

Osteosarcoma of the skull base presenting as a petrocavernous pseudoaneurysm and masquerading as an intracranial abscess: illustrative case

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BACKGROUND Telangiectatic osteosarcoma (TOS) is a rare and aggressive high-grade malignant neoplasm composed of blood-filled or empty cystic spaces resembling aneurysmal bone cysts. Uncommonly, TOSs can occur in the skull base.

OBSERVATIONS The authors present a case of a TOS that presented as a petrocavernous carotid pseudoaneurysm and then masqueraded as an intracranial abscess. The prognosis for TOSs with intracranial involvement is typically unfavorable and inversely related to the degree of intracranial involvement.

LESSONS Skull-based malignancies should be part of the differential diagnosis for a rapidly progressing lesion. Recovery of polymicrobial organisms during endoscopic sinus surgery should prompt reconsideration of the differential diagnosis. Postinflammatory changes from endovascular coiling have been described and can confound imaging and clinical findings.

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KEYWORDS osteosarcomas; pseudoaneurysm; intracranial abscess

This study describes a challenging clinical case in which the diagnosis was ultimately obscured by multiple clinical factors. This case illustrates the importance of maintaining a broad differential diagnosis in the setting of confounding clinical findings.

Illustrative Case

A 65-year-old male presented with a 2-month history of headaches, diplopia, and pain behind the left eye. His clinical examination was significant only for a left sixth cranial nerve palsy. Multiple imaging studies showed a large unruptured left petrocavernous segment internal carotid artery pseudoaneurysm (Fig. 1). Initial magnetic resonance imaging (MRI) did not raise any concerns for a surrounding mass. His symptoms were thought to be from an aneurysmal mass effect. He underwent balloon test occlusion and left internal carotid endovascular sacrifice for the treatment of the large pseudoaneurysm (Fig. 2). He had complete resolution of his

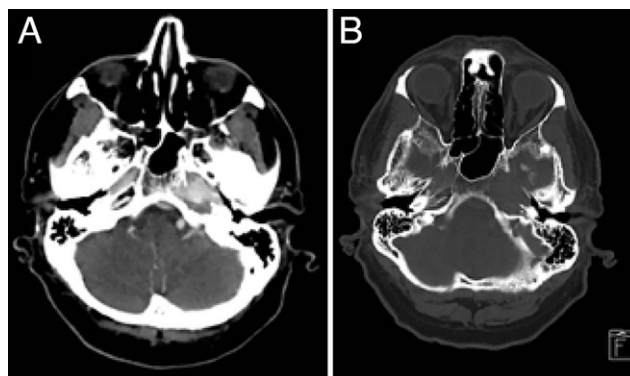


FIG. 1. A: Initial computed tomographic angiography demonstrating aneurysm. **B:** Bone kernel of (A) showing aggressive lytic change of the bony clivus (ill-defined margin).

ABBREVIATIONS MRI = magnetic resonance imaging; TOS = telangiectatic osteosarcoma.

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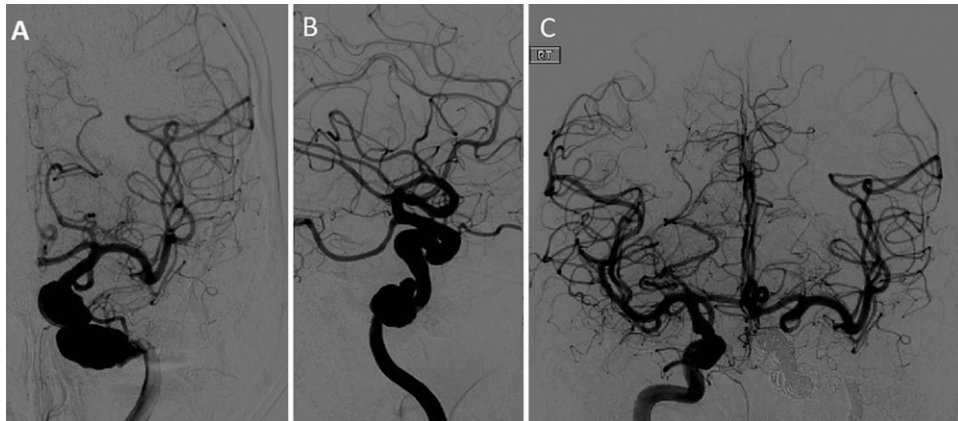


FIG. 2. A: Pretreatment left internal carotid artery (ICA) anteroposterior (AP) angiogram demonstrating left petrous pseudoaneurysm with concomitant cavernous segment aneurysmal dilation. **B:** Pretreatment left lateral ICA angiogram demonstrating left petrous pseudoaneurysm with concomitant cavernous segment aneurysmal dilation. **C:** Post-treatment right ICA anteroposterior angiogram demonstrating parent vessel sacrifice of left ICA with robust and synchronous filling of the hemispheres.

