

## Case Report

# Inflammatory Pseudotumor of the Temporal Bone and Parapharyngeal Space: A Clinical Case

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Cite this article as: D'Orazio F, Falcioni M, Waskiewicz J, Tredici M, Khoury C, Mancini C. Inflammatory pseudotumor of the temporal bone and parapharyngeal space: a clinical case. *J Int Adv Otol*. 2025, 21(4), 1790, doi: 10.5152/iao.2025.241790.

We present the case of a 42-year-old female who experienced gradually worsening pain in the left ear, accompanied by hearing impairment and occasional ear discharge. These lesions rarely occur in the temporal bone. Computed tomography and magnetic resonance scans identified a mass of soft tissue located in the left mastoid, with intracranial invasion and mastoid erosion. Initial treatment involved surgery followed by histopathology, which confirmed the diagnosis of a plasma cell granuloma, also known as an inflammatory pseudotumor. After surgery, the pseudotumor invaded the ipsilateral parapharyngeal space and became surgically inaccessible. Steroid therapy and radiotherapy were proceeded with. When feasible, aggressive surgical intervention is advised as the primary treatment, supplemented by steroids and radiation therapy for persistent or recurrent conditions. At the last follow-up, the patient was not completely free of disease but symptom-free and in good general health.

**KEYWORDS:** Inflammatory pseudotumor, otalgia, parapharyngeal space, plasma cell granuloma, steroid therapy, temporal bone

## INTRODUCTION

This case describes an inflammatory pseudotumor originating in the temporal bone and extending into infratemporal and parapharyngeal regions.

Inflammatory pseudotumors are unencapsulated, benign growths initially identified by Bahadori and Liebow in 1973.<sup>1</sup> In the head and neck, the most common location is the orbit, followed by the meninges, paranasal sinuses, infratemporal fossa, and soft tissues. Involvement of the temporal bone, skull base, and facial nerve is uncommon.<sup>2</sup>

## CASE PRESENTATION

In July 2021, a 42-year-old woman was admitted to the emergency department with complaints of persistent left ear pain and hearing loss that did not improve with local or systemic antibiotics or pain medication for a month. The clinical history was remarkable for an osteosarcoma of the left arm that required multiple surgeries and adjuvant chemotherapy (methotrexate, cisplatin, and adriamycin). She was free of disease for the last 2 years. Written informed consent was obtained from the patient prior to publication.

Otomicroscopy showed a poly-lobulated and bleeding mass completely occupying the left external auditory canal (EAC) (Figure 1).

Tonal audiometry showed a severe mixed hearing loss with a large conductive component (ABG 45dB).

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Received: November 26, 2024 • Revision Requested: March 4, 2025 • Last Revision Received: April 9, 2025 •

Accepted: May 18, 2025 • Publication Date: July 28, 2025

Available online at [www.advancedotology.org](http://www.advancedotology.org)



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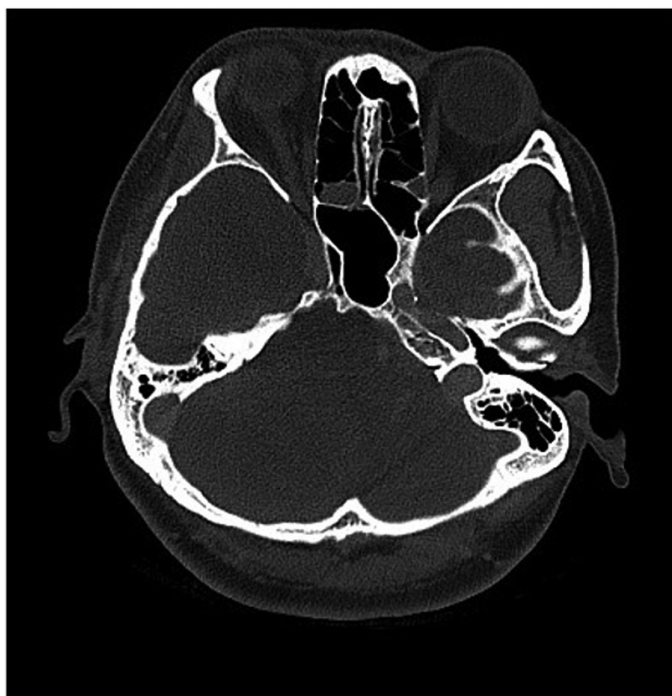


