Case Report

Flow Diversion for an Extracranial Infectious Internal Carotid Pseudoaneurysm Secondary to Exudative Otitis Media: Illustrative Case

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Objective: Treatment of infectious aneurysms is challenging because of the fragility of the vessel walls. Surgical trapping and endovascular parent artery occlusion are the treatments of choice for medically intractable infectious aneurysms. Here, we describe a patient with an infectious aneurysm at the extracranial petrous segment of the internal carotid artery that was secondary to exudative otitis media; it was successfully treated with reconstructive endovascular interventions using a flow redirection endoluminal device (FRED).

Case Presentation: A 57-year-old man was administered antibiotics for exudative otitis media. After 6 months, the patient underwent CT screening, which revealed destruction of the petrous bone and a pseudoaneurysm at the petrous segment of the left internal carotid artery. Catheter angiography, including a balloon occlusion test, revealed a 47-mm wide-necked pseudoaneurysm at the distal cervical segment of the left internal carotid artery with poor collateral blood supply to the left internal carotid artery. We performed reconstructive endovascular treatment using a FRED. A follow-up catheter angiogram after 6 months confirmed a minor neck remnant of the aneurysm.

Conclusion: Flow diversion may be a useful treatment approach for extracranial infectious aneurysms, despite concerns about incomplete obliteration of the aneurysm and the persistent risk of re-rupture before complete obliteration.

Keywords flow diversion, infectious aneurysm, exudative otitis media

Introduction

Infectious aneurysms are rare cerebrovascular lesions that pose a significant therapeutic challenge due to their angiographic and pathophysiologic features.[1,2)]

Infectious aneurysms are typically found after or concomitantly with systemic bacterial infections, especially infective endocarditis.[1,2)] Infectious aneurysms are found in 3%–10% of patients with infectious endocarditis, develop because of septic embolisms, and are commonly found in the distal cerebral arteries.[1,2)]

Infectious aneurysms are typically pseudoaneurysms that are prone to rupture due to their fragile vascular walls.[1,2)] The treatment of infectious aneurysms is challenging. The first-line treatment is intensive antibiotic therapy.[1,2)] Infectious aneurysms can spontaneously resolve, decrease in size, or thrombose after antibiotic treatment.[1,2)] Surgical and/or endovascular interventions should be considered for medically intractable infectious aneurysms.[1,2)] Surgical neck clipping and intraaneurysmal coil embolization are not feasible because of the potential risk of rupture during the procedure.[1,2)] Surgical trapping and endovascular parent artery occlusion are general treatments for infectious pseudoaneurysms.[1,2)] However, this treatment strategy is challenging in cases of infectious aneurysms of the internal carotid arteries (ICAs), whose antegrade flow must be maintained. In

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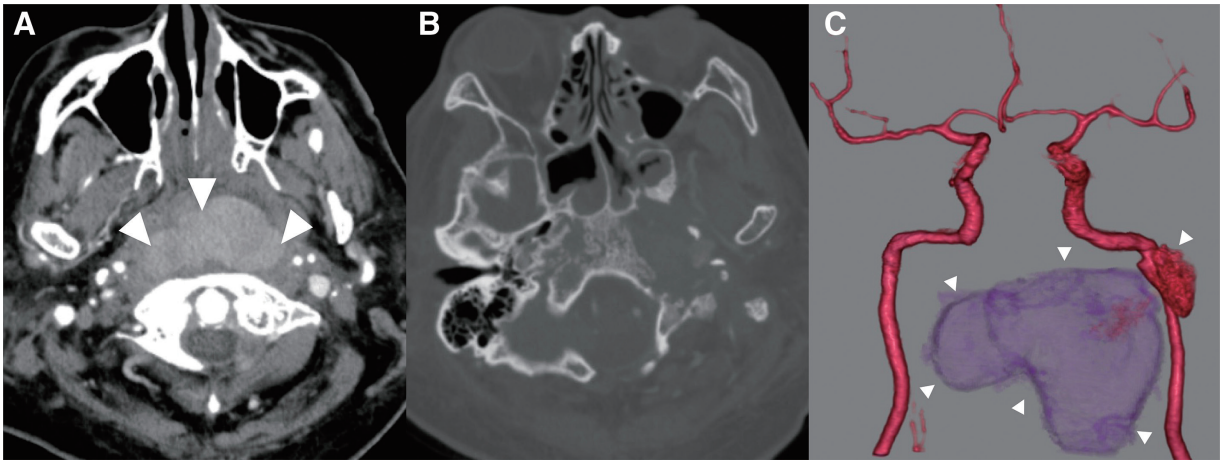

