Phosphaturic Mesenchymal Tumor of the Olfactory Groove

Rita Snyder, MD[1]; Leomar Y. Ballester, MD, PhD[1]; Sujit Prabhu, MD[1]
[1]University of Texas, MD Anderson Cancer Center

Abstract

Background: Phosphaturic mesenchymal tumors (PMT) are rare, typically benign neoplasms which most frequently arise in the extremities.[1,2] However, they may also occur intracranially and mimic other extra-axial neoplasms.

Objective: To improve recognition of a rare etiology of skull base neoplasia and review subsequent treatment considerations.

Methods: Case report and review of the literature.

Results: Our review of the literature returned 9 published cases to date of PMT arising from the anterior cranial fossa. Complete cure was obtained with gross total resection. Postoperative Dotatate-PET was helpful in confirming no residual tumor, and normalized laboratory results supported these findings.

Conclusion: PMT are rare tumors which may mimic skull base meningiomas. Surgical resection is curative. Residual may be detected on specialized imaging and managed medically if necessary.

Introduction

Phosphaturic mesenchymal tumors (PMT) are rare, typically benign neoplasms which most frequently arise in the extremities.[1,2] Patients often present with osteomalacia due to decreased renal reabsorption of phosphate and downregulation of 25-hydroxyvitamin D3-1a-hydroxylase caused by Fibroblast growth factor 23 (FGF-23) excreted by the tumor.[2] Serum and urine studies may help confirm the diagnosis. Considering this etiology when patients present with a history of unexplained recurrent fractures in the presence of an extra-axial skull base lesion may influence treatment planning as complete resection is curative.[2] Here we discuss a case of PMT mimicking an olfactory groove meningioma and subsequent clinical management.

Methods and Materials

A review of the literature was performed. The PUBMED database was searched for articles with the keywords: "phosphaturic mesenchymal tumor" AND "skull base". We also included a case study of a patient treated at our institution.

Results

A total of 18 studies were returned, 5 of which were review articles. Of the 37 published cases in the literature to date, only 9 were found isolated to the anterior cranial fossa.[2]

Regarding our case, a 59-year-old woman was referred to endocrinology after presenting with multiple spontaneous fractures. FGF-23 level was elevated, prompting work-up for an underlying malignancy. Serum Vitamin D and phosphorus were deficient; however, bone density scans of the spine and forearm were normal. A right frontal extra-axial lesion arising from the olfactory groove was discovered on MRI brain (**Figure 1**). The patient was otherwise asymptomatic. She underwent uneventful resection of this lesion, and her phosphate levels normalized within one month postoperatively. Intraoperatively and histopathologically the lesion was locally invasive (**Figure 2**). Dotatate-PET confirmed complete resection (**Figure 3**).