symptoms for 3 weeks. Recurrence of his symptoms prompted readmission. MRI/magnetic resonance angiography of his brain showed a large mass of heterogeneous tissue with nodular peripheral enhancement centered on the coil mass and involving the left cavernous sinus, left orbital apex, and left fifth cranial nerve (Fig. 3). Due to concern for a potential infectious process of the coil mass, bilateral ethmoidectomies and sphenoidotomies were performed to sample tissue for cultures. Intraoperative cultures grew *Klebsiella oxytoca*, *Cutibacterium acnes*, and *Corynebacterium* species. He was discharged to receive 6 weeks of metronidazole and ceftriaxone. When his symptoms continued to worsen over the next week, he was readmitted, and his antimicrobials were broadened. A postendovascular coiling inflammatory process was considered in the differential diagnosis. He continued to have rapid worsening of his symptoms, including complete ophthalmoplegia and loss of pupillary light reflex. At this point, an invasive fungal infection was considered, and liposomal amphotericin B was added. Due to these infectious concerns, he underwent an extended endonasal debridement with coil mass evacuation after surgical trapping of the carotid artery (Fig. 4). Intraoperatively during coil removal, the coils were noted to have shifted with the mass growth, and no vessel wall was

appreciated. The finding in tissue from this biopsy that was sent for broad-range polymerase chain reaction for infectious organisms (including bacterial, fungal, and mycobacterial targets) was negative. Histopathology demonstrated giant cells (Fig. 5). Follow-up bacterial cultures of this second debridement were without growth. The final pathology report demonstrated malignant telangiectatic osteosarcoma (TOS) of the left petrous bone and clivus. The patient died shortly after being evaluated for salvage chemotherapy.

Discussion

Observations

This case demonstrates the challenge in diagnosis of skull base malignancies as well as the difficulty that confounding clinical factors can present in assessing these patients. In this particular case, a unique challenge was weighing the likelihood of postoperative infection versus inflammatory changes originating from his aneurysm coiling procedure.

TOSs make up 2%–12% of all osteosarcomas.¹ Typically, TOSs develop in younger populations (15–20 years of age) with a 2:1 male predominance.² The exact cause of the disease and risk factors are relatively unknown, given its rarity.³ Some authors propose

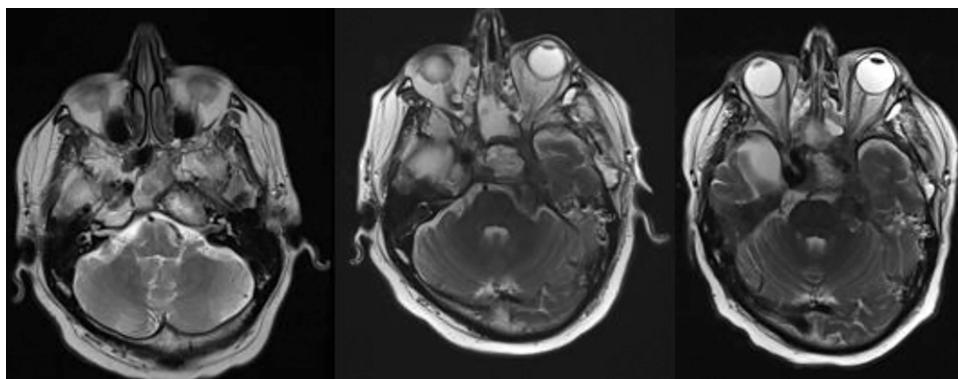


FIG. 3. Sequential axial T2-weighted images showing progression of the process with development of exophytic component initially into sphenoid sinus cavity and later extending intracranially into the cerebellopontine angle.

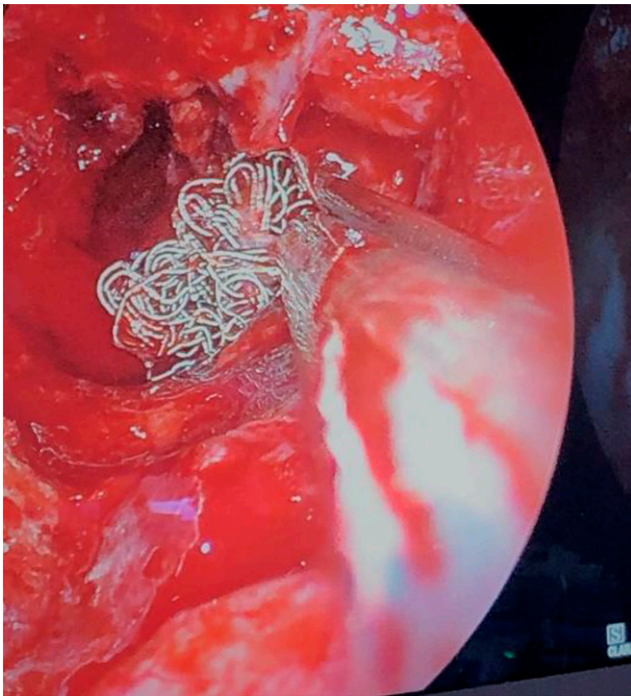


FIG. 4. Intraoperative photograph of endonasal evacuation of presumed infected coil mass from the previously sacrificed carotid artery.

