

## Case Report

# Resection of a Large Cavernous Hemangioma Following Preoperative Embolization in a Child's Temporal Bone

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Primary intraosseous cavernous hemangiomas (PICHs) in the temporal bone are very rare. Most treated cases are in adult patients, and there are very few reports involving children. To our knowledge, no cases reported have ever actually adopted embolization in children, although several reports recommend preoperative embolization of the feeding arteries. Here, we present a case of a child with a large cavernous hemangioma developing in the temporal bone. The patient was an 11-year-old girl whose chief complaints were hearing loss, aural fullness, and otalgia. Based on imaging, a lymphoma was initially suspected, but extensive arterial bleeding occurred during biopsy under local anesthesia. Angiography was performed to evaluate the blood flow in the tumor, and revealed the middle meningeal artery as a main feeding artery to the tumor. A huge PICH at the temporal bone was successfully resected following preoperative embolization of the feeding artery.

**KEYWORDS:** Primary intraosseous cavernous hemangioma, temporal bone, external auditory canal, embolization, angiography

## INTRODUCTION

Primary intraosseous hemangiomas are rare bone tumors comprising 0.7% to 1.0% of all bone tumors.<sup>1</sup> Most of them are seen in the vertebral column and are rarely found in the skull. They are mostly encountered in adults in the fourth and fifth decades of life.<sup>2</sup>

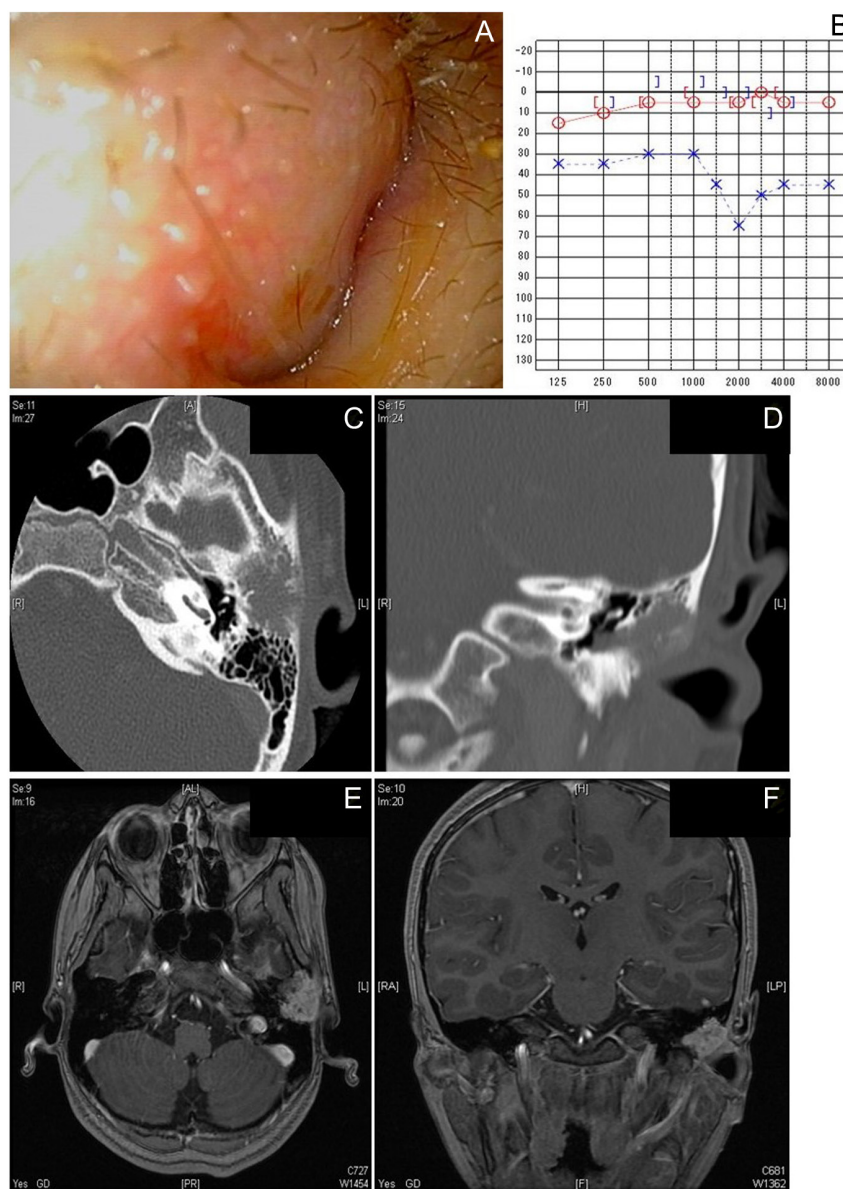
Hemangiomas can be histologically divided into 3 types: cavernous, capillary, and mixed. Most hemangiomas in the skull are of the cavernous type (PICHs). In contrast, hemangiomas in the spine are usually of the capillary type.