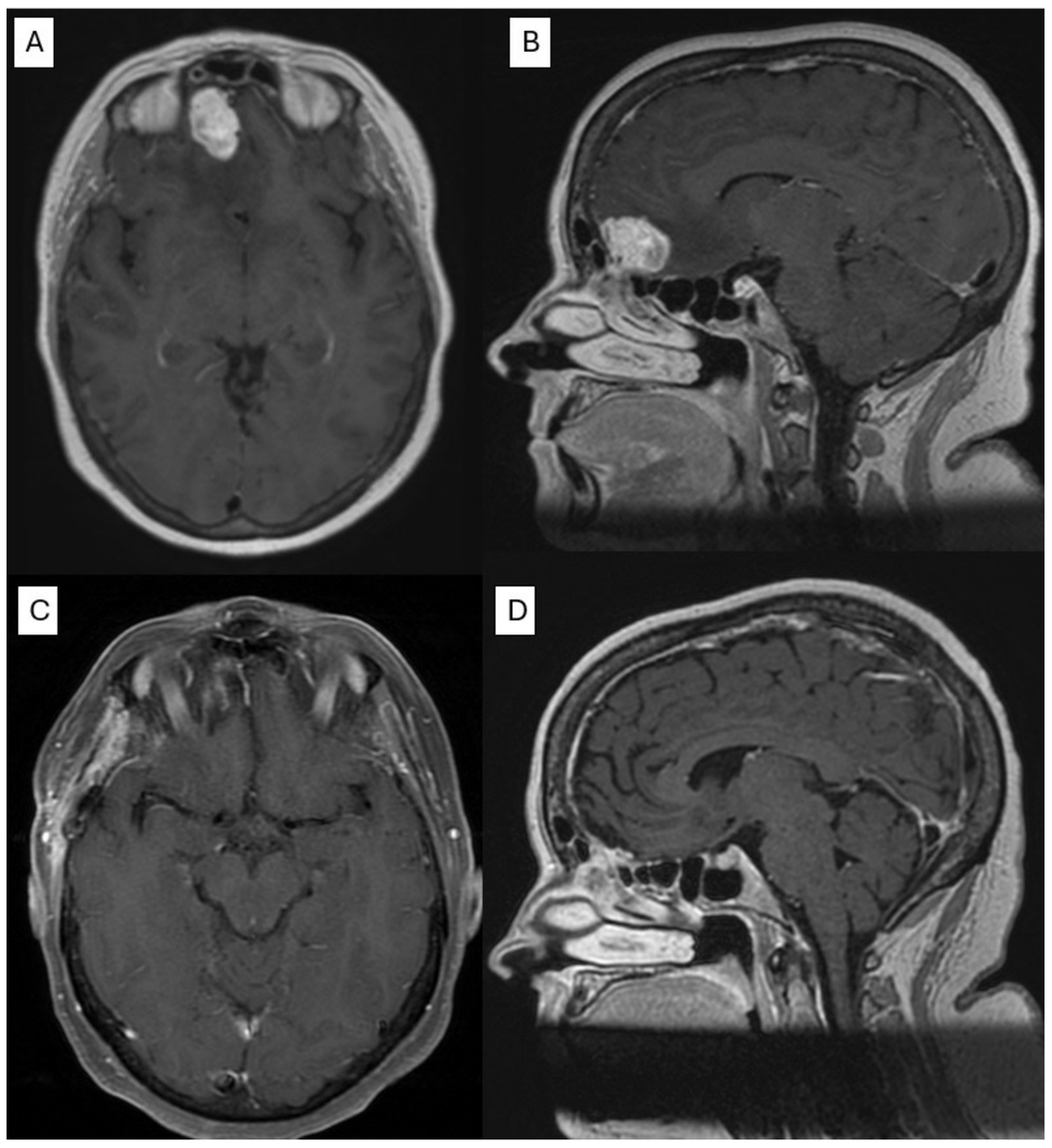

Figure 1. Preoperative (A&B) and postoperative (C&D) contrasted MRI scans of a PMT mimicking an olfactory groove meningioma.

