

[ CASE REPORT ]

## Ipsiversive Ocular Torsion, Skew Deviation, and Hearing Loss as Initial Signs of Anterior Inferior Cerebellar Artery Infarction

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### Abstract:

A 67-year-old man with hypertension and type 2 diabetes mellitus was admitted to our hospital because of left hearing loss and vertical diplopia. A neurological examination showed ocular torsion, skew deviation, and sensorineural hearing loss in the left ear. Brainstem and cerebellar neurological signs were not observed. Left middle cerebellar peduncle infarction was evident on magnetic resonance imaging. He was treated with antiplatelet, however, the infarct progressed after this administration. Ocular tilt reaction (OTR) involves the triad of ocular torsion, skew deviation, and head tilt. Ipsiversive OTR components associated with hearing loss can be early diagnostic signs of anterior inferior cerebellar artery infarction.

**Key words:** ocular tilt reaction, ocular torsion, skew deviation, head tilt, middle cerebellar peduncle, inner ear

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### Introduction

Anterior inferior cerebellar artery (AICA) infarction may mimic peripheral audiovestibular disease, as hearing loss or vertigo is a predominant symptom (1). Hearing loss associated with AICA infarction is observed in 27-92% in the acute phase (1-3). AICA infarction is clinically diagnosed in combination with peripheral audiovestibular and brainstem or cerebellar neurological signs. Patients with AICA infarction sometimes show ocular tilt reaction (OTR), which involves the triad of ocular torsion, skew deviation, and head tilt (4-8). OTR results from the unilateral disorder of the inner ear (9), vestibular pathway in the brainstem (10, 11), or the cerebellum (12-15). Such unilateral lesion causes an imbalance in the utricle related vestibulo-ocular reflex (VOR) in the roll plane, which results in ipsiversive or contraversive OTR. OTR accompanied by audiovestibular dysfunction can be important symptoms for diagnosing AICA infarction. However, OTR sometimes spontaneously improves and is occasionally overlooked. Therefore, the clinical significance

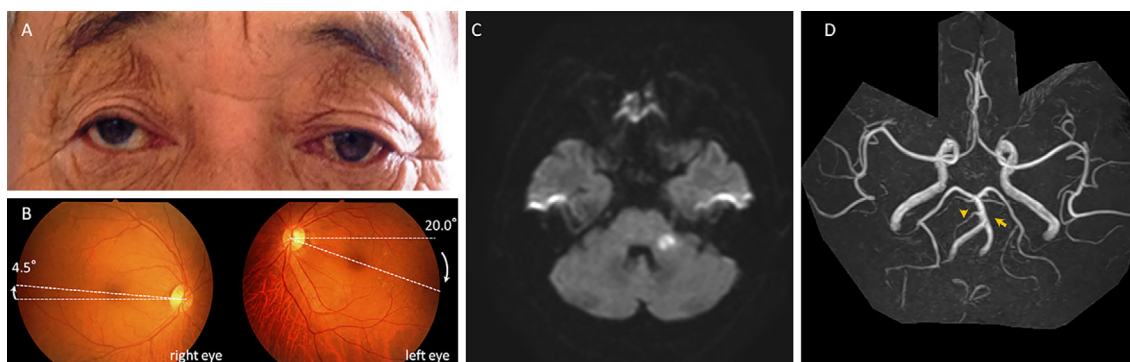
of OTR in AICA infarction has not yet been well discussed.

We herein report a case of ipsiversive partial OTR and sensorineural hearing loss due to AICA infarction and review the literature.

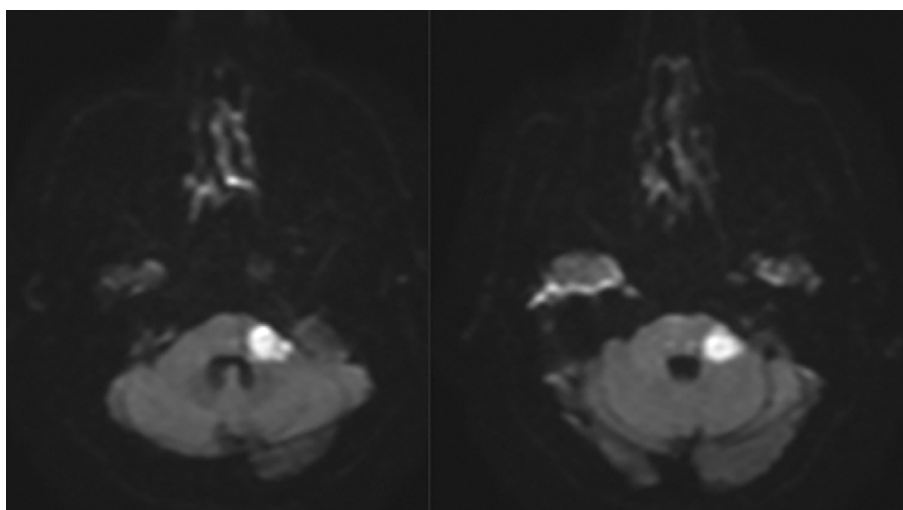
### Case Report

A 67-year-old man with hypertension and type 2 diabetes mellitus visited an otorhinolaryngologist due to waking up with hearing loss in the left ear and vertical diplopia. On admission to our hospital 3 days later, he was alert with normal vital signs except for a high blood pressure of 162/86 mmHg. Neurological examinations revealed gaze-evoked horizontal nystagmus to the right side and skew deviation with right eye hypertropia (Fig. 1A). The pupils were isocoric and direct light reflexes were prompt. Head tilt was not observed. He complained of dizziness, but was able to walk straight. Cerebellar dysarthria, ataxia, and sensory disturbance were not observed. Fundus photography revealed binocular torsion (Fig. 1B). Sensorineural hearing loss in the

