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Cryptogenic Vascular Dissection as a Potential Cause of Intractable Otalgia

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Abstract

Introduction: To highlight the diagnostic journey and management of otalgia linked to cryptogenic vascular dissection.

Methods: A 68-year-old male experienced intractable right otalgia for 53 years, initially triggered by water ingress into his ear. This otalgia was a persistent pain that can worsen when exposed to cold, coughing, or exercise. It has been treated with multiple painkillers for a long time. and has been treated for presumed otitis and trigeminal neuralgia in multiple hospitals for 53 years, but the cause has not been identified. Dynamic contrast enhanced computed tomography (DCE-CT) revealed a long-segment cryptogenic vascular dissection with characteristic endometrial flaps in the intracranial vertebral artery, which was not detected by standard imaging.

Results: Endovascular stent repair addressed the vascular dissection, resulting in the normalization of ischemic areas and the complete resolution of otalgia. Follow up for about almost 12 months after surgery showed good recovery for the patient, so there is no need to take painkillers anymore.

Conclusions: This case illustrates the diagnostic complexity of secondary otalgia, highlights cryptogenic vascular dissection as a potential etiology, and illustrates the effectiveness of endovascular treatment for cryptogenic vascular dissection-associated otalgia, advocating for advanced imaging techniques in elusive cases.

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Introduction

Primary otalgia, which commonly originates from conditions such as otitis media and otitis externa [1,2], is often readily diagnosable for its characteristic presentation and well-defined diagnostic criteria. However, secondary otalgia, which can arise from a broader spectrum of etiologies-including neuralgias, nervus intermedius neuralgia [3], Bell's palsy, Ramsay Hunt syndrome, carotidynia, dental and temporomandibular joint disorders [4], pharyngitis, myofascial pain, and cervical spine arthritis [1]-introduces a layer of complexity that complicates diagnosis. This complexity is largely due to the intricate innervation of the auricle and the diverse origins of referred pain. In this case presentation, we explore a rare etiology of intractable otalgia: cryptogenic vascular dissection (CVD) [5]. To our knowledge, this is the inaugural report detailing the application of endovascular therapy to resolve CVD as a novel treatment approach for intractable otalgia.

Materials and Methods

Case presentation

A 68-year-old male patient presented with a 53-year history of chronic otalgia that originated from water ingress into his right ear during a swimming incident. The pain, which fluctuated from mild to severe, exhibited a pattern of persistence and exacerbation, particularly during colds, coughing, or physical exertion. Over the course of 53 years, the patient has been managed with a variety of analgesics. Despite multiple hospital visits and treatments for suspected otitis and trigeminal neuralgia, the underlying cause remained elusive, and no abnormalities were detected upon examination of the ears, neck, and brain.

Thirteen months prior to the current presentation, the patient underwent stent implantation to address severe stenosis at the origin of the left extracranial vertebral artery, which was causing dizziness, followed by a regimen of antiplatelet drugs. One year ago, he experienced right-sided

hemiplegia due to a cerebral infarction in the left basal ganglia. Throughout the 53-year duration of his condition, continuous analgesic therapy was maintained.

Almost six months ago, the patient reported a persistent, dull otalgia on the right side, which subsequently intensified. This exacerbation was unresponsive to treatment with antibiotics, carbamazepine, a range of analgesics, or tramadol injections. The intractable nature of the pain and the lack of response to conventional therapies underscore the complexity of the case and the need for a comprehensive, multidisciplinary approach to diagnosis and management (Table 1).

Test method

Preoperatively (Figure 1A-D), despite the presence of diffuse hypoperfusion in the posterior circulation (Figure 1C), initial physical examinations and diagnostic imaging modalities, including conventional MRI, computed tomography angiography (CTA, Figure 1A), and digital subtraction angiography (DSA, Figure 1D), failed to disclose any aberrant vascular morphology or additional lesions. However, dynamic contrast-enhanced computed tomography (DCE-CT) revealed the CVD with the telltale intimal flaps characteristic of the dissection within the right vertebral artery (Figure 1B). This elusive ischemic dissection, referred to as CVD [5], was identified as the culprit behind the patient's worsening pain. Following adequate preparation with dual antiplatelet therapy, the right-sided CVD was addressed with stent implantation and reconstruction under general anesthesia, with no intraoperative neurologic complications encountered.

Results and Follow-Up

After stent repair (Figure 1E-I), the postoperative CT perfusion follow-up revealed a normalization of the previously ischemic regions in the right cerebellum and brainstem, as indicated by the shift from red to blue in Figure 1, which coincided with the complete resolution of right otalgia observed both immediately postoperatively and throughout the nearly 6-month follow-up period. Notably, the left-sided hypoperfusion was attributed to a separate instance of CVD, as depicted in (Figure 1H).