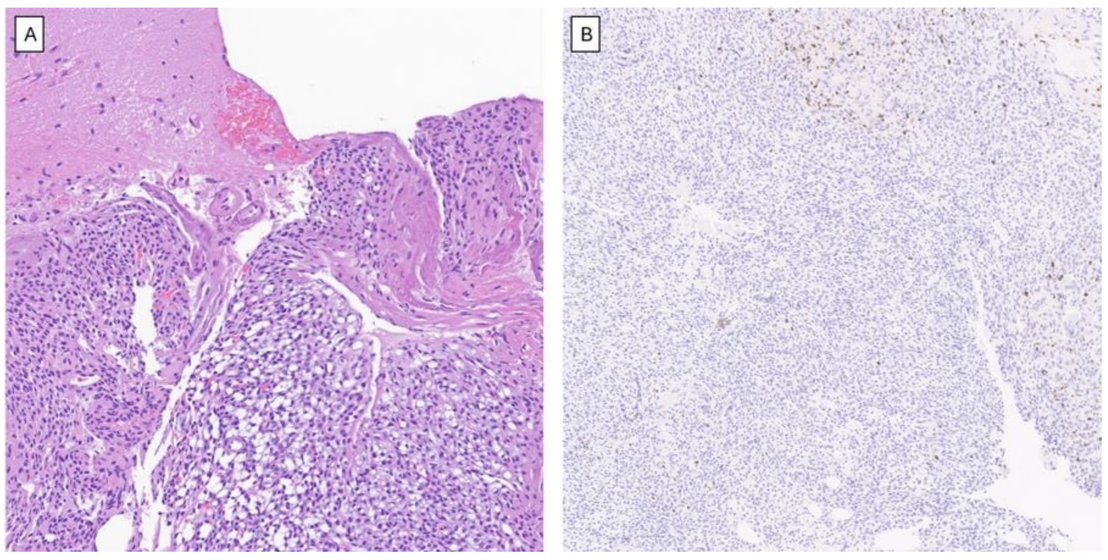

Figure 2. Histopathology slides of resected PMT demonstrating A) bland spindle cells, a vascular rich network, and alternating mature adipocytic areas on H&E stain and B) low field Ki67 antigen (MIB1) labelling index of 7.68% (1,003 nuclei counted), with an average index of 5.17%.

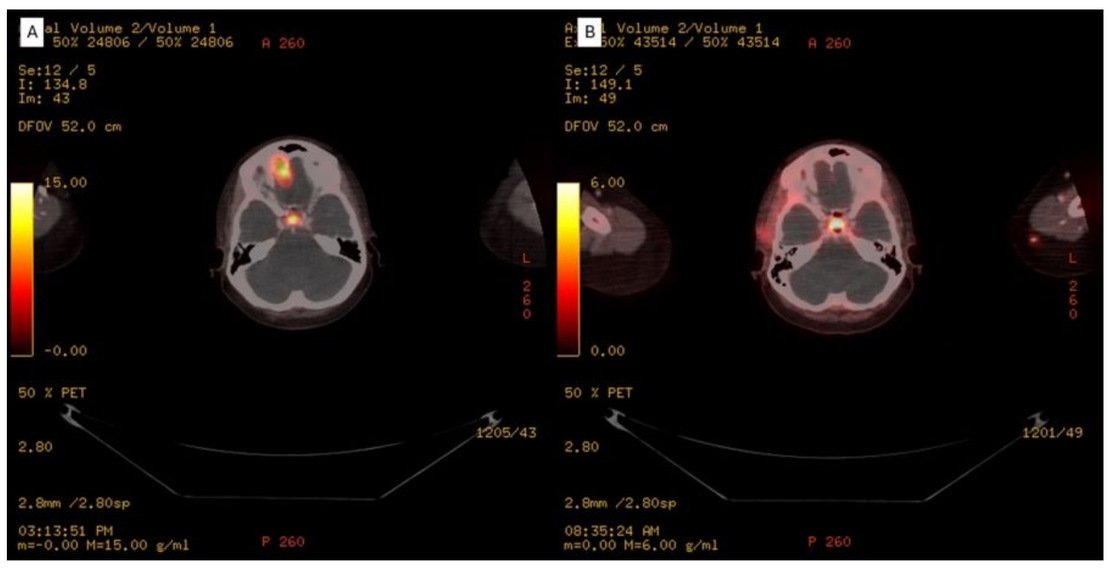

Figure 3. A) Preoperative and B) postoperative Dotatate-PET study demonstrating complete resection of the highly avid PMT.

Conclusions

Skull base phosphaturic mesenchymal tumors are a rare etiology in which patients typically present with osteomalacia. They may mimic meningioma on both MRI and PET imaging, therefore serum and urine phosphate levels are helpful for further characterization. Gross total resection is curative. Confirmation of complete resection may be obtained with a postoperative Dotatate-PET scan.

Contact

Rita Snyder, MD
1515 Holcombe Blvd.
Houston, TX 77030
rsnyder1@mdanderson.org

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