To date, approximately 100 cases of PICHs of the skull have been reported. Most of these reports describe PICHs in the frontal bone; PICHs in the temporal bone are less common.<sup>3</sup> While 50 cases of cavernous hemangiomas in the internal auditory canal have been reported,<sup>4</sup> only 8 cases have been reported in the external auditory canal (EAC), including the tympanic membrane.<sup>5</sup> Thus, to the best of our knowledge, there are no reported cases of a cavernous hemangioma in the EAC of a pediatric patient.

Here, we present the first report of a pediatric case of PICH of the temporal bone, with an extremely hypervascular lesion requiring preoperative embolization of the tumor.

## CASE PRESENTATION

An 11-year-old female patient presented to our department with hearing loss, aural fullness, and otalgia in the left ear. Her left EAC was obscured almost completely by the tumor. The tumor had a smooth surface (Figure 1A). Audiometry indicated a conductive hearing loss in the left ear (Figure 1B). A computed tomography (CT) scan revealed a mass extending from the left ventral mastoid air cell into the EAC (Figure 1C and D). Magnetic Resonance Imaging (MRI) performed using contrast revealed a mass, and the

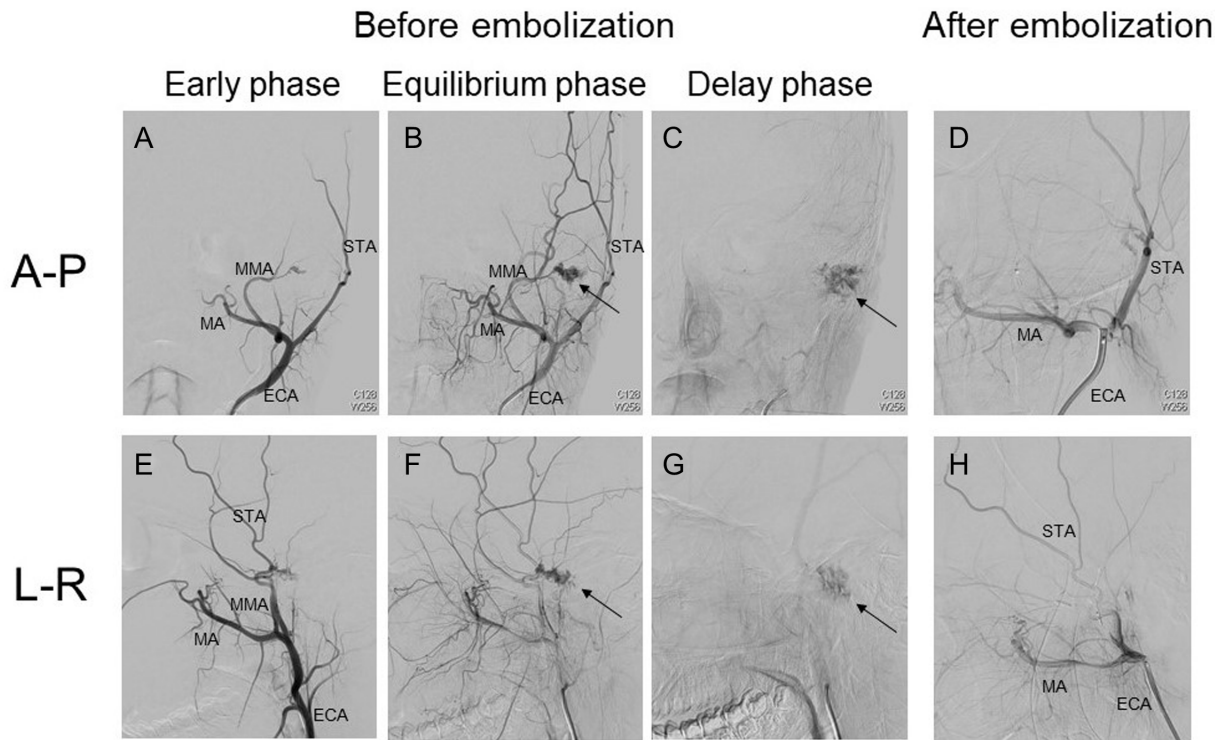


**Figure 1.** At the first consultation. (A) The patient's left EAC is nearly completely closed. (B) Audiometry shows conductive hearing loss in the left ear. (C and D) Axial and coronal CT scans show soft tissue density in the left temporal bone involving the EAC, with needle-like irregularities in the periphery and microcalcifications in the tumor. (E and F) Axial and coronal MRI scans show a well-enhanced tumor lesion in the left temporal bone involving the EAC, measuring 23 mm x 21 mm x 16 mm.

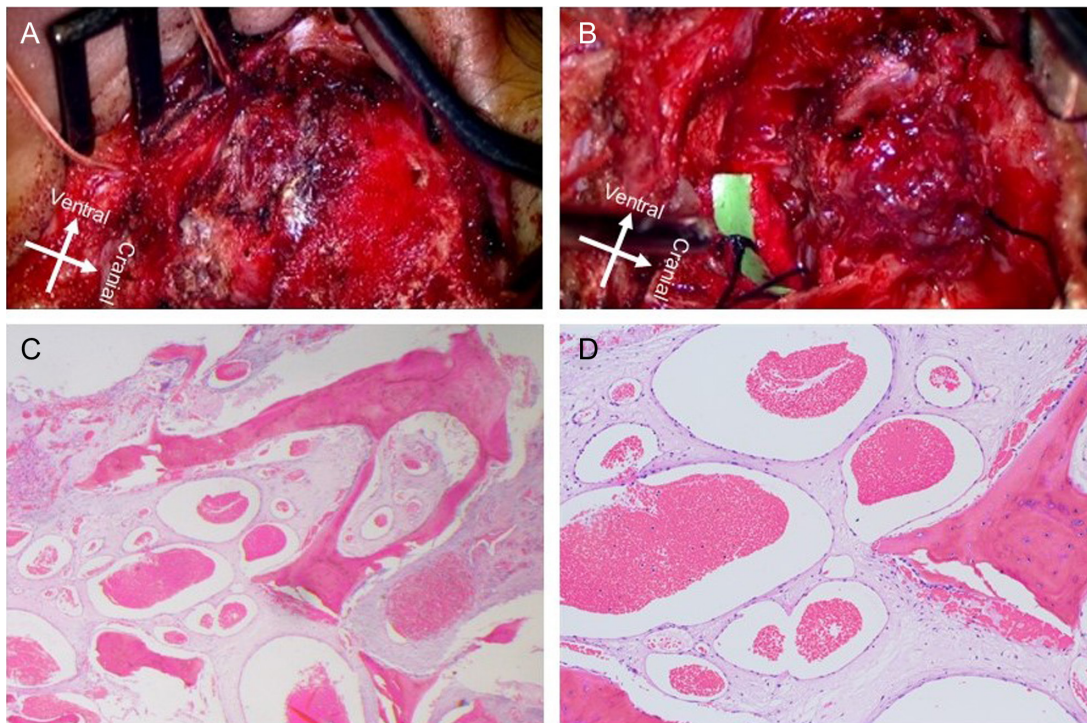
thickened dura in the left middle cranial fossa, measuring 23 mm x 21 mm x 16 mm (Figure 1E and F). Based on these images, we suspected lymphoma and planned a biopsy from the EAC under local anesthesia.