**Figure 1.** Left otoscopy image depicting a mass obturating the external auditory canal.

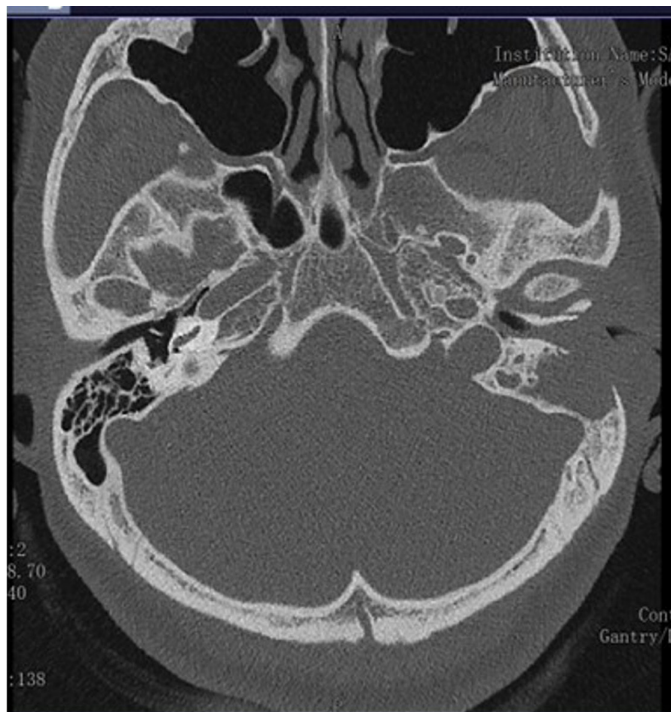
A cranial computed tomography (CT) scan performed as a routine follow-up for the osteosarcoma 7 months before the observation (October 2020) was completely negative. (Figure 2).

A new high-resolution CT of the temporal bone (Figure 3) revealed an osteo-destructive lesion at the level of the left mastoid, measuring approximately 22 × 14 mm, with extension into the EAC through an erosion of the posterior wall.

A magnetic resonance imaging (MRI) (Figure 4) confirmed the presence of a solid neoformation, hypointense in T1 and T2 sequences, with homogeneous enhancement after gadolinium administration.

