

# Unilateral sudden sensorineural hearing loss with vertigo as a first symptom of bilateral internal auditory canal metastases: a case report

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## Abstract

There have been relatively few reports of bilateral internal auditory canal metastases of asymptomatic primary pulmonary adenocarcinoma presenting as unilateral sudden sensorineural hearing loss and vertigo. We report a case of a 60-year-old male patient who complained of sudden hearing loss in the right ear and vertigo. Upon a physical examination, no definite neurological signs or nystagmus were observed. Pure-tone audiometry showed deafness in the right ear at all frequencies and high-frequency sensorineural hearing loss in the left ear. The video head impulse test suggested bilateral vestibulopathy. Magnetic resonance imaging of the brain (with gadolinium contrast) revealed bilateral internal auditory canal enhancement and a variable-sized nodular and peripheral-enhancing lesion in the cerebrum and the right cerebellum. A computed tomographic and bronchoscopic biopsy identified asymptomatic primary pulmonary adenocarcinoma in the left upper lobe of the lungs. This is a rare report of bilateral internal auditory canal metastases in an asymptomatic patient with primary pulmonary adenocarcinoma who initially presented with symptoms of unilateral sudden sensorineural hearing loss with vertigo.

## Keywords

Sudden sensorineural hearing loss, vertigo, internal auditory canal, brain metastasis, pulmonary adenocarcinoma, lesion, pure-tone audiogram

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## Introduction

Sudden sensorineural hearing loss (SSNHL) is defined as a hearing loss of at least 30 dB at three sequential frequencies, as measured in a standard pure-tone audiogram, occurring over 72 hours.<sup>1</sup> The incidence of SSNHL is between 5 and 30 cases/100,000 people per year.<sup>2,3</sup>

The prevalence of accompanying vertigo or balance disorder with SSNHL has been reported as 31%.<sup>3</sup> The possible etiologies of SSNHL with vertigo are diverse. These etiologies include Ménière's disease, anterior inferior cerebellar artery stroke, herpes zoster oticus, labyrinthitis, labyrinthine infarct, perilymphatic fistula, SSNHL with benign paroxysmal positional vertigo, and any tumorous lesion in the internal auditory canal (IAC).<sup>4-7</sup> A main diagnostic test for SSNHL with vertigo is magnetic resonance imaging (MRI) with a gadolinium contrast for evaluating the IAC space.

One identifiable etiology of SSNHL is any neoplasm in the IAC. The incidence of IAC neoplasms was reported as 2% in a meta-analysis of 23 studies.<sup>8</sup> Neoplasms in the IAC compromise the cochlear nerve and can cause progressive or sudden hearing loss. The most common IAC neoplasm is vestibular schwannoma, and a few cases are diagnosed as metastatic neoplasm. Half of these metastatic tumors are diagnosed as bilateral metastases.<sup>9</sup>

We report a case of asymptomatic primary pulmonary adenocarcinoma with bilateral IAC metastases presenting as unilateral sudden sensorineural hearing loss and vertigo. To the best of our knowledge, this is the first report of bilateral IAC metastases in a patient with asymptomatic primary pulmonary adenocarcinoma who first presented with symptoms of unilateral SSNHL with vertigo.

## Case presentation

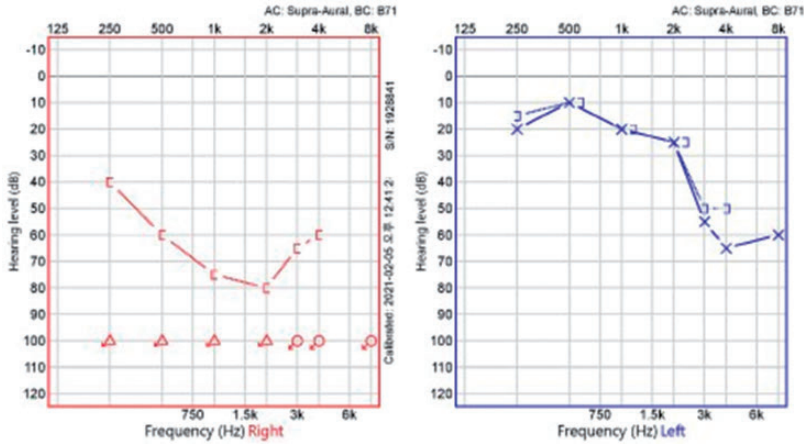
The manuscript was prepared and revised according to the CARE Checklist (2016), and the reporting of this study conforms to the CARE guidelines.<sup>10</sup>