Fig. 1 CT on admission. (**A**) Axial CT scan showing a large mass (arrowhead) with contrast enhancement in the upper and middle pharynx at admission. (**B**) Axial bone imaging from the CT scan showing osteolysis of the left petrous bone. (**C**) CT angiogram showing the formation of a pseudoaneurysm (arrowhead) in the petrous segment of the left internal carotid artery.

such cases, surgical trapping or endovascular parent artery trapping should be combined with bypass surgery. Another treatment strategy could be flow diversion; however, previous reports on its use for treating infectious aneurysms are limited.[3–5)]

Here, we describe the case of a patient with an extracranial infectious aneurysm due to exudative otitis media who was successfully treated with endovascular reconstructive therapy with the deployment of a flow redirection endoluminal device (FRED).

Case Presentation

A 57-year-old man was admitted to our hospital with left otorrhea and left facial palsy. The patient was diagnosed with exudative otitis media. Therefore, antibiotic therapy was initiated. After 6 months, he was emergently hospitalized because of severe dyspnea. During screening of the entire body for infection, a CT scan revealed a large mass with contrast enhancement in the upper and middle pharynx (**Fig. 1A**). Destructive changes were also evident in the petrous bone (**Fig. 1B**). A CT angiogram was performed to evaluate vascular injuries caused by exudative otitis media, demonstrating a pseudoaneurysm at the distal cervical part of the left ICA (**Fig. 1C**). The diagnosis was an infectious aneurysm of the left extracranial ICA due to the destruction of the petrous bone after osteoclastic otitis media. The patient also experienced pneumonitis due to coronavirus disease 2019 (COVID-19). Intensive care was provided, including empiric antibiotic therapy, intubation, and respiratory management. The infectious large aneurysm was incidentally found during a screening test for systemic infection. The management of this patient requires intubation due to respiratory failure caused by concomitant COVID-19 infection, not due to the infectious large aneurysm. *Corynebacterium matruchotii* and *Pseudomonas aeruginosa* were detected in the otorrhea. After antibiotic therapy with piperacillin, tazobactam, and ceftolozane for 2 weeks, catheter angiography combined with a balloon occlusion test was performed. A 47.3-mm-diameter wide-necked aneurysm was evident in the distal part of the cervical segment of the left ICA (**Fig. 2A**). The balloon occlusion test of the left ICA further demonstrated poor collateral blood supply via the anterior and left posterior communicating arteries (**Fig. 2B** and **2C**). The patient had impaired consciousness after occlusion of the left ICA, indicating that the patient could not tolerate left ICA occlusion.

Herein, we discuss the treatment strategies employed. Catheter angiography suggested that the anterograde flow of the left ICA should be maintained due to insufficient collateral flow. For cases of surgical trapping, a high-flow bypass should be created if collateral flow is insufficient.[6)] We were concerned about the occlusion of the high-flow bypass because of the osteoclastic otitis media. Therefore, a flow diversion device was implanted. The patient was pretreated for 10 days with oral antiplatelet drugs (3.75 mg/day prasugrel and 100 mg/day aspirin). After the introduction of general anesthesia, intravenous heparin was administered during the procedure to maintain an activated coagulation time of between 250 and 300 seconds.