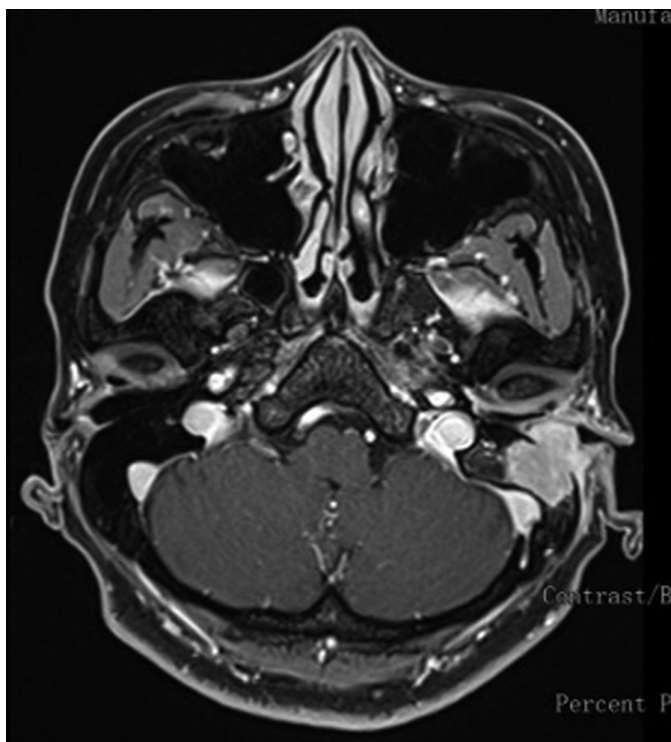


**Figure 2.** Follow-up cranial computed tomography for the osteosarcoma scan was completely negative.

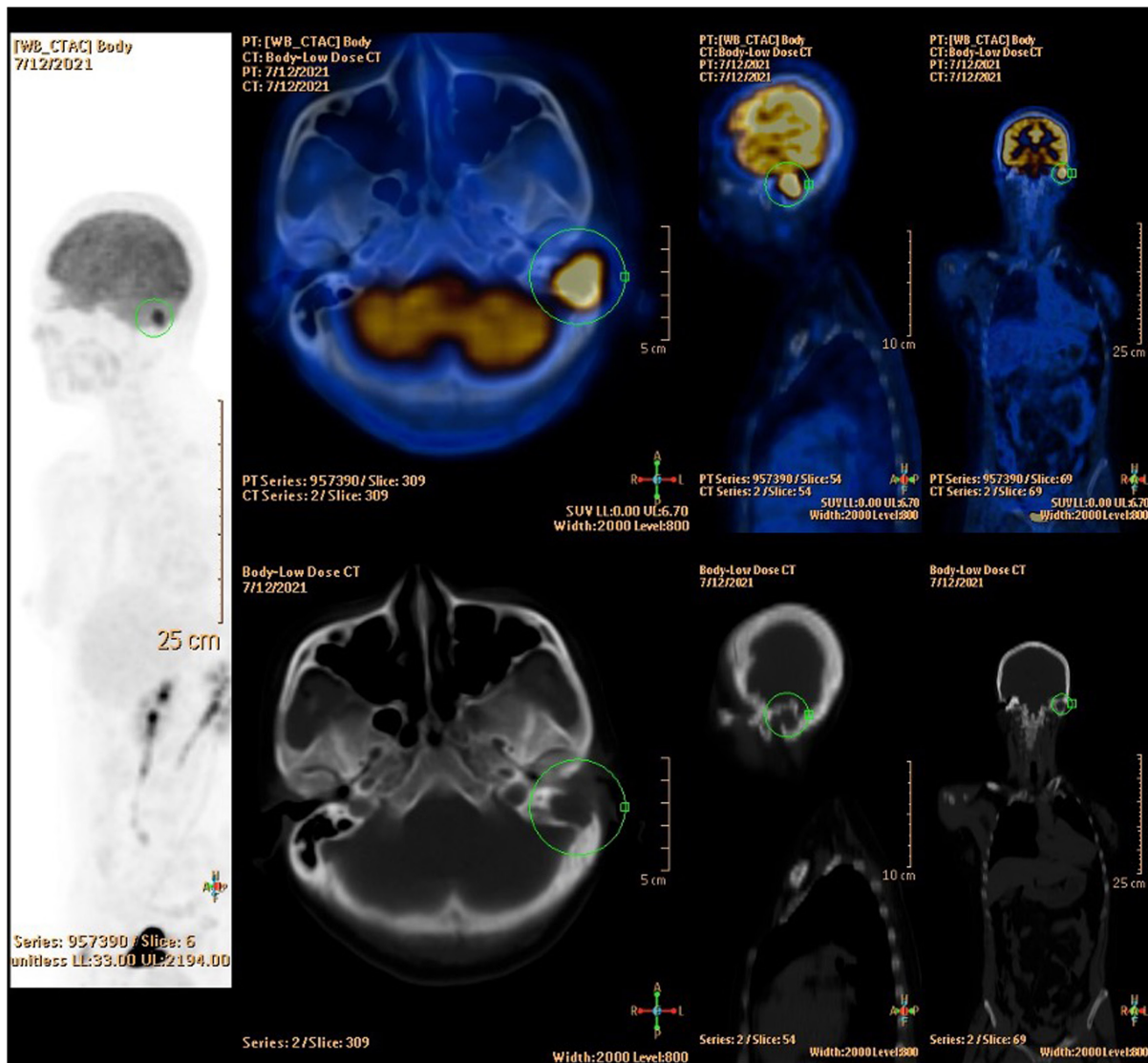


**Figure 3.** Ear computed tomography scan showing the osteo-destructive lesion at the level of the left mastoid.

Because of a clinical suspicion of an osteosarcoma metastasis, a positron emission tomography (PET) scan was performed (Figure 5) to exclude other locations; however, the left temporal bone was the only area of intense and pathological hyperaccumulation of the radiopharmaceutical agent (SUVmax 12.1-SUV median 1.9).



