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Surgical Neurology International

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SNI: Neuro-Oncology

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Case Report

Combined endovascular therapy and surgery for central giant cell granuloma in the temporal bone: A case report

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Received: 20 June 2024
Accepted: 13 August 2024
Published: 20 September 2024

DOI
10.25259/SNI_487_2024

Quick Response Code:

ABSTRACT

Background: Central giant cell granuloma (CGCG) is an uncommon, benign intraosseous lesion that most frequently occurs in the mandible and maxilla.

Case Description: A 31-year-old female with a medical history of Kawasaki disease presented to our hospital complaining of a clogged right ear. Head computed tomography revealed a mass in the squamous part of the right temporal bone, with osteolytic changes and invasion of the external auditory canal, middle ear, temporomandibular joint, and mastoid air cells. Enhanced magnetic resonance imaging (MRI) showed a strong signal in the intraosseous lesion. Digital subtraction angiography revealed tumor staining from multiple feeders, including the middle meningeal, posterior deep temporal, and posterior auricular arteries. Preoperative feeder embolization using a detachable coil and Embosphere Microspheres were performed for the middle meningeal artery under general anesthesia. After the endovascular treatment, we operated on the temporal bone lesion. Postoperative enhanced MRI showed subtotal resection and residual tumor near the external auditory canal, which was left in place to prevent opening the external auditory canal. The histopathological examination showed proliferation of mononuclear cells intermingled with osteoclast-like multinucleated giant cells. A diagnosis of CGCG was made. The postoperative course was uncomplicated, and the patient was discharged on day 10 of hospitalization.

Conclusion: We reported a rare case of CGCG in the temporal bone, managed by endovascular therapy and surgical resection. This combination therapy resulted in subtotal resection, preserving surrounding normal structures, such as the external auditory canal and tympanic cavity.

Keywords: Central giant cell granuloma, Feeder embolization, Temporal bone

INTRODUCTION

Central giant cell granulomas (CGCGs) are rare benign lesions originating from bone tissue and occurring mostly in the mandible and maxilla.[4] The World Health Organization classified CGCG as a bone-related lesion rather than a tumor because its clinical behavior and radiographic features are those of a benign tumor.[18] In clinical practice, it is difficult to distinguish giant cell tumors (GCTs) from CGCGs, and both rarely involve the temporal bone.[14] Both types are benign osteolytic lesions that can sometimes be aggressive, with local recurrence rates of 10–20%.[5,15] Due to the local recurrence rates, surgical resection is the main treatment option. We report a rare case of CGCG in the temporal bone, managed by a combination of endovascular therapy and surgical resection.

CASE REPORT

A 31-year-old female patient with a history of fully-healed Kawasaki disease and no head trauma presented to our outpatient Department of Otolaryngology complaining of a clogged right ear. Head computed tomography (CT) revealed an osteolytic lesion in the right temporal bone that extended to the external auditory canal (EAC), middle ear, the roof of the temporomandibular joint (TMJ), and mastoid air cells [Figures 1a-c]. Head magnetic resonance imaging (MRI) T1 and T2 sequences revealed a solid extra-axial isointense well-delimited mass in the right temporal bone [Figures 1d and e]. A gadolinium contrast MRI T1 sequence showed a strong signal in the lesion of the temporal bone [Figure 1f]. Digital subtraction angiography of the right external carotid artery (ECA) showed multiple feeders of the bone tumor, including the posterior convexity and petrosquamous branches of the middle meningeal artery (MMA), posterior deep temporal artery (PDTA), and posterior auricular artery [Figures 2a-c]. Based on these imaging studies, we planned surgical resection of the bone tumor by the middle fossa approach. Given that MMA devascularization would be the last procedure and that hemorrhage during tumor removal would prevent radical resection, we performed endovascular feeder embolization before the surgical resection.