A 60-year-old male patient visited our outpatient clinic for treatment of unilateral SSNHL accompanied by vertigo. He had first presented with SSNHL of the right ear 2 months previously and visited another ear, nose, and throat clinic. He had been prescribed high-dose prednisolone and treated for 2 weeks, but his hearing did not fully recover and he visited our clinic.

The patient had a previous medical history of type 2 diabetes mellitus and hypertension. Seven years before the examination, he suffered from myocardial infarction and had coronary stents inserted. He was medicated for diabetes mellitus and hypertension, but this was poorly controlled because of poor drug compliance. He had a history of 10 pack-years of smoking since the age of 20 years. He drank alcohol eight times a month every month for 40 years. There was neither a history of a previous diagnosis of malignancy nor clinical evidence of a current malignant tumor.

The patient reported unilateral hearing loss of the right ear, with positional vertigo and intermittent headaches. Otoscopic findings were normal. No other neurological symptoms were observed.

An audiogram showed deafness of the right ear at all frequencies and high-frequency sensorineural hearing loss of the left ear (Figure 1). Under videonystagmography, there was no definite nystagmus at rest or after a head-shake test. The video head impulse test was performed and showed decreased gain and covert saccade in the right posterior canal test, and decreased gain in the left posterior canal



**Figure 1.** Pure-tone audiometry shows deafness in the right ear (red) and high-frequency sensorineural hearing loss in the left ear (blue).

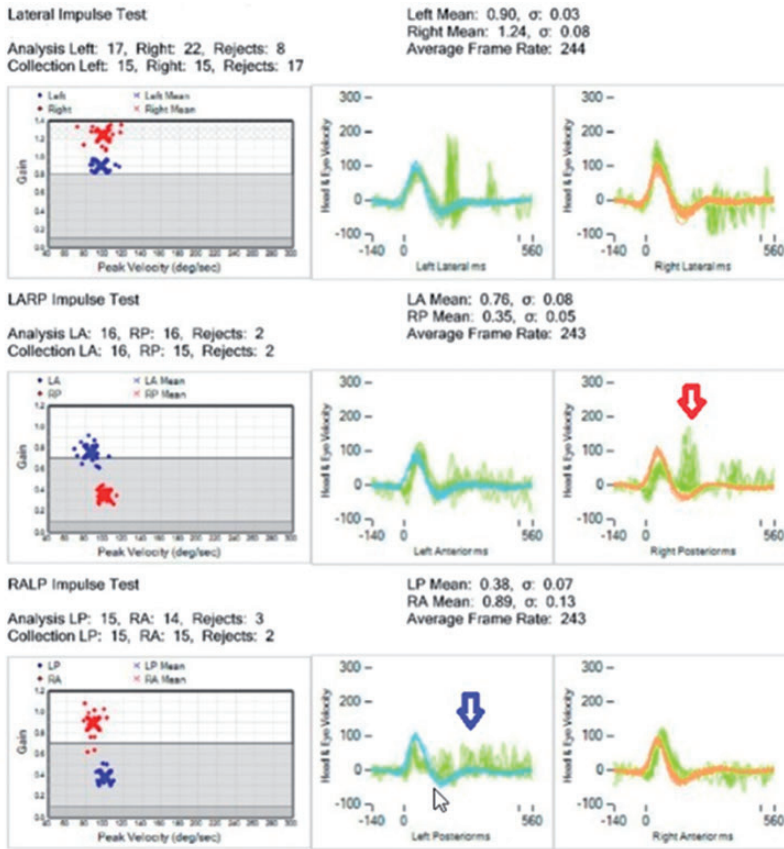
test. An overt saccade was observed on the left lateral canal test (Figure 2).