**Figure 4.** Axial contrast-enhanced T1 magnetic resonance imaging (at the time of diagnosis) showing a solid, hypointense formation, with homogeneous enhancement in the mastoid after gadolinium administration.



**Figure 5.** Positron emission tomography–computed tomography showing pathological hyperaccumulation of the radiopharmaceutical agent in the left temporal bone.

A biopsy of the lesion component protruding into the EAC resulted inconclusive.

In the persisting suspicion of an aggressive neoplastic lesion, a transmastoid removal was then planned; the option to intraoperatively convert the surgery to a subtotal petrosectomy (SP) was explained to the patient.

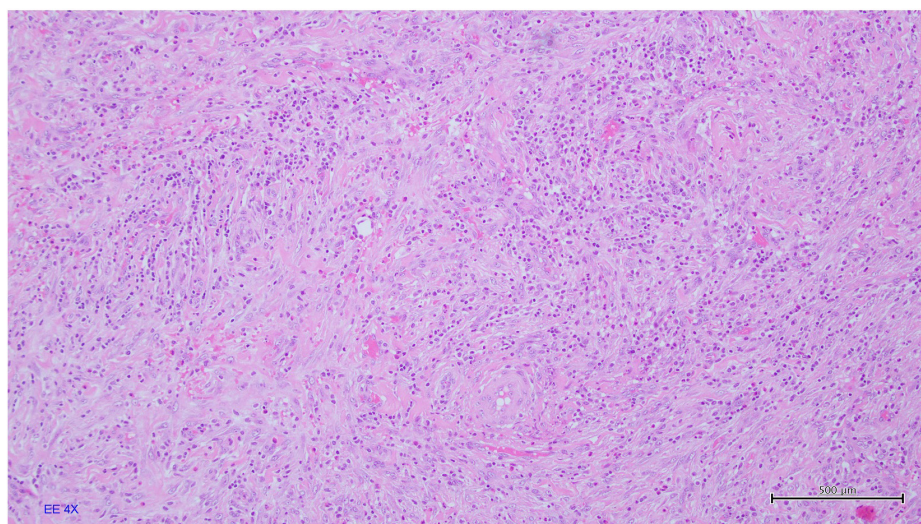
Intraoperatively the pathological tissue appeared extremely hemorrhagic and occupied the entire mastoid cavity, sparing the attic and the middle ear cleft but infiltrating the periosteal muscle plane and eroding the posterior wall of the EAC. The dura of the middle cranial fossa and the sigmoid sinus were focally uncovered. Lateral to the tympanic membrane, a secondary cholesteatoma entrapped in the EAC was also removed.

The result of the intraoperative histopathological examination was not conclusive; however, the aggressive behavior of the lesion suggested switching the operation to an SP, removing as much

perilesional bone as possible. The definitive histological examination allowed reaching the final diagnosis of plasma cell granuloma (inflammatory pseudotumor) (Figure 6). Immunostaining for light chains showed polyclonal plasma cells and IgG4+/IgG+ ratio <40% (Figure 7).

In view of the histological diagnosis and in accordance with the current scientific literature, the patient was treated with steroid therapy i.v. (methylprednisolone 125 mg/day i.v. for 2 weeks and then 80 mg/die for 3 weeks). At the 2 months follow-up clinically, the patient showed good healing of the retroauricular wound as well as the blindsac closure of the ear canal but complained of severe pain in the left cheek and temporal area, which was controlled with oxycodone and pregabalin twice a day. The follow-up MRI (Figure 8A) showed regular outcomes of the SP with adipose tissue filling all the surgical cavity. However, a large area of post-gadolinium enhancement and diffusion restriction was present along the skull base, with infiltration of the parotid gland, the temporomandibular joint, reaching the left parapharyngeal space till the nasopharynx, with





**Figure 6.** Histological examination (EE 10X) shows a proliferation of spindle cells with a storiform pattern, marked vascularity, and a mixed inflammatory cell infiltrate, including numerous reactive plasma cells in a collagenous background

involvement of the internal carotid artery and the internal jugular vein. Small lymph nodes were present close to the masseter muscle and at levels I and II in the neck. There were no signs of endocranial infiltration.