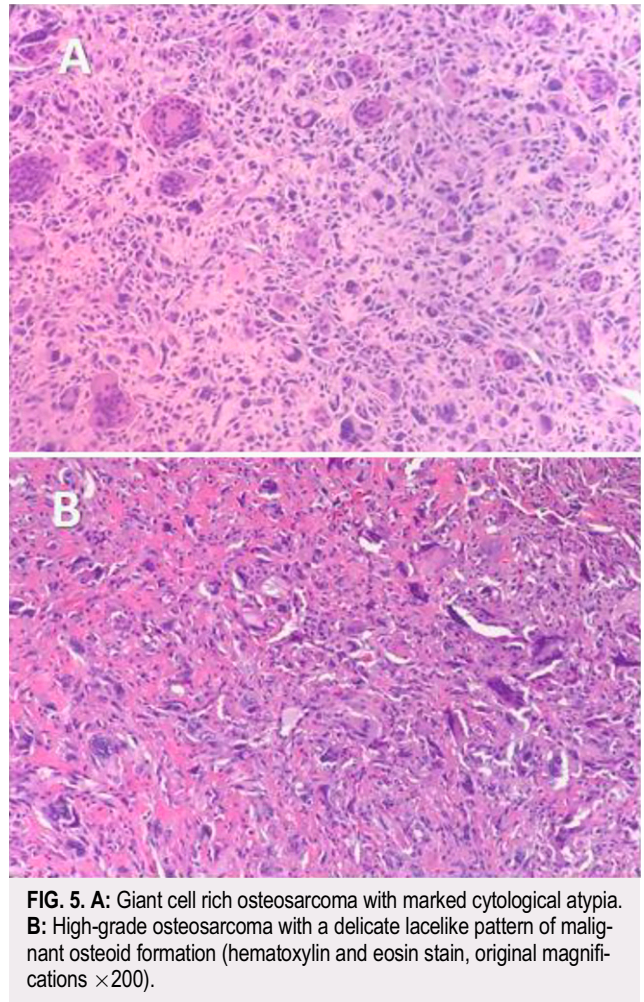


FIG. 5. A: Giant cell rich osteosarcoma with marked cytological atypia. **B:** High-grade osteosarcoma with a delicate lacelike pattern of malignant osteoid formation (hematoxylin and eosin stain, original magnifications $\times 200$).

the possibility of genetic predisposition in rare instances of multiple family members having the disease.³ These tumors typically present in the distal femoral metaphysis (42%), followed by the proximal tibia (17%), proximal humerus (9%), and proximal femur (8%).^{3,4} TOSs occur in skull bones in only 1.5% of cases.³ The most common symptoms are local pain, a soft tissue mass, or both. The best imaging modality is often plain radiographs, which typically demonstrate lytic destructive lesions and occasionally minimal intralésional sclerosis.

Histology of TOSs demonstrates osteoblast-like and fibroblast-like features at the ultrastructural level that are presumed to have derived from multipotent stem cells of mesenchymal origin. Macroscopic histology of resected specimens reveals the classic description as “a bag of blood,” given that they are composed of 90% cystic components before treatment.¹ The septa generally consist of pleomorphic cells and osteoclast-like giant cells.² The lesions are largely hemorrhagic.⁴ Treatment for TOSs usually consists of neoadjuvant chemotherapy typically consisting of at least two of the following: methotrexate, ifosfamide, cisplatin, carboplatin, and doxorubicin. This is followed by resection. TOS 5-year survival rates are generally 65% with contemporary chemotherapy protocols.² However, the prognosis for TOSs with intracranial involvement is typically unfavorable and inversely related to the degree of intracranial involvement.²

Lessons

Our case illustrates the importance of early recognition and rapid progression of this malignancy and the difficulty in doing so with confounding clinical clues. TOSs are commonly misdiagnosed as aneurysmal bone cysts.² In this case, the location of the presentation, the proximity to a giant pseudoaneurysm, and the initial bacterial cultures all confounded the final diagnosis.^{5–7} Specifically, the

confounding question was whether the mass was a postsurgical abscess or a local reaction to the endovascular coiling procedure. With the knowledge provided by the ultimate diagnosis, our patient’s intraoperative cultures are best explained by the microbiology of the nasal cavity.^{7,8} Published case reports have documented a variety of radiographic findings thought to be related to reactive processes after endovascular coiling, which in this case made the patient’s ultimate diagnosis challenging.⁹

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Kiley, Morton. Acquisition of data: Morton. Analysis and interpretation of data: Kiley, Bakken. Drafting the article: Kiley, Washington. Critically revising the article: Kiley, Morton. Reviewed submitted version of manuscript: Kiley, Morton. Approved the final version of the manuscript on behalf of all authors: Kiley. Administrative/technical/material support: Kiley, Morton.

Supplemental Information

Previous Presentations

This work was previously presented in abstract form at the San Antonio Uniformed Services Health Education Consortium Research Day.

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