Under general anesthesia, a 6 Fr FUBUKI catheter (Asahi Intecc, Nagoya, Aichi, Japan) was navigated to the right ECA. An intermediate catheter (Guidepost; Tokai Medical Products, Aichi, Japan) was then advanced into the orifice of the MMA. An Excelsior 1018 microcatheter (Stryker, Kalamazoo, MI, USA) was inserted into the posterior convexity branch of MMA using a microwire (SynchroSELECT standard; Stryker); [Figure 3a]. MMA embolization was performed using Embosphere Microspheres (300–500 μm) through the Excelsior 1018 microcatheter. We did not use n-butyl-2-cyanoacrylat because MMA angiography showed an anastomosis between the recurrent meningeal and ophthalmic arteries. After embolization, the posterior convexity branch of MMA could not be visualized. Next, we performed embolization of the main MMA trunk using a detachable coil [Figure 3b]. After this procedure, ECA angiography revealed diminished MMA and enhanced staining from the PDTA [Figure 3c]. Subsequently, we performed tumor resection using the middle cranial fossa approach. The tumor eroded into the zygomatic arch, and the vertical segment of the temporal bone was distended. The tumor was firm, partially bony, and reddish brown [Figure 3d]. The mass was avascular in its cranial component but bled easily in its deep, caudal component. The resection was limited internally by the

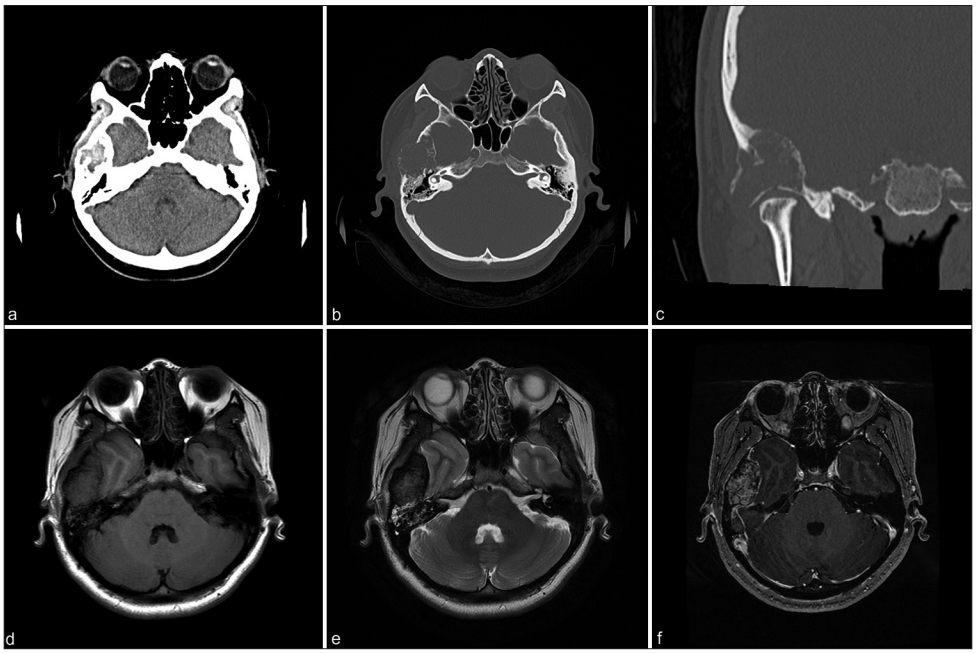

Figure 1: (a) Computed tomography (CT) demonstrating a temporal bone tumor. (b, c) CT bone window shows an expansive and destructive process in the temporal bone that extends to the external auditory canal, middle ear, temporomandibular joint roof, and mastoid air cells. (d, e) Initial head magnetic resonance imaging (MRI) revealed a well-delineated solid extra-axial isointense signal in the right temporal bone in T1 and T2 sequences. (f) A gadolinium contrast MRI T1 sequence shows strong enhancement.