During the biopsy, unexpected severe arterial bleeding occurred immediately after incising the tumor. As we had to finish the procedure abruptly, only fragmented samples were obtained. The pathological diagnosis concluded a hemangioma. After the biopsy, we considered angiographic embolization of the tumor. Angiography revealed a hypervascular lesion in the left temporal bone, which was fed mainly by the middle meningeal artery. The draining vein was not identified because of contrast agent congestion (Figure 2). Based on these findings, we decided to perform a tumor resection under general anesthesia following embolization of the feeding artery of the tumor.

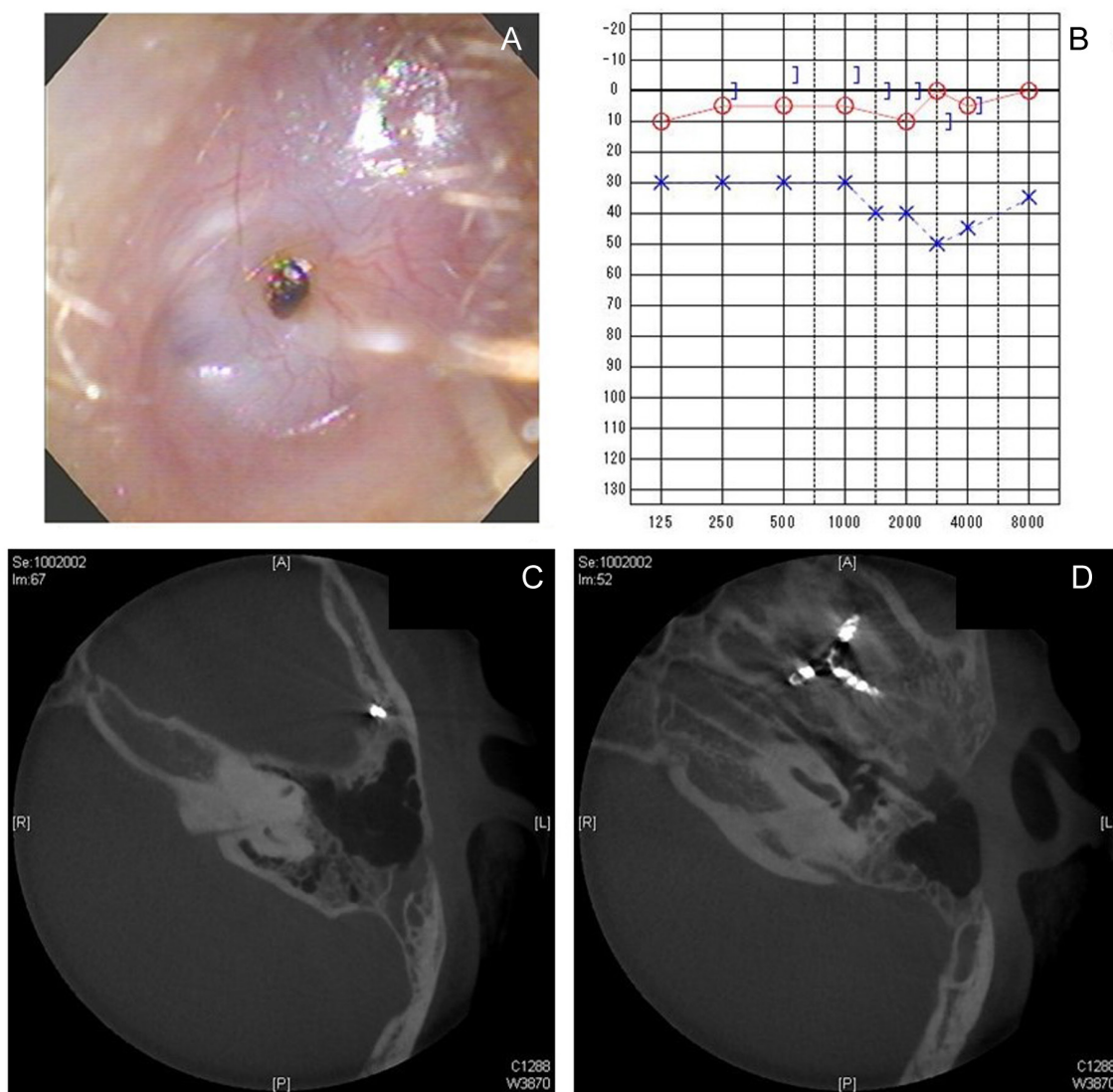
One month later, a temporal bone tumor resection was performed the day after embolizing the feeding artery. We pinned the patient's head so that we could move to craniotomy quickly if needed. The resection first involved a postauricular incision. The tumor was found in the subcutaneous soft tissue of the upper EAC (Figure 3A). We had to raise the skin flap carefully because the tumor adhered to the surrounding soft tissue. The tumor extended from the left ventral mastoid air cell to the EAC. The main tumor was found in the mastoid air cells around the epitympanum, and the tumor reached the mastoid air cells above the temporomandibular joint anteriorly. We subsequently resected the thin bone covering the cranial surface of the tumor and removed the tumor from the middle cranial base (Figure 3B). There was no bleeding from the middle cranial base. The dura of the middle cranial base was successfully preserved intact. Because the EAC was filled by the tumor and the epithelium was thinned, a part of the epithelium from the EAC wall was removed