Gadolinium contrast-enhanced MRI of the head revealed bilateral IAC enhancement, with an 11 × 6-mm lesion on the right and a 9 × 6-mm lesion with cystic change on the left (Figure 3). Additionally, multiple, variable-sized, nodular and peripheral-enhancing lesions in the cerebrum and the right cerebellum were observed.

Laboratory tests for cancer markers showed elevated carcinoembryonic antigen (172.0 ng/mL; normal range: 0–3.8 ng/mL) and cytokeratin 19 fragment (4.7 ng/mL; normal range: 0–3.3 ng/mL) concentrations. Esophagogastroduodenoscopy and colonoscopy showed no abnormal findings. A positron emission tomography-computed tomographic scan showed multiple instances of lymph node uptake in the mediastinum and left pulmonary hilum, and a suspicious primary tumor on the left upper lobe (1.8-cm lobulated nodule). Adenocarcinoma was diagnosed on the basis of a fluoroscopy-guided percutaneous needle biopsy of the left upper lobe nodule.

Anaplastic lymphoma kinase and programmed death-ligand 1 mutations were negative and epidermal growth factor receptor mutation (exons 18 and 20) was positive. On the basis of the biopsy findings of the left upper lobe nodule and the imaging findings from a gadolinium contrast-enhanced MRI of the head, the patient was diagnosed with primary pulmonary adenocarcinoma (epidermal growth factor receptor-positive) with bilateral IAC metastases and brain metastases.

We initiated treatment after we obtained the patient’s consent to treat with oral administration of afatinib and whole-brain radiation therapy to control the brain metastases. The patient reported a decreased incidence of headache and vertigo following the administration of afatinib. No adverse or unanticipated events of any type were reported during the first 2 months of afatinib treatment. In a 2-month follow-up after the brain MRI, multiple, variable-sized, nodular and peripheral-enhancing lesions in the cerebrum and the right cerebellum were markedly decreased.



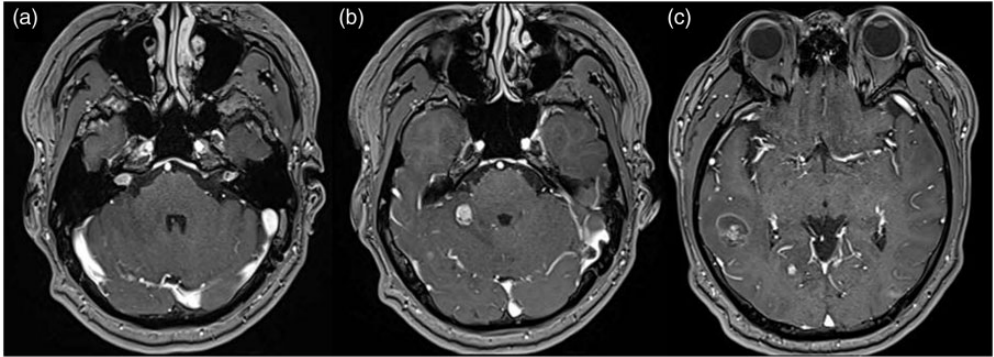
**Figure 2.** The video head impulse test shows decreased gain (vestibulo-ocular reflex gain: 0.35) and covert saccade in the right posterior canal test (red arrow), and decreased gain (vestibulo-ocular reflex gain: 0.38) and overt saccade in the left posterior canal test (blue arrow).

The enhancement of both IAC lesions was attenuated, and the size of the bilateral IAC lesions was also decreased, with an  $8 \times 6$ -mm lesion on the right and a  $7 \times 5$ -mm lesion on the left.