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**Figure 1.** Ophthalmological and brain magnetic resonance imaging (MRI) findings. A) Hypertropia of the right eye in the mid-position. B) Pathological evidences of  $4.5^{\circ}$  intorsion in the right eye and  $20.0^{\circ}$  extorsion in the left eye in the fundus photograph. C) An acute middle cerebellar peduncle infarction was evident on diffusion-weighted MRI. D) The left anterior inferior cerebellar artery (AICA) was not visualized (arrow), whereas the right AICA could be seen (arrow head); no stenosis or occlusion were observed in the main cerebral arteries.



**Figure 2.** MRI on day 4. The extent of infarction was evident.

left ear was observed on pure tone audiometry. Hyperglycemia (189 mg/dL) and elevated acetylated hemoglobin (7.5%) were observed. Long-term electrocardiogram monitoring and transthoracic echocardiogram showed no abnormalities. A high density lesion was observed in the left middle cerebellar peduncle (MCP) on diffusion-weighted imaging (Fig. 1C) on day 1. The left AICA was not visualized on magnetic resonance (MR) angiography (Fig. 1D). Given these findings, he was diagnosed with having had an acute ischemic stroke due to the left AICA occlusion.

Oral aspirin and intravenous ozagrel sodium were immediately administered. On day 4, however, slurred dysarthria, facial dysesthesia on the left side, left limb ataxia, and an ataxic gait were obvious. The extent of the infarct was evident based on a comparison with the initial magnetic resonance imaging (MRI) findings (Fig. 2). Clopidogrel was added to aspirin and ozagrel sodium to prevent any further progression of the infarction. The skew deviation disappeared within a week. However, his dizziness and gait insta-

bility showed no improvement. One month later, he was able to gait independently and was discharged home. His sensorineural hearing loss remains as a sequela.

## Discussion

The present patient showed ipsiversive ocular torsion, skew deviation and sensorineural hearing loss as the initial neurological signs of AICA infarction. We detected neurological progression with a stepwise expansion of the ischemic lesion.

OTR associated with AICA infarction is a rare condition, with only 10 cases proven on MRI identified thus far (4-8). All cases have shown at least one of the components of OTR-sensorineural hearing loss. The direction of the OTR components was ipsiversive in all cases. The most commonly affected sites on brain MRI were the MCP and lateral pons. Ipsiversive ocular torsion and skew deviation were observed in 9 of 10 patients (90%), head tilt was noted in 5 of

10 patients (50%), complete OTR was seen in 4 of 10 patients (40%). As per previous studies OTR components in the brainstems were found with the following frequencies: ocular torsion (83%), skew deviation (31%), and complete OTR (20%) (10, 11). The OTR components associated with AICA infarction might be frequently observed, although in fewer cases than cases who showed OTR components in the brainstem in the previous studies (10, 11).

Sensorineural hearing loss in patients with AICA infarction reflects inner ear ischemia because the inner ear is supplied by the labyrinthine artery, a branch of the AICA (1). Partial ischemia of the AICA can easily cause inner ear ischemia because the labyrinthine artery is an end artery with minimal collaterals from other major arterial branches. In the present case, vertical diplopia and hearing loss were the initial chief complaints. Therefore, we speculate that hypoperfusion of the AICA initially caused labyrinthine infarction, which caused ipsiversive signs of OTR. A previous study also suggested that the inner ear plays a role in the mechanisms of ipsiversive OTR in patients with AICA infarction (7). Most of the unilateral cerebellar lesions present with contralateral OTR signs (15). The mechanism underlying contraversive OTR is to involve the interruption of the inhibitory projection from the lesioned cerebellum to otolith related neurons in the ipsilesional vestibular nucleus (12-15). However, this mechanism cannot explain ipsiversive OTR in patients with AICA infarction. We should therefore predict more expansive and progressive ischemia than MRI findings may suggest in such patients, although current radiological modalities cannot reveal inner ear ischemia.

AICA infarction is often associated with atherosclerosis of the basilar artery and neurological progressions sometimes are observed than initial neurological findings (2). The mechanisms of most frequently found to underlie AICA infarction are the thrombotic narrowing of the AICA itself or the spread of basilar artery plaque into the AICA origin (16). In the present case, the left AICA was not visualized on MRA, suggesting that atherosclerotic narrowing or plaque at the AICA origin caused hypoperfusion of the AICA, which resulted in stepwise progression.

Patients with ipsiversive OTR accompanied by audiovestibular symptoms, even when no brainstem or cerebellar signs are noted, should undergo MRI. Such patients are at risk of progression of AICA infarction, so an immediate diagnosis and appropriate treatment are required to prevent a further progression of the infarction.

The authors obtained a statement of informed consent to publish a photograph of the patient.

**The authors state that they have no Conflict of Interest (COI).**

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