanic cavity and the major vascular structures, including the ICA and the sigmoid bulb of the internal jugular vein [2-4,8]. High-resolution temporal bone CT is regarded as the preferred modality when a retrotympanic mass is seen on otological examination [8]. Magnetic resonance imaging and MRA have limitations in this setting, as they do not depict bony dehiscences of the middle or inner ear [9]. In our patient, CT clearly demonstrated the bony defect in the carotid canal with protrusion of the artery into the tympanic cavity, and CTA confirmed the finding. As this clinical picture can mimic other middle-ear pathologies such as cholesterol granuloma, tympanic or jugulotympanic paraganglioma, dehiscent jugular bulb, meningioma, meningocele or meningoencephalocele, or even superimposed hemorrhage secondary to infection, obtaining accurate imaging prior to any surgical intervention is crucial to prevent potentially life-threatening complications [2,3].

Conservative therapy is the preferred approach and was the treatment chosen for our patient [2]. Conservative management has likewise been adopted in other reported cases in which surgical correction was not considered feasible without compromising hearing or eustachian tube function [9]. This condition is not usually linked to abnormal auditory function [2,5]. However, our patient exhibited bilateral sensorineural high-frequency hearing loss, which we attribute to occupational noise-induced hearing loss given his frequent exposure to intense noise as a subway worker.

Various anomalies affecting the carotid artery circulation have been documented, with congenital absence of the ECA being an extremely rare occurrence [6,7]. In our patient, it was identified as an incidental imaging finding. In this condition, a single carotid vessel is present, lacking the characteristic bifurcation, with

absence of the ECA. Consequently, the branches that would normally originate from the ECA arise directly from the ICA [6,7,10], as observed in our case. Its prevalence has been reported at approximately 0.2% in an MRA series, although with a definition broader than true non-bifurcation [11].

Atherosclerotic stenosis has been described in the few reported cases of absent ECA [6]. The development of atherosclerotic plaque in these vessels seems to involve factors beyond abnormal flow hemodynamics, and carotid endarterectomy has been performed successfully for symptomatic stenosis in this variant [11,12]. This finding emphasizes the importance of early diagnosis to prevent potential occlusions, which could have severe consequences for patients with this anomaly. Although our patient presented with stenosis of the proximal C2 segment of the left ICA, no intra- or extraluminal obstructive cause was identified, suggesting that the narrowing may be congenital in origin, possibly reflecting arterial hypoplasia.

This highlights the importance of follow-up, given the potential for atherosclerotic plaque formation in a vessel with an already reduced calibre. Recognition of the anomaly is also relevant before head and neck surgery and intra-arterial procedure planning [11]. Since the carotid bifurcation was only partially depicted on CTA in our patient, MRA was essential to confirm the diagnosis.

Conclusions

Dehiscence of the ICA canal is a rare cause of PT and pulsatile retrotympanic mass, which can mimic other middle-ear pathologies. Temporal bone CT, complemented by CT angiography, is essential to establish the diagnosis and to avoid potentially dangerous surgical interventions.

Congenital absence of the ECA is an extremely rare anomaly that may be found incidentally. It can be associated with stenosis of the carotid system and should be confirmed with angiographic techniques such as MRA when the carotid bifurcation is not completely depicted. Radiologists and clinicians should be aware of these entities to ensure an accurate diagnosis, as well as appropriate patient management and follow-up.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Carolina Tim-Tim, Jose Pais

Acquisition, analysis, or interpretation of data: Carolina Tim-Tim, Jose Pais

Drafting of the manuscript: Carolina Tim-Tim, Jose Pais

Critical review of the manuscript for important intellectual content: Carolina Tim-Tim, Jose Pais

Supervision: Jose Pais

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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