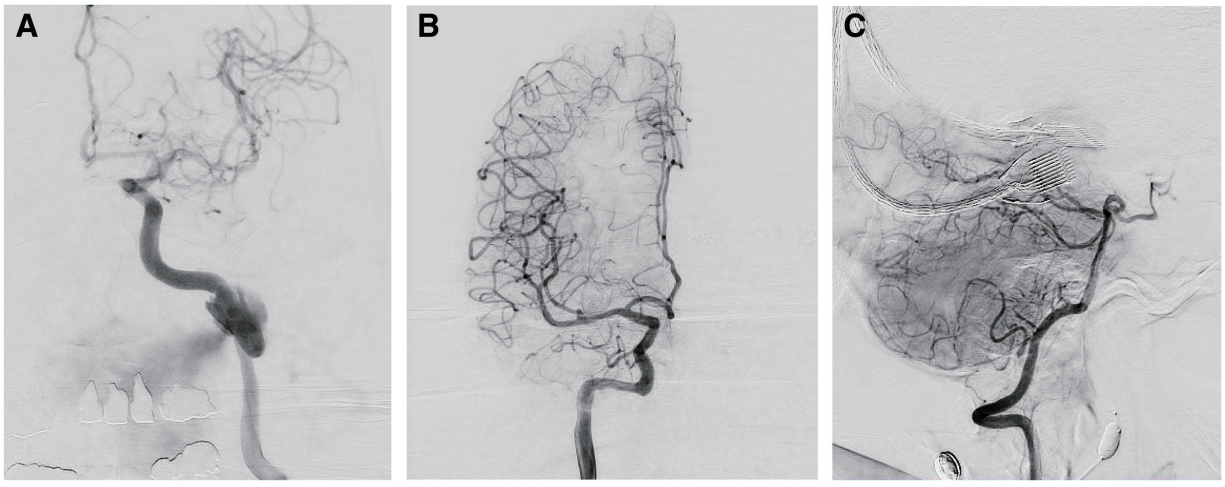

Fig. 2 Catheter angiogram with balloon occlusion test. (**A**) Left internal carotid angiogram showing a pseudoaneurysm and extravasation in the proximal petrous segment of the ICA. (**B**) Right carotid artery angiogram combined with transient occlusion of the left ICA using a balloon, showing no collaterals via the anterior communicating artery. (**C**) Left vertebral artery angiography combined with transient occlusion of the left ICA using a balloon, showing no collaterals via the left posterior communicating artery. ICA, internal carotid artery

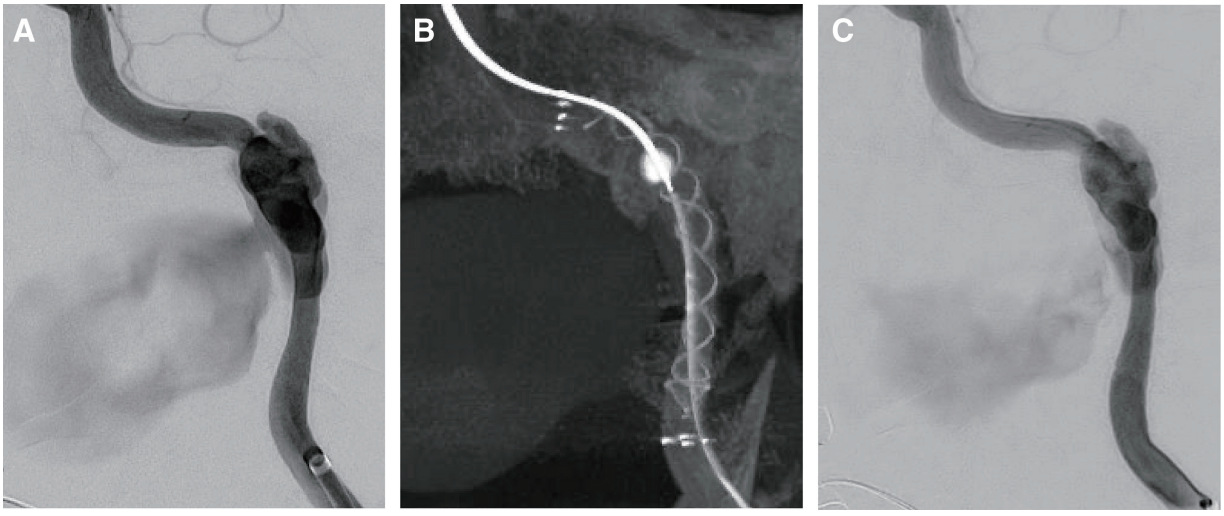

Fig. 3 Flow diversion. (**A** and **B**) Deployment of the FRED from the petrous segment into the distal cervical segment of the left internal carotid artery. (**C**) Internal carotid angiogram just after deployment of FRED. FRED, flow redirection endoluminal device

A 6-French Fubuki guiding catheter (ASAHI INTECC, Aichi, Japan) was placed in the ICA. A Sofia Select EX (Terumo MicroVention, Tokyo, Japan) was advanced into the ICA proximal to the aneurysm using a coaxial technique with a microcatheter over a microwire. Next, a Headway27 microcatheter (Terumo MicroVention) was inserted coaxially and navigated through the petrous part of the left ICA using a 0.014 microguidewire. A FRED measuring 5.5 × 32 mm was deployed by unsheathing over the microcatheter with a slow push/pull technique (**Fig. 3A** and **3B**). A catheter angiography performed immediately after FRED deployment confirmed that contrast stagnation in the aneurysm was not evident (**Fig. 3C**). A CT scan and MRI performed 1 day after endovascular treatment confirmed no evidence of hemorrhagic or thromboembolic infarction (data not shown).