**Figure 2.** Angiography before surgery. (A-D) A-P view. (E-H) L-R view. (A and E) early phase. (B and F) equilibrium phase. (C and G) delay phase. Before embolization, angiography of the middle meningeal artery shows that the contrast agent flows directly into the lesion (*black arrow*) in the early phase, congestion of the contrast agent in the lesion in the equilibrium phase, and no clear outflow. (D and H) After embolization, there is no inflow of contrast agent into the lesion.



**Figure 3.** A and B Intraoperative image. (A) The tumor was found in the subcutaneous soft tissue of the upper EAC. (B) The dissection was completed from the skull base, mastoid air cells, and anterior subcutaneous tissue. (C and D) Pathological finding shows a cavernous hemangioma with fibrous connective tissue, extended vascular lumen, and bone tissue.



**Figure 4.** After the operation. (A) The patient's left EAC is shallow, and a normal tympanic membrane cannot be observed. (B) Audiometry shows conductive hearing loss in the left ear, which remained at the same level as before the operation. (C and D) Axial and coronal CT scans show the coil used for embolization in the left temporal bone. The tumor was completely resected.

with the tumor. The tympanum remained intact. We removed the entire tumor, except for parts of the tumor that extended to the temporomandibular joint, which were separately removed completely. After removing the tumor, soft wall reconstruction with the fascia of temporal muscle was performed on the EAC. Despite severe arterial bleeding at the biopsy where only fragmented samples had been obtained, bleeding was completely controlled during the entire course of the resection due to preoperative embolization. Specifically, the amount of bleeding was 50 mL at the biopsy but only 30 mL at the time of resection.

