

Case Report

Ruptured aberrant internal carotid artery pseudoaneurysm presenting with spontaneous massive ear bleeding following a single sneeze: a case report

Seiichiro HIRONO¹⁾ Eiichi KOBAYASHI¹⁾ Koichi EBIHARA²⁾ Michihiro HAYASAKA²⁾
Homare SUZUKI³⁾ Yoshitaka OKAMOTO³⁾ Naokatsu SAEKI¹⁾

- 1) Department of Neurological Surgery, Chiba University Graduate School of Medicine
2) Department of Neurological Surgery, Kimitsu Chuo Hospital
3) Department of Otorhinolaryngology-Head and Neck Surgery, Chiba University Graduate School of Medicine

●Abstract●

Objective: An aberrant internal carotid artery (ICA) is a rare congenital vascular malformation and many previous studies have emphasized this anomaly because of an increased risk of injury during otological surgical procedures. However, neither spontaneous massive bleeding nor infectious pseudoaneurysm associated with aberrant ICA has been reported to date.

Case presentation: We present the case of a 54-year-old healthy female patient who experienced sudden massive bleeding from the ear following a single sneeze. This patient had suffered from ipsilateral otorrhea due to acute otitis media for one month. Based on a radiological examination in combination with past medical history, infectious pseudoaneurysm associated with aberrant ICA was diagnosed as the cause of bleeding. Although an embolic infarction during the endovascular procedure induced slight hemiparesis, the pseudoaneurysm was successfully managed with endovascular coil trapping.

Conclusion: Although aberrant ICA has been reported as a possible cause of ear bleeding during otological procedures in the past, this is the first report of a spontaneously ruptured pseudoaneurysm associated with aberrant ICA. This case indicates that it is important to consider ruptured pseudoaneurysm in the differential diagnosis of ear bleeding, and also provides evidence that endovascular treatment is suitable for this condition.

●Key Words●

aberrant internal carotid artery, coil trapping, ear bleeding, infectious pseudoaneurysm

<Correspondence Address : Seiichiro HIRONO, 1-8-1 Inohana, Chuo-ku, Chiba-city, Chiba, 260-8670, Japan E-mail: seiichirohirono@chiba-u.jp>

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Introduction

An aberrant internal carotid artery (ICA) is a rare vascular anomaly that is thought to develop from early agenesis of the first embryonic segment of the ICA and compensatory hypertrophy of the inferior tympanic branch of the ascending pharyngeal artery and the caroticotympanic branch of the ICA⁷⁾. Although this anomaly, which mimics a middle ear mass, is often reported as causing pulsatile tinnitus or ear bleeding following traumatic injury due to myringotomy or other

surgical procedures, no case of aberrant ICA with spontaneous massive bleeding has been previously reported. Furthermore, infectious pseudoaneurysm (i-pseudoaneurysm) associated with aberrant ICA is extremely rare, and an optimal treatment strategy has not yet been established.

Case report

A 54-year-old female patient who had a one-month history of right otorrhea hemorrhaged heavily from the right ear soon after a single sneeze. Upon arrival at our