Postoperatively, the patient received antiplatelet therapy with prasugrel and aspirin for 6 months and antibiotic therapy with tazobactam and ceftolozane for 3 months. One month after deployment, a catheter angiogram confirmed shrinkage of the pseudoaneurysm (**Fig. 4A**). Follow-up catheter angiography performed at 1 and 5 months after flow diversion confirmed gradual thrombosis of the pseudoaneurysm, and it showed entry remnants of the pseudoaneurysm 6 months after FRED deployment (**Fig. 4B**).

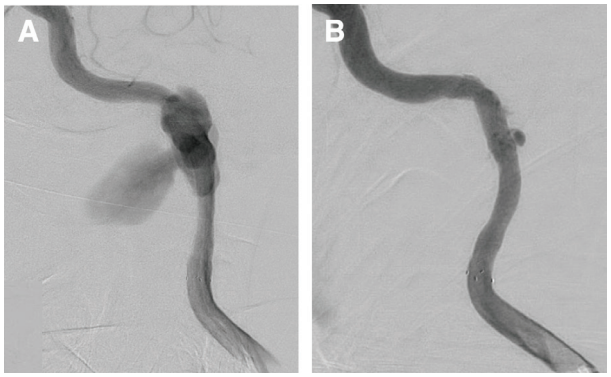

Fig. 4 Follow-up catheter angiogram. Catheter angiogram at 1 (**A**) and 6 months (**B**) after flow diversion.

Discussion

Here, we describe a rare case of an infectious extracranial ICA aneurysm caused by exudative otitis media that was successfully treated with reconstructive endovascular therapy using FRED. There are several case reports on ruptured pseudoaneurysms at the cervical-petrous part of the ICA secondary to infection around the petrous bone.[7–11] Exudative otitis media and otitis externa can invade the petrous bone and carotid arterial wall, leading to the formation of a pseudoaneurysm. *Pseudomonas aeruginosa* appears to be the most common causative organism for pseudoaneurysms secondary to exudative otitis media and otitis externa.[11,12] These case reports highlight the need to evaluate the ICA in cases of chronic or exudative otitis media and otitis externa, which can lead to destructive changes in the petrous bone.

The treatment of infectious aneurysms of the major arteries is challenging. Surgical or endovascular trapping of infectious aneurysms in the distal intracranial arteries is usually performed. It is critical to determine whether the antegrade flow of the parent artery can be sacrificed in cases of infectious aneurysms of major arteries.[6,8] A balloon occlusion test is mandatory to determine the necessity of bypass surgery or reconstructive endovascular interventions.[8] Based on a previous hemodynamic study by Shimizu et al.,[6] we discussed the requisite for bypass. The patient had poor collaterals via the anterior and posterior communicating arteries on the balloon occlusion test. The patient did not tolerate it during the balloon occlusion test. Unfortunately, we did not obtain hemodynamic data from the balloon occlusion test. If we take the bypass and trapping strategy, a high-flow bypass is best for securing the flow in the ICA territory. We considered that the superficial temporal artery (STA)-middle cerebral artery (MCA) double bypass or bypass to the proximal M2 is insufficient to supplement the MCA territory. However, there is a concern regarding the infection due to exudative otitis media, which can cause vascular injuries to the STA. Thus, a high-flow bypass was discouraged in this case. Therefore, flow diversion was selected as the first-line treatment for an infectious large ICA aneurysm at the distal cervical portion.