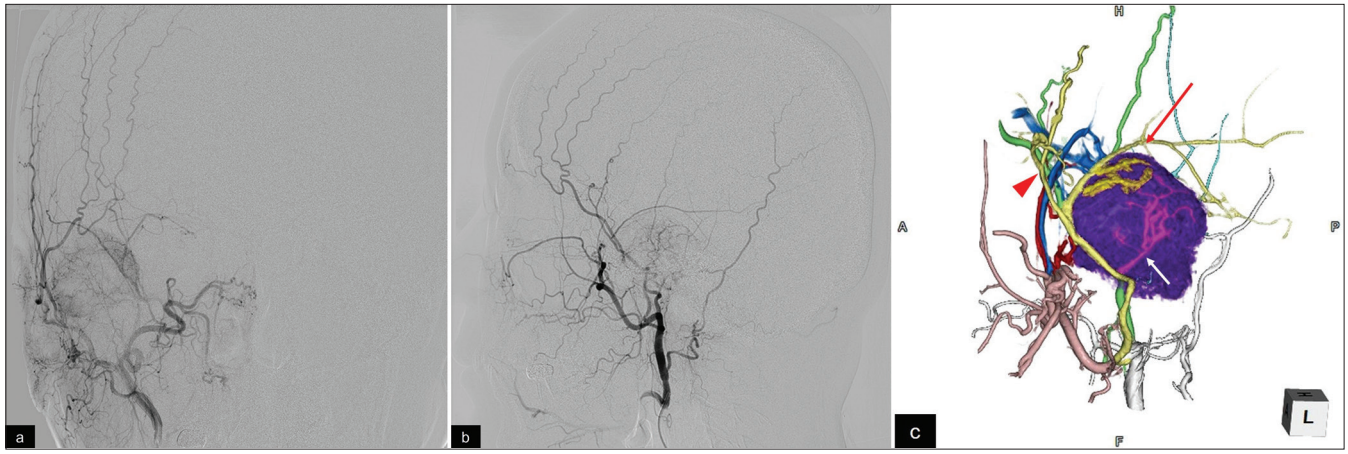

Figure 2: (a, b)Right external carotid artery angiography reveals multiple feeders for the bone tumor, including the posterior convexity and petrosquamous branches of the middle meningeal artery, posterior deep temporal artery, and posterior auricular artery (a, anterior view; b, lateral view). (c) Three-dimensional volume rendering shows tumor (purple) and frontal (red arrow head), petrosquamous (red arrow), and petrosal (white arrow) branch of MMA.

TMJ roof and caudally by the mastoid air cells and the tympanic cavity. Microscopically, the tumor margin was ossified, and the dura remained intact. Subtotal resection by piecemeal removal was achieved. The 3 × 3 cm defect in the temporal bone was filled with adipose tissue harvested from the abdomen.

A CGCG was suspected based on the pathology report of an intraoperative frozen sample. Hematoxylin-eosin staining of the resected lesion showed multinucleated osteoclast-like giant cells with spindle-shaped or polygonal stromal cells [Figure 3e]. Prussian blue staining detected hemosiderin [Figure 3f]. Based on these, CGCG was diagnosed. Postoperative MRI revealed subtotal tumor resection. The residual tumor was near the EAC [Figures 3g and h]. The patient was discharged on day 10 of hospitalization with no neurological deficits.

DISCUSSION

CGCGs were first reported by Jaffe[7] in 1953 as non-aggressive, bony lesions commonly occurring in the mandible and maxilla.[4] CGCGs in the temporal bone are rare, and distinguishing them from GCT in clinical practice is challenging.[14] Li *et al*.[9] reported that the most common initial symptom was an expanding mass in the temporal region or EAC (91.3%). In this case, the initial symptom reported by the patient was a feeling of a clogged right ear. The cause was an expanding mass in the EAC. The typical CT manifestation of CGCG is an expansive, well-defined lesion with scattered calcifications surrounded by a thin, calcified shell.[16] Preoperative diagnosis of CGCG is difficult, but some investigators indicated that it has characteristic MRI findings, including a thick low T1- and T2-weighted signal rim with variable central signal.[8,12] Enhanced MRI scans of early CGCG revealed septations, which are rarely found in GCTs.[9]

The pathogenesis of CGCG is unclear. It is assumed to be a hyperplastic reparative and self-healing reaction to intra-osseous trauma-induced hemorrhage that triggers the reactive granulomatous process.[6,17] On histological examination, GCTs are distinguished from CGCGs as they show more giant cells and higher mitotic activity.[6,11] Behjati *et al*.[1] reported novel histone H3.3 mutations in various bone and cartilage tumors. They suggested that 95% of chondroblastoma tumors harbor p.Lys46Met alterations in H3F3B and that 92% of the GCTs show H3.3 alterations in H3F3A. These mutations were found in a stromal cell population rather than osteoclasts or their precursor. Bernard *et al*.[2] reported recurrent somatic H3F3A and H3F3B mutations in CGCGs.

CGCG and GCT differ in their clinical course. Higher recurrence rates and risk of metastasis and malignant transformation were reported in GCT.[18] The standard treatment for CGCGs is complete surgical resection, with recurrence rates of 10–20%.[17] However, the vital structures around the tumor, including the TMJ capsule, EAC, mastoid air cells, and facial nerve, render a complete surgical excision difficult and sometimes impossible, especially when in the temporal bone. When radical resection is not performed, salvage radiation therapy should be considered.[3] However, one must remember that radiotherapy is less effective and harbors the risk of malignant transformation.[3,6,17] In this case, subtotal resection was performed. As the residual tumor was negligible, we decided not to administer any additional therapy.