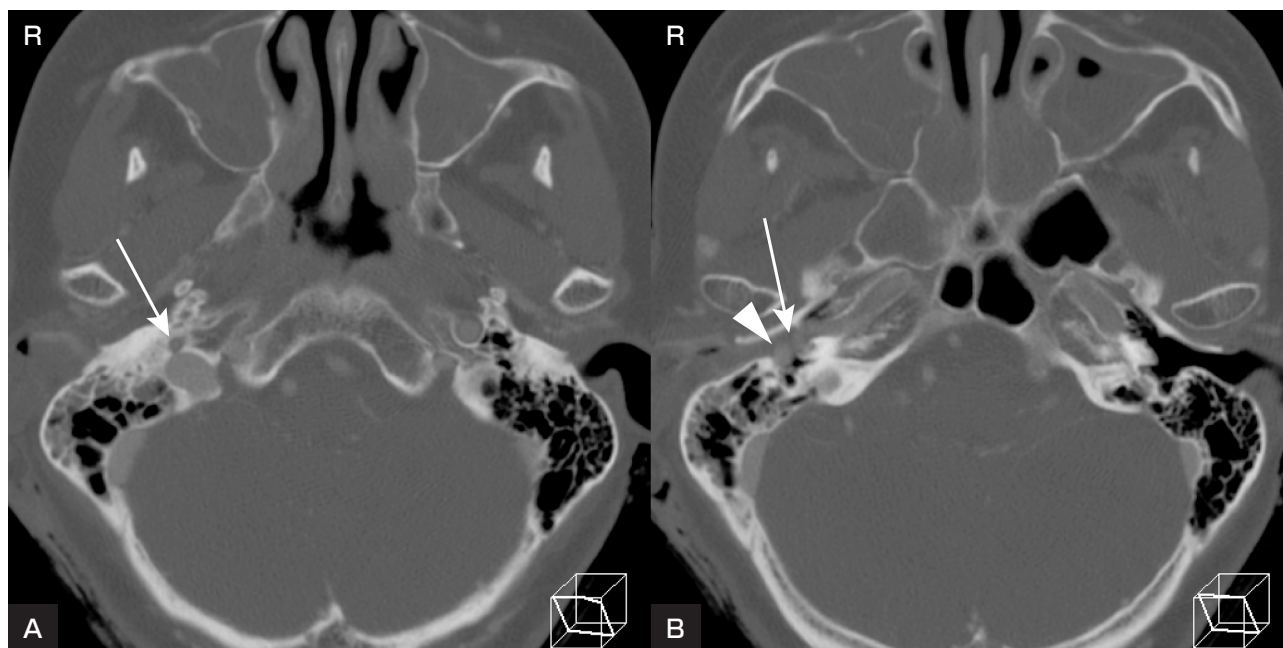


Fig. 1 CT scan on arrival.

- A** : The aberrant internal carotid artery (ICA, arrow) running through the enlarged inferior tympanic canaliculus (ITyC).
B : The aberrant ICA (arrow) runs from the posterior in the hypotympanum to the mesotympanum, without a bony plate of the carotid canal. The protruded pseudoaneurysm (arrowhead) is also of note.

institute, a physical examination showed hypovolemic shock due to massive bleeding from the right ear. After the external ear canal was packed and sufficient red cell concentrates were transfused to stabilize circulation, computed tomography (CT) of the ear (**Fig. 1**) revealed that the right ICA was markedly situated posterolaterally with the absence of a bony plate between the middle ear cavity and the carotid canal; furthermore, the inferior tympanic canaliculus (ITyC) was larger than usual. Carotid angiography (**Fig. 2**) showed a laterally protruding 4.0×6.0 mm aneurysm at the genu of the vertical and horizontal segment of the ICA, and a narrowing of the ICA where it entered the ITyC. Based on these findings together with the patient's one-month history of right otorrhea, a diagnosis of spontaneous ruptured i-pseudoaneurysm with aberrant ICA was given. Further angiography of the left carotid artery and the dominant left vertebral artery demonstrated good filling of the right cerebral hemisphere through the Circle of Willis collaterals in addition to flow from the right middle meningeal artery (MMA) to the intracranial ICA.

To obtain complete hemostasis, a decision was made to interrupt the flow to the i-pseudoaneurysm with coil embolization. A 7Fr sheath (Terumo-Clinical Supply, Tokyo, Japan) was introduced into the right common femoral artery. A 7Fr guiding catheter with a balloon (Patlive; Terumo-Clinical Supply) was advanced into the proximal right cervical ICA in case proximal flow control was needed. A microballoon catheter with a 4.5 mm diameter balloon (Attendant; Terumo-Clinical Supply) was positioned in the aberrant ICA proximal to the pseudoaneurysm, and temporary occlusion was performed. Since the patient showed no obvious neurological deficits during temporary occlusion, the trapping procedure was continued. However, a small amount of contrast extravasation from the aneurysm was observed when a microcatheter (Excelsior 1018; Boston Scientific, Natick, MA, USA) was advanced distal to the aneurysm. The patient demonstrated slight left hemiparesis immediately after the extravasation, which was probably caused by embolic infarction.

Temporary inflation of the 7Fr balloon catheter in the



Fig. 2
Right internal carotid angiogram, frontal view (A) and lateral view (B). A 4.0 × 6.0 mm pseudoaneurysm (arrow) at the genu of a remarkably lateralized ICA. The arrowhead shows the narrowed ICA where it enters the ITyC.

cervical ICA successfully controlled the extravasation and trapping at the sites proximal and distal to the aneurysm was accomplished with 12 coils. Post-trapping angiography demonstrated complete embolization of the ICA and sufficient flow to the right cerebral hemisphere via the anterior communicating artery (Acom), the posterior communicating artery (Pcom) and the MMA (Fig. 3). Dosing of Cilostazol was held until post-trapping day 7 because of mild oozing from the external ear canal. The patient showed mild left hemiparesis and the post-treatment MRI scan (Fig. 4) confirmed the existence of an infarction in the right frontal and parietal lobe. After intensive physical therapy and rehabilitation, the patient was able to walk without any help and returned home as modified Rankin Scale 2.

Discussion

To our knowledge, this is the first report on spontaneously ruptured aberrant ICA pseudoaneurysm. Since many cases with bleeding from an aberrant ICA as a result of traumatic injury following myringotomy, biopsy or middle ear surgery have been reported^{4,10,12,15},

this anomaly is well-known in combination with traumatic pseudoaneurysm (t-pseudoaneurysm). In our patient, since there was a one-month history of ipsilateral otorrhea, we concluded that this aneurysm was an i-pseudoaneurysm, which has histological characteristics of complete disruption of the vascular wall by neutrophil infiltration throughout all layers of the arterial wall.⁹ The i-pseudoaneurysm in our patient ruptured easily after a single sneeze. In addition to the fragile nature, a defect of the bony plate between the aberrant ICA and the middle ear cavity may also have been involved in the ear bleeding observed in our patient.