Surgical trapping is one of the treatment strategies.[5] In this case, high-flow bypass was needed in combination with surgical trapping because of poor collateral blood supply to the vascular territories of the left ICA. However, we were concerned about the inflammation caused by the osteoclastic otitis media, which could lead to stenosis or occlusion of the high-flow bypass, as previously reported.[13] Therefore, we avoided performing surgical trapping with a high-flow bypass. As endovascular therapy was considered, coil embolization was difficult due to the size of the aneurysm. Given the limitations of other treatment options, flow diversion emerged as a viable alternative. However, case reports on this are limited.[14,15] After deployment, flow-diverting stents change the hemodynamics of the aneurysm, causing thrombosis within the aneurysm and acting as a scaffold to promote intimal growth.[16] So far, there are no reports demonstrating the detailed mechanisms of flow diversion's efficacy for infectious aneurysms. Previous studies reported the efficacy of the Pipeline embolization device (Medotronic, Minneapolis, MN, USA) for intracranial and extracranial pseudoaneurysms.[17–19] Bounajem et al. reported a multi-institutional case series on ruptured ICA pseudoaneurysms, including blister, dissecting, and iatrogenic pseudoaneurysms.[19] Complete occlusion was obtained by the deployment of the Pipeline Flex embolization device (PED) in 82.6% of the patients in this case series.[19] There was no thromboembolic event or aneurysmal rupture after the deployment of the PED.[19] These results indicate the safety and efficacy of the use of the PED for pseudoaneurysms.[19] However, the detailed mechanisms of the cure of pseudoaneurysms by flow diversion are not discussed in those reports. Infectious aneurysms are considered pseudoaneurysms that are brittle and fragile due to all 3 vascular layers being injured by high levels of inflammation and injuries caused by bacteria and their toxins.[1,17] Promotion of intimal formation would be the basic mechanism underlying the cure of pseudoaneurysms after flow diversion, which is the same as flow diversion for saccular or fusiform aneurysms. One case of ICA pseudoaneurysm due to malignant otitis externa was successfully treated with a

Surpass flow diverter (Stryker, Portage, MI, USA).[10] In our case, using a PED was not feasible because the diameters of the extracranial arteries were larger than those of the intracranial arteries. Therefore, we adopted reconstructive endovascular treatment using FRED to preserve the anterograde flow of the left ICA and prevent the growth and rupture of the infectious pseudoaneurysm. Collectively, these observations support the notion that flow diversion could be a therapeutic option for infectious aneurysms of major arteries in which antegrade flow should be preserved.

Several issues are associated with flow diversion in infectious pseudoaneurysms. A critical issue is the persistent risk of rupture before complete occlusion occurs. It typically takes several weeks to months for complete obliteration to occur after the deployment of flow-diverting stents for intracranial aneurysms, while endothelization across the flow-diverting stents occurs.[16,20] Second, the outcome after flow diversion for infectious aneurysms remains unknown. Greco et al. reported that the complete obliteration rate for pseudoaneurysms was 79% in the short term (<1 year) and 84% in the long term (>1 year), respectively.[17] Therefore, close follow-up is mandatory to confirm the thrombotic changes in aneurysms. In cases of incomplete obliteration, the rupture of infectious aneurysms remains a concern. In such cases, the deployment of another flow-diverting stent[21] or performing surgical trapping or parent artery occlusion is required.[6] Third, dual antiplatelet therapy with the use of a flow-diverting device can induce hemorrhagic complications. The safety of dual antiplatelet therapy for those with infectious pseudoaneurysms remains unknown. Fourth, the uncertainty of the efficacy of flow diversion for infectious aneurysms poses the risk of life-threatening events, such as thrombotic occlusion of the ICA and rupture of the infectious aneurysm. A safety STA-MCA bypass may be helpful in cases of thrombotic occlusion of the ICA. Therefore, flow diversion for infectious aneurysms needs approval from the institutional review board because of the off-label use of flow-diverting stents and the uncertainty of the safety and efficacy of this treatment.

Conclusion

This is a rare case report of successful flow diversion for an infectious extracranial internal carotid aneurysm with poor collaterals. Despite the successful clinical course in our case, there were critical concerns, such as uncertainty about its flow-disrupting effect and the risk of re-rupture. Further studies should investigate the safety and efficacy of flow diversion for infectious aneurysms of major extracranial/intracranial arteries.

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Ethics Approval

This study was approved by the ethics committees of the National Hospital Organization Sendai Medical Center. (Identification Number: 23-104).

Disclosure statement

The authors declare that they have no conflicts of interest.

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