The in-operative bleeding tendency of CGCG makes it a risk factor that hinders complete resection.[13] Although

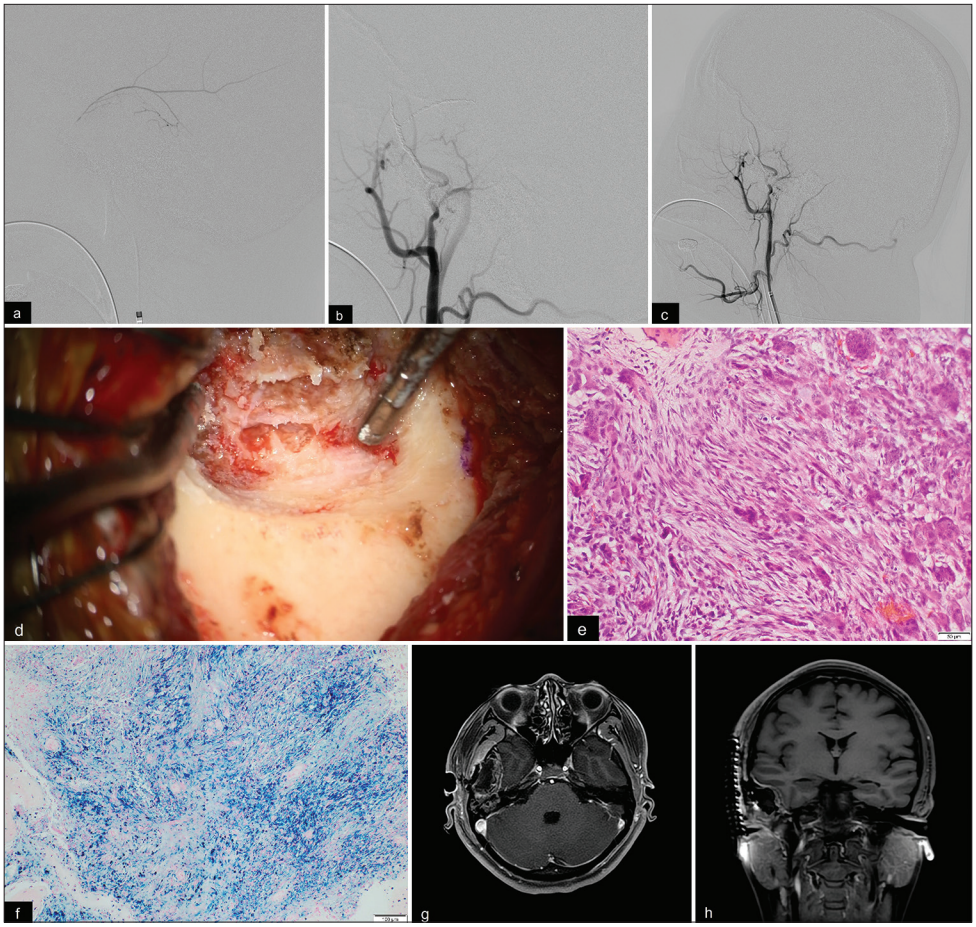

Figure 3: (a) Microcatheter was introduced into the posterior convexity branch of the middle meningeal artery (MMA), and an Embosphere microsphere (300–500 μm) was injected. (b) Embolization for the main MMA trunk, using a detachable coil. (c) External carotid artery angiography shows that the stain disappeared from the MMA. (d) Operative images show a firm, partially bony, reddish-brown tumor. (e) Hematoxylin-eosin staining shows multinucleated osteoclast-like giant cells with spindle-shaped or polygonal stromal cells. (f) Prussian blue staining shows the presence of hemosiderin. (g, h) Postoperative enhanced magnetic resonance imaging shows subtotal resection of the temporal bone tumor.

there is only one report about preoperative embolization with polyvinyl alcohol and detachable coils,[10] preoperative embolization of feeders is a useful way to achieve radical resection. We first reported the preoperative endovascular therapy that occluded the feeders of MMA by Embosphere and coils. These were de-vascularized in the final phase of the surgery. Like the surgery for meningiomas, the decreased hemorrhage during the surgery assisted in increasing the removed lesion percentage and shortening the operative time. It is controversial whether preoperative embolization should be performed. However, the previous report suggests that feeder embolization would be effective in preventing massive bleeding from the tumor.[10] In fact, we performed resection of the tumor without massive bleeding, and it would spend more operation time without preoperative embolization.