## Discussion

The prevalence of vertigo or imbalance has been reported to be approximately 30% in subjects with SSNHL.<sup>3</sup> In one study, researchers analyzed the audio-vestibular test results of 27 subjects who presented

with acute vertigo and SSNHL.<sup>4</sup> They found that 23 of the 27 subjects were diagnosed with probable labyrinthitis/labyrinthine infarction. Perilymphatic fistula is also a possible cause of acute vertigo with SSNHL in which the characteristic presentation in patients is descending-type hearing loss (either sudden or progressive) accompanied by or following dizziness. Dizziness in perilymphatic fistula is characterized by exacerbation as a result of a positional change; nystagmus can be observed in positional tests.<sup>5</sup> Abnormal IAC lesions are one



**Figure 3.** Magnetic resonance imaging of the brain. (a) Bilateral internal auditory canal enhancement, with an  $11 \times 6$ -mm lesion on the right and a  $9 \times 6$ -mm lesion with cystic change on the left. (b) and (c) Variable-sized nodular and peripheral-enhancing lesions in the cerebrum and the right cerebellum.

of several differential diagnoses of these etiologies. Therefore, MRI should be considered with gadolinium contrast to detect any IAC lesions in patients presenting with acute vertigo along with SSNHL. Our patient presented with unilateral sensorineural hearing loss with vertigo. The video head impulse test suggested bilateral vestibulopathy. On the basis of these results, we performed an MRI with gadolinium contrast and detected bilateral IAC tumor lesions.

An identifiable cause of SSNHL is a neoplasm in the IAC or the cerebellopontine angle. The most common tumor in these sites is vestibular schwannoma. Among the non-schwannomatous lesions, meningiomas, arachnoid cysts, metastases, carcinomatous meningitis, hamartomas, lipomas, neuritis/arachnoiditis, cavernomas, hemangiomas, hemangioblastomas, jugular bulb diverticulum, and traumatic neuromas may be found. Among these various etiologies, metastatic carcinoma accounts for only 0.3% to 0.7% of all lesions in these spaces.<sup>11</sup>

The current case showed bilateral IAC metastatic carcinoma with primary lung adenocarcinoma. A systematic review of the literature showed that bilateral IAC

metastatic carcinoma accounted for 52.9% of the 102 reported cases of IAC metastatic carcinoma.<sup>9</sup> The prevalence of the common primary tumor sites for IAC metastases was as follows: the lungs was 21.2%, skin was 18.6%, and breasts were 16.7%, and lung and skin cancers had the highest rates of bilateral metastasis.<sup>9</sup> The malignant melanoma (69.6%) and adenocarcinoma (57.5%) histological subtypes showed the highest rate of bilaterality.<sup>9</sup>

Diagnosing these IAC tumors is challenging because their radiological findings are similar to those of schwannoma. Histological confirmation of any diagnosis is even more challenging because of the difficulty in obtaining tumor tissue from the IAC. Metastatic tumors usually show rapid growth and patients who present with rapidly progressing symptoms tend to have a known history of cancer malignancy.<sup>12</sup> A main finding of the differential diagnosis between schwannoma and metastases is the presence of adjacent extranodular extensions in metastases, while schwannoma typically shows contrast enhancement within the mass itself.<sup>13,14</sup> Computed tomographic evaluation of the temporal bone has a limited benefit for differential diagnosis.<sup>15</sup>



Treatment of metastatic IAC lesions depends on the primary origin of the tumor. Recently, several targeted therapeutic agents for malignant tumors have been developed and some are capable of penetrating the blood–brain barrier, such as lorlatinib, in anaplastic lymphoma kinase-positive non-small cell lung cancer or afatinib in epidermal growth factor receptor-positive non-small cell lung cancer. These targeted agents are effective and well-tolerated, and increase progression-free survival. Therefore, identifying target mutations is crucial to administering these target-specific agents properly.<sup>16</sup>

The current case is unique and important because the patient first presented with unilateral SSNHL with vertigo and there was a suspicion of bilateral vestibulopathy in the video head impulse test. When such non-specific clinical findings are observed, an MRI with gadolinium contrast should be considered to detect any IAC lesions.

#### Declaration of conflicting interest

The authors declare that there is no conflict of interest.

#### Ethics statement

Study protocol approval was not required because this is a case report. Written informed consent was obtained from the patient for publication of the case report and accompanying images, and all details have been deidentified to protect the patient's identity.

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