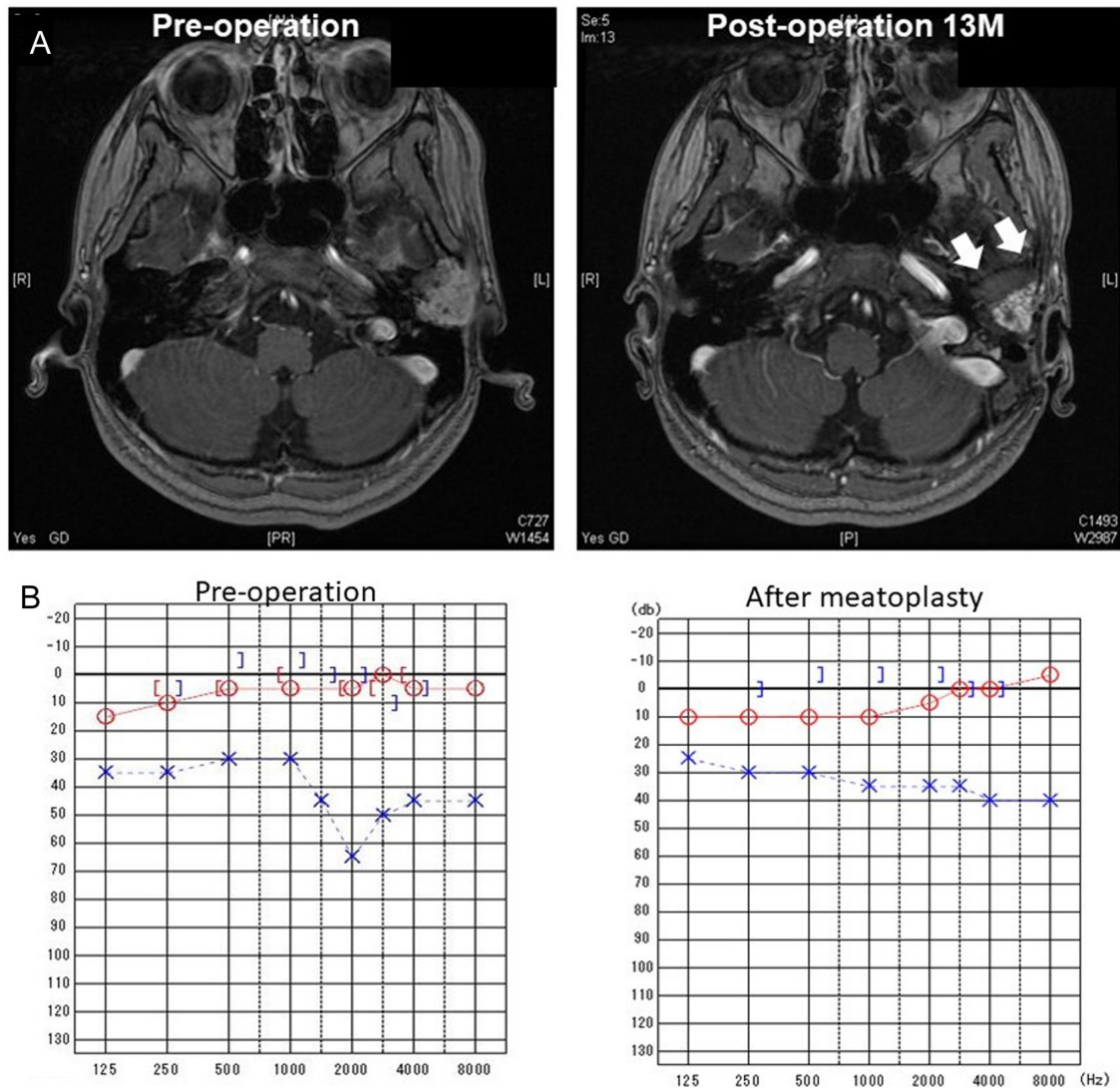
Pathological findings showed a cavernous hemangioma with fibrous connective tissue and an extended vascular lumen (Figures 3C and D).

The postoperative course was excellent, and the patient was discharged in approximately 1 week. The otalgia disappeared 2 weeks after the operation.

However, the hearing loss did not improve because the reactive proliferation of soft tissues in the EAC occurred postoperatively (Figure 4). The patient is reviewed at our hospital regularly. The meatoplasty was performed 12 months after the first total resection. Unfortunately, a recurrent lesion was found on the MRI taken 1 month after the meatoplasty. We are planning the revision surgery for total resection and the improvement of the remaining conductive hearing loss (Figure 5).