CONCLUSION

We reported a rare case of CGCG in the temporal bone. The patient presented with a complaint of a clogged right ear. Combining endovascular therapy and surgical resection resulted in subtotal resection while preserving the surrounding normal vital structures, such as the external auditory canal and tympanic cavity.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

1. Behjati S, Tarpey PS, Presneau N, Scheipl S, Pillay N, Van Loo P, *et al.* Distinct H3F3A and H3F3B driver mutations define chondroblastoma and giant cell tumor of bone. Nat Genet 2013;45:1479-82.
2. Bernard F, Troude L, Bouvier C, Roche PH. Giant cell reparative tumor: An exceptional differential diagnosis for a lytic lesion of the temporal bone. Neurochirurgie 2016;62:332-5.
3. Boedeker CC, Kayser G, Ridder GJ, Maier W, Schipper J. Giant-cell reparative granuloma of the temporal bone: A case report and review of the literature. Ear Nose Throat J 2003;82:926-9, 933-4, 936-7.
4. Chuong R, Kaban LB, Kozakewich H, Perez-Atayde A. Central giant cell lesions of the jaws: A clinicopathologic study. J Oral Maxillofac Surg 1986;44:708-13.
5. Dai WY, Tian C, Liu L. Case reports of a giant cell reparative granuloma and a giant cell tumor on temporal bone. Chin Med J (Engl) 2018;131:2254-6.
6. Dimitrakopoulos I, Lazaridis N, Sakellariou P, Asimaki A. Giant-cell granuloma in the temporal bone: A case report and review of the literature. J Oral Maxillofac Surg 2006;64:531-6.
7. Jaffe HL. Giant-cell reparative granuloma, traumatic bone cyst, and fibrous (fibro-oseous) dysplasia of the jawbones. Oral Surg Oral Med Oral Pathol 1953;6:159-75.
8. Lewis ML, Weber AL, McKenna MJ. Reparative cell granuloma of the temporal bone. Ann Otol Rhinol Laryngol 1994;103:826-8.
9. Li X, Wen Y, Zhang J, Wu N, Shen W, Yang S, *et al.* Imaging features, staging system, and surgical management of giant cell lesions of the temporal bone. Acta Otolaryngol 2022;142:553-61.
10. Matsui T, Iwamuro K, Ishikawa T, Asano T, Itoyama S, Tabe H. Large giant cell reparative granuloma of the petrous bone--case report. Neurol Med Chir (Tokyo) 2002;42:232-6.
11. Montero EH, Navarro JS, Pueyo JL, Garca FM, Sampériz LC, Garca AO. Giant-cell reparative granuloma in the temporal bone. Am J Otolaryngol 2003;24:191-3.
12. Moser A, Hoffmann KM, Walch C, Sovinz P, Lackner H, Schwinger W, *et al.* Intracranial reparative giant cell granuloma secondary to cholesteatoma in a 15-year-old girl. J Pediatr Hematol Oncol 2008;30:935-7.
13. Nemoto Y, Inoue Y, Tashiro T, Mochizuki T, Katsuyama J, Hakuba A, *et al.* Central giant cell granuloma of the temporal bone. AJNR Am J Neuroradiol 1995;16:982-5.
14. Orosz Z, Athanasou NA. Giant cell-containing tumors of bone. Surg Pathol Clin 2017;10:553-73.
15. Sepulveda I, Alzerreca J, Villalobos P, Ulloa JP. Giant cell tumor of the temporal bone with skull base and middle ear extension. J Med Cases 2021;12:149-51.
16. Shrestha S, Zhang J, Yan J, Zeng X, Peng X, He B. Radiological features of central giant cell granuloma: Comparative study of 7 cases and literature review. Dentomaxillofac Radiol 2021;50:20200429.
17. Souter MA, Bird PA, Worthington JP. Giant cell reparative granuloma of the temporal bone treated with calcitonin. Otol Neurotol 2006;27:999-1002.
18. Thompson L. World Health Organization classification of tumours: Pathology and genetics of head and neck tumours. Ear Nose Throat J 2006;85:74.

How to cite this article: Morimoto T, Ko S, Shimada K, Nishioka T, Tokunaga H. Combined endovascular therapy and surgery for central giant cell granuloma in the temporal bone: A case report. Surg Neurol Int. 2024;15:335. doi: 10.25259/SNI_487_2024