Methylprednisolone i.v. was continued at 125 mg a day for 10 days and then progressively subsided during the following 13 weeks. The therapy was temporarily suspended as the patient began to show collateral effects such as fatigue, ipsilateral eye swelling, Cushingoid face, and iatrogenic diabetes.

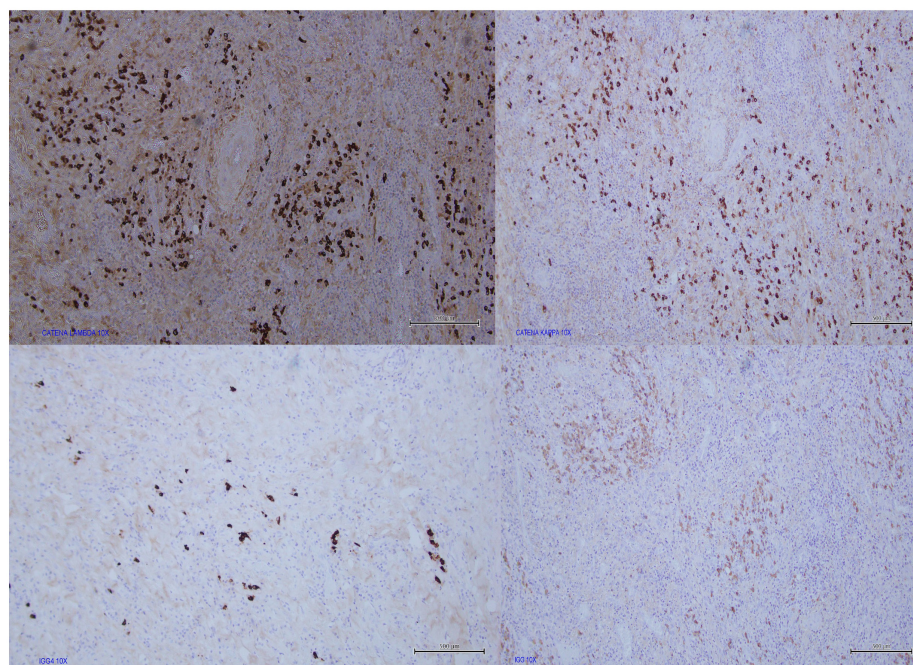
When the iatrogenic diabetes was pharmacologically controlled with Metformin 500 mg twice a day, methylprednisolone was restarted at a dosage of 80 mg a day for a further 2 months.

Because a new MRI (November 2021) failed to show any regression of the lesion, the methylprednisolone dosage was again increased to 96 mg a day but still without any radiological and clinical regression of the disease at 2 months (January 2022).

After a multidisciplinary discussion, radiation therapy (RT) was indicated.

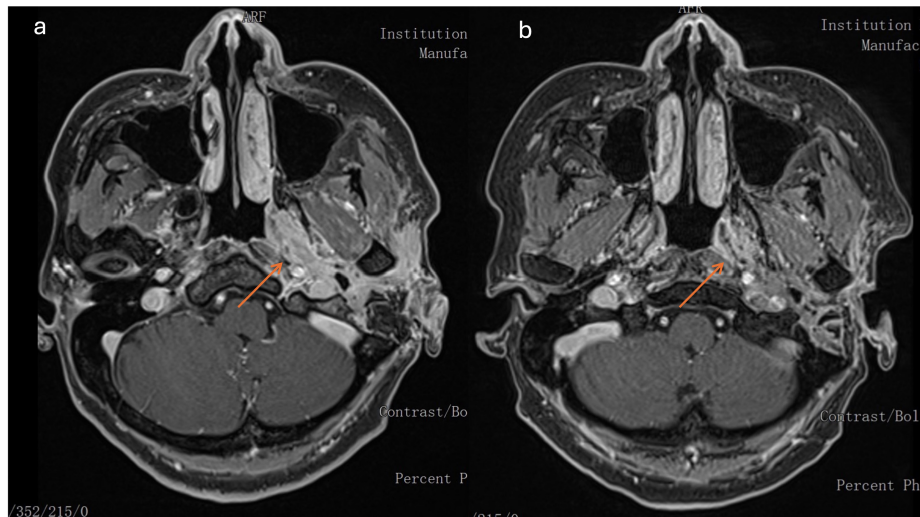
The patient underwent 30 RT sessions over 6 weeks with 2 Gy per day up to 60 Gy using the VMAT IMRT technique.