Discussion

This case report illustrates the experience of a 68-year-old male patient suffering from the continuous right otalgia over 53 years, presenting a diagnostic challenge resolved only through the identification of CVD (Table 1, Figure 1) [5]. Hence, a comprehensive analysis of the nerves and vertebrobasilar arteries implicated in otalgia, including vertebral artery, the trigeminal, glossopharyngeal, nervus intermedius (geniculate), auriculotemporal, lesser occipital, greater auricular, and vagal nerves, is crucial for developing diagnostic and therapeutic strategies [6].

For decades, the patient had been treated for presumed diseases such as otitis media and trigeminal neuralgia, utilizing a range of analgesics, carbamazepine, and antiplatelet medications without achieving otalgia relief. This case underscores the critical need for etiologic diagnosis, as treatments targeted at assumed conditions proved ineffective without identifying the underlying cause. Traditional examination or imaging modalities, such as DSA and MRI, showed normal vertebral artery morphology, failing to account for the observed cerebellar and brainstem hypoperfusion (Figure 1C). DCE-CT, however, elucidated the long-segment vascular dissection

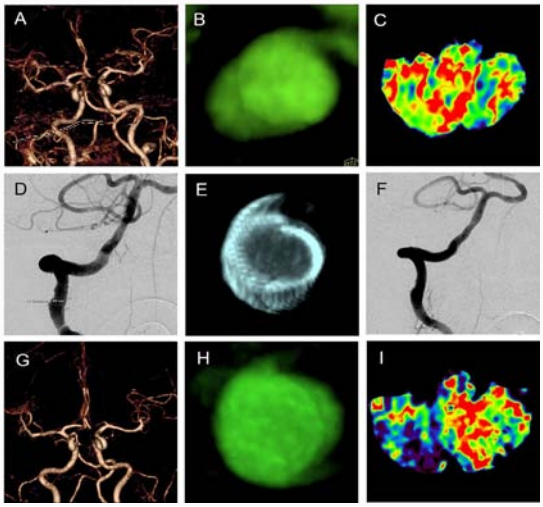

Figure 1: Perioperative evaluation and endovascular treatment of cryptogenic vasculara dissection (cvd)**.** Cta reveals normal morphology and almost similar diameters of the intracranial segments of the bilateral vertebral arteries (Figure A), (Figure B) shows the intimal flap of the dissection, (Figure C) demonstrates diffuse hypoperfusion in the posterior circulation, with a more severe impact on the right side; A stent reconstruction treatment was provided for the CVD in the V4 segment of the right vertebral artery (Figure D-G), Postoperatively, the normal morphology of the right vertebral artery and the hypoperfusion in the right posterior circulation returned to normal (Figure C *vs.* Figure I), while the hypoperfusion on the left side is caused by the dissection in the left vertebral artery (Figure H).

Table 1: Years, Y; Months, M; No relief, NR; Gradually worsening, GW *Worsening otalgia for colds, coughs, or exercise& Stenting for severe stenosis for dizziness at the opening of the left extracranial vertebral artery*& Cerebral infarction in the left basal ganglia.

Patient symptoms, onset time, and changes before and after intervention						
Symptoms	Onset time	Before intervention	After intervention			
			Worsened	No change	Partial improvement	Completely resolved
Persistent otalgia	53 Y	NR				+
Worsening otalgia*	53 Y	NR				+
Dizziness&	13M	NR			+	
Cerebral infarction (L)	1 Y	NR			+	
Otalgia recurred (R)	6M	GW				+

within the intracranial segments of the bilateral vertebral arteries (Figure 1B and 1H). To some degree, the symptoms of dissection mainly depend on its size and the site of ischemia. Therefore, the symptoms of different locations may be diverse. In this case, the right dissection-related to otalgia was more serious (Figure 1B and 1C), which was well demonstrated by the complete pain relief lasting for approximately 12 months follow-up after endovascular treatment (Figure 1E-1G) [7,8].

The successful endovascular treatment of this case illuminates CVD as a potential etiology for intractable otalgia, suggesting a pathogenic mechanism involving ischemic and hypoxic functional disorders of local brain tissue and nerves. This study demonstrates the importance of considering vascular causes in intractable otalgia and the effectiveness of targeted endovascular treatment. Future research should focus on elucidating the role of CVD, particularly in intractable cases lacking clear neurovascular conflict on imaging.

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Author Contributions

Guarantors of integrity of entire study: Jing Pan, Kai-Jun Zhao and Jian-Min Liu.

Study concepts/study design: all authors.

Data acquisition or data analysis/interpretation: all authors.

Manuscript drafting or manuscript revision for important intellectual content: all authors.

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Significance

1. Cryptogenic vascular dissection is one of the rare causes of secondary otalgia.

2. Endovascular treatment of cryptogenic vascular dissection is an effective method for intractable otalgia.

9. Supplementary Data (Video): Changes in Patient's Condition Before and After Treatment (Video).

Before treatment: Intractable right otalgia for 53 years.

1-hour post-treatment: Absence of right otalgia.

8 hours post-treatment: Continued absence of right otalgia, patient begins to smile.

Before discharge: Complete resolution of right otalgia.

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