Although ear bleeding is commonly associated with cranial trauma or a perforated eardrum, in most cases the amount of bleeding is very small. The present case showed massive ear bleeding with hypovolemic shock on arrival, which suggested arterial bleeding. Therefore, our findings in this patient indicate that a ruptured pseudoaneurysm of an aberrant ICA is important in the differential diagnosis of ear bleeding.

Compared to a ruptured t-pseudoaneurysm, which only occurs in hospitals following ear-related procedures, an



Fig. 3
Post-trapping right internal carotid angiogram, frontal view (A) and lateral view (B). The right ICA was completely embolized, and there was good collateral flow from the middle meningeal artery (arrowhead) to the territory perfused by the right ICA.

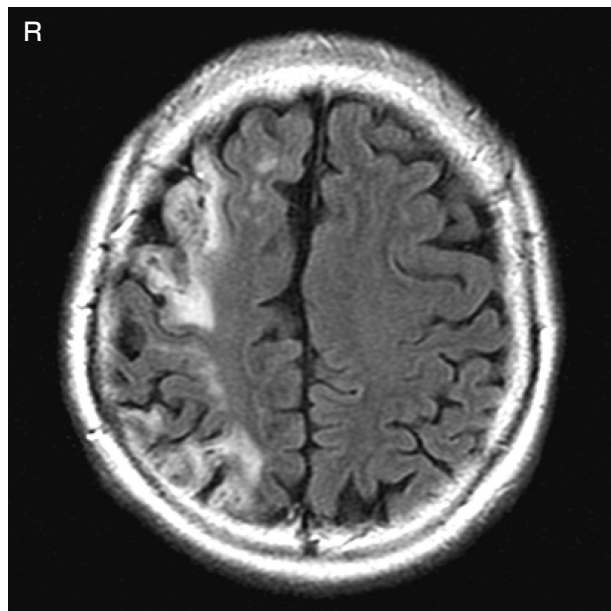


Fig. 4
Post-treatment MRI scan revealing the thrombotic infarction in the right frontal and parietal lobe.

immediate and accurate diagnosis of ruptured i-pseudoaneurysm is essential even though it is more difficult. A temporal bone CT scan should be performed

first to ensure the accurate diagnosis of aberrant ICA^{7,12}. The posterolateralization of the ICA in the middle ear cavity and enlarged ITyC are the key findings of an aberrant ICA. From an angiographic point of view, it is often difficult to distinguish a lateralized ICA (a normal anatomic variant) from an aberrant ICA, since some angiographic features are shared². Therefore, a temporal bone CT scan must be performed in combination with cerebral angiography. Briefly, lateralized ICA is not normally associated with enlarged ITyC, because it is not the result of partial agenesis of the ICA. A genu of the lateralized ICA protrudes into the anterior mesotympanum, whereas the aberrant ICA runs from the posterior of the hypotympanum across the middle ear cavity to the anterior mesotympanum along the posterior cochlear promontory.

Following a CT scan, carotid artery angiography is immediately required for confirmation of the diagnosis and to determine the origin of the bleeding, and again following endovascular treatment. Before the endovascular treatment era, direct surgery or ligation of the ICA in the neck was the only option to control

bleeding from an aberrant ICA t-pseudoaneurysm^{3,13}). However, these were associated with relatively high morbidity and mortality^{1,5,6}). After the first case treated by a balloon catheter was reported in 1983¹¹), endovascular treatment has been the first choice to control bleeding from an aberrant ICA t-pseudoaneurysm^{4,12,14}). Regardless of whether open surgery or endovascular treatment is performed, ICA occlusion is widely accepted for providing hemostasis, since a rupture in the pseudoaneurysm may cause life-threatening bleeding as in our patient. A review of literature regarding the management of non-infectious pseudoaneurysm suggests that covered stent-graft implants show promising results with no serious complications. However, this method has not been used in the treatment of a patient with any acute infectious processes⁸).

Although the patient showed good flow to the territory perfused by the ICA via the Acom, Pcom and MMA, she developed mild left hemiparesis probably due to embolic infarction. In this case, the risk of embolism was extremely high. First, the patient could not be treated with anticoagulant intravenous heparin during the procedure because of massive ear bleeding. Second, the aberrant ICA in our patient formed a sharp curve at the genu of the ICA, and we encountered difficulties in navigating a microcatheter distal to the i-pseudoaneurysm, which contained unstable thrombi. Those thrombi could easily migrate to the distal ICA area and cause infarction. Since an infectious process was present in our patient, this in itself might have increased the risk of thrombosis. All of these factors may have been involved in the embolic infarction in our patient.

Conclusion

We report here the first case of spontaneous ear bleeding from i-pseudoaneurysm of an aberrant ICA, which is important in the differential diagnosis of spontaneous ear bleeding. This case report provides evidence that a ruptured i-pseudoaneurysm of an

aberrant ICA can be managed successfully with endovascular trapping.

The authors declare that they have no conflict of interest.

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