## DISCUSSION

PICs in the temporal bone are relatively rare, especially in children, and differential diagnosis is challenging. CT and MRI images are useful for the diagnosis of temporal bone lesions. Despite the typical imaging findings, we first considered the lesion to be a lymphoma in this case. In children, lymphomas are much more common than cavernous hemangiomas. Typical CT findings of PICs include a honeycomb tumor with surrounding needle-like irregularities and microcalcifications within the tumor.<sup>6,7</sup>



**Figure 5.** (A) MRI scans 13 months after the tumor resection reveals a recurrent lesion (arrows in the preoperative image indicate the limit of an initial tumor). (B) Audiometry after the meatoplasty shows a conductive hearing loss in the left ear, which showed slight improvement compared to the preoperative hearing level.

With MRI, PICHs are well enhanced, heterogeneous, isointense to the brain on T1-weighted, and isointense or hyperintense to the brain on T2-weighted images.<sup>6,7</sup> In this case, the CT images showed needle-like irregularities in the periphery, and microcalcifications were also found in the tumor. The MRI images showed a well-enhanced lesion, which was isointense to the brain on the T1-weighted and hyperintense to the brain on the T2-weighted image. These features are representative of the typical imaging findings of cavernous hemangiomas. In contrast, malignant lymphomas have almost the same imaging findings. Therefore, it is challenging to distinguish cavernous hemangiomas from lymphomas based on imaging alone. Another common differential disease of temporal bone tumor in children is rhabdomyosarcoma. Rhabdomyosarcoma appears as a homogeneous mass on CT with very mild contrast enhancement. With MRI, rhabdomyosarcoma enhances significantly with contrast and has an intermediate signal on both T1-weighted- and T2-weighted images.<sup>8</sup> PICHs and rhabdomyosarcoma can be distinguished by imaging.

In this case, we embolized the feeding artery of the cavernous hemangioma in the temporal bone before the removal of the tumor. Most intracranial hemangiomas are asymptomatic and rarely embolized. Although angiography should be considered when massive bleeding is expected, to our knowledge, no pediatric cases of embolization of a feeding artery have been reported for cases of temporal bone PICH. Because the tumor resection progressed from the surface to the deep part, it was considered important to reduce the bleeding as much as possible by embolizing the feeding artery coming from the deep part that could not be processed early during the operation. In this case, the angiographic image clearly showed the main feeding artery and the congestion of contrast agent in the lesion. Therefore, this is the first report of angiographic images of PICH in the temporal bone.

The only method to treat symptomatic hemangioma is complete surgical excision. Some papers report cases of recurrence of cavernous hemangioma in sinus or orbit after surgery.<sup>9,10</sup> PICHs in the

temporal bone are often asymptomatic, although frequently associated with chief complaints of bloody otorrhea, aural fullness, pulsatile tinnitus, and hearing loss. In this case, the patient suffered from hearing loss, aural fullness, and otalgia in the left ear. Unfortunately, while the otalgia disappeared, the conductive hearing loss remained postoperatively because of reactive connective tissue proliferation. Nevertheless, resolution of the otalgia improved the quality of life of the young girl.

## CONCLUSION

We report a pediatric case of complete resection of a cavernous hemangioma involving the EAC and the temporal bone, following preoperative embolization. Large temporal bone PICHs are very rare, difficult to diagnose, and challenging to treat, especially in children. However, embolization of the feeding artery led to optimal control of the bleeding, enabling the tumor to be successfully removed. The tumor resection improved otalgia and contributed to a better quality of life for the patient.

**Ethics Committee Approval:** This study was conducted in accordance with the approval of the ethics committee of the Keio University School of Medicine (approval no. 20200033).

**Informed Consent:** Written informed consent were obtained from the patient who participated in this study.

**Peer Review:** Externally peer-reviewed.

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**Conflict of Interest:** The authors have no conflict of interest to declare.

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