RT was completed in 6 weeks and the tolerance was good. The patient reported grade (G) 2 radiodermatitis, G2 xerostomia, and G2 mucositis (RTOG Radiation Therapy Oncology Group [RTOG] scoring) on the left cheek mucosa and homolateral lingual edge.



**Figure 7.** Immunostaining for light chains and polyclonal plasma cells and IgG4+/IgG+ ratio <40%.





**Figure 8.** (a) Axial contrast-enhanced T1 magnetic resonance imaging (MRI) (third month of follow-up) showing a large area of post-gadolinium enhancement along the skull base, with infiltration of the parotid gland, the temporomandibular joint, reaching the left parapharyngeal space till the nasopharynx, with involvement of the internal carotid artery and the internal jugular vein (orange arrow). (b) Axial contrast-enhanced T1 MRI image (1-year follow-up) showing the parapharyngeal space less contrasted, in contact with the external and internal carotid artery in its extracranial part, involving the carotid space (orange arrow).

RT was performed with concomitant corticosteroid therapy (methylprednisolone) at an initial dose of 96 mg a day, which was tapered each week to try to reduce the side effects.

At the end of RT (June 2022), a follow-up MRI (Figure 8B) showed a significant reduction of the lesion.

In consideration of the clinical and radiological improvement, it was decided to progressively decrease the corticosteroid therapy to 2 mg daily for 6 months.

This led to a resolution of the iatrogenic diabetes, while the radiological images appeared stable (December 2022), so the therapy was suspended.

At the last follow-up visit (August 2023), the patient appeared in excellent health and pain-free, without any medical therapy.

The PET examination performed more than 1 year after the end of radiotherapy showed the absence of areas of pathological accumulation of the radiopharmaceutical and therefore an absence of hypermetabolic pathology in the temporal bone and parapharyngeal space.

## DISCUSSION

Inflammatory pseudotumor is a rare non-cancerous process that mimics malignant processes and is found in almost all organs and has been found in almost all organ systems.<sup>3</sup> But in the differential diagnosis of this pathology, osteomyelitis of the skull base should also be considered. A mass of tissue of this type in the EAC can often be observed in patients with cholesteatoma and can lead to widespread inflammation and osteomyelitis in an immunocompromised patient. In this case, secondary cholesteatoma was also found during surgery.

Radiographically, thickening and enhancement of the dura are non-specific reactive findings that may also appear in cases of malignant

tumors. Nonetheless, preservation of the inner ear structures argues against an aggressive neoplasm and more likely indicates a chronic inflammatory process.<sup>4</sup>

After a review of the literature on inflammatory pseudotumors, no real guidelines were found on the attack therapy of this pathology, which does not frequently involve the temporal bone and parapharyngeal space.

First-line treatment must always be surgical if possible, and corticosteroid therapy must always be carried out in high doses, controlling any side effects that may arise until the symptoms have completely improved.

Finally, radiotherapy can stabilize an unresectable disease<sup>5</sup> and any residual neoformations evident on MRI after RT can be consequently metabolically inactive.

This second factor can therefore be regarded as a relevant prognostic index.

**Data Availability Statement:** The data that support the findings of this study are available on request from the corresponding author.

**Ethics Committee Approval:** N/A.

**Informed Consent:** Verbal informed consent was obtained from the patient who agreed to take part in the study.

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

**Author Contributions:** Concept – F.D.; Supervision – M.F.; Materials – C.M.; Data Collection and/or Processing – F.D., M.F., J.W.; Analysis and/or Interpretation – M.T.; Literature Search – C.K.; Writing – F.D., M.F.; Critical Review – M.F.

**Declaration of Interests:** The authors have no conflicts of interest to declare.

**Funding:** The authors declare that this study received no financial support.

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