

## Neuroanatomical Studies

# Malignant primary intraosseous meningioma in a pediatric patient: A case report and review



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## ABSTRACT

**Introduction:** This is the first case of a malignant, pediatric PIOM reported in the United States.

Primary extradural meningiomas (PEM) arise in locations other than dura, outside the intracranial compartment. Primary Intraosseous Meningiomas (PIOMs) are a rare subset of PEM that arise in bone.

**Case report:** A 16 year old female, neurologically intact, mildly tender swelling over her right forehead that increased in size, accompanied by headaches and dizziness. Imaging demonstrated a lesion infiltrating the frontal calvarium and the adjacent intracranial and extracranial soft tissues. Surgical resection was performed. Pathology supported the diagnosis of anaplastic meningioma (WHO grade III).

**Discussion:** PIOMs are a rare subset arising in bone. In an extensive review, less than 150 cases of PIOMs were found. It likely originates from entrapment of arachnoid cap cells, congenitally or after development. Treatment is similar to intradural meningiomas, namely a potentially curative resection. Resection and cranioplasty should be performed simultaneously. Adjuvant radiation, chemotherapy and bisphosphonates are recommended.

## 1. Introduction

Most meningiomas are considered primary intradural lesions and are located in the subdural space. In contrast, primary extradural meningiomas (PEM) arise in locations other than the dura, outside the intracranial compartment [16,24]. In a comprehensive study in 2000 at MD Anderson, 1.6% of meningiomas were PEMs, and only 142 patients with PEM were found in a literature search from 1976 to 2000 [24]. They are further divided into primary intraosseous meningiomas (PIOMs), which describes those that arise in bone [31]. In the vast majority of PIOMs the bone of origin arises in cranial bones, with a few reported to have originated in the mandible [26].

This is the first case of a malignant, pediatric PIOM reported in the United States [2,3,6,8,18,20,22,38,42].

## 2. Case report

### 2.1. History

A 16 year old, 50 kg, white female noticed a swelling over her right forehead. She had bumped her head several times getting into and out of her car. The bump resolved but soon began to increase in size and was accompanied by headaches several times per week for the next 2 months. She experienced dizziness for 3 days. This prompted a visit to her primary care physician. A CT of her head demonstrated an extra-calvarial lesion (Fig. 1). She was referred to neurosurgery for evaluation.

The patient had no significant medical history. She had an unremarkable neurological exam. She had a well-defined, mildly tender,

**Abbreviations:** PEM, Primary Extradural Meningioma (PEM); PIOM, Primary Intraosseous Meningioma (PIOM); WHO, World Health Organization; CT, Computed Tomography; MRI, Magnetic Resonance Imaging; HU, Hounsfield Unit; ECA, External Carotid Artery; DWI, Diffusion Weighted Imaging; ADC, Apparent Diffusion Coefficient; FLAIR, Fluid-attenuated Inversion Recovery; H&E, Hematoxylin & Eosin

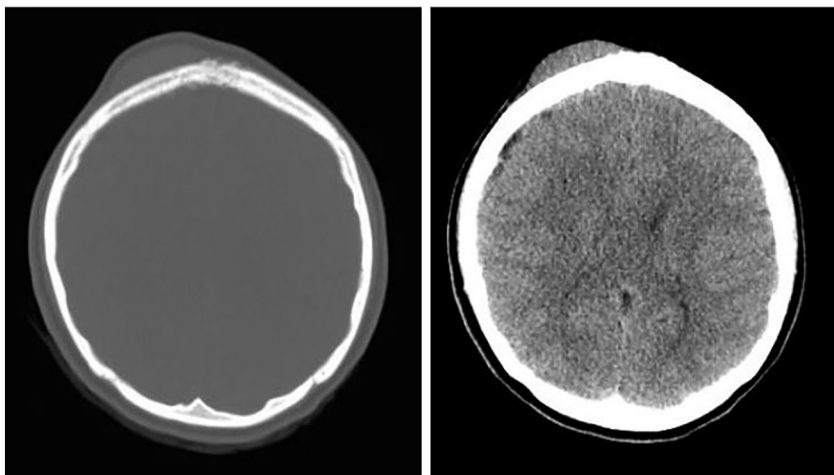
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**Fig. 1.** Non-contrast Head CT. *Left:* Bone filter images demonstrate an ill-defined osseous lesion involving the diploic space, internal and external table of the right frontal calvarium with permeative pattern of osseous destruction and irregular cortical osseous and subperiosteal remodeling. *Right:* Soft tissue filter images demonstrate crescent shaped extra-cranial and intracranial soft tissue components at the level of the right frontal calvarial lesion.

somewhat fluctuant mass overlying her right forehead. The remaining physical exam was benign. We elected to observe the lesion without immediate plans for resection. The mass continued to grow over the next 2 weeks and an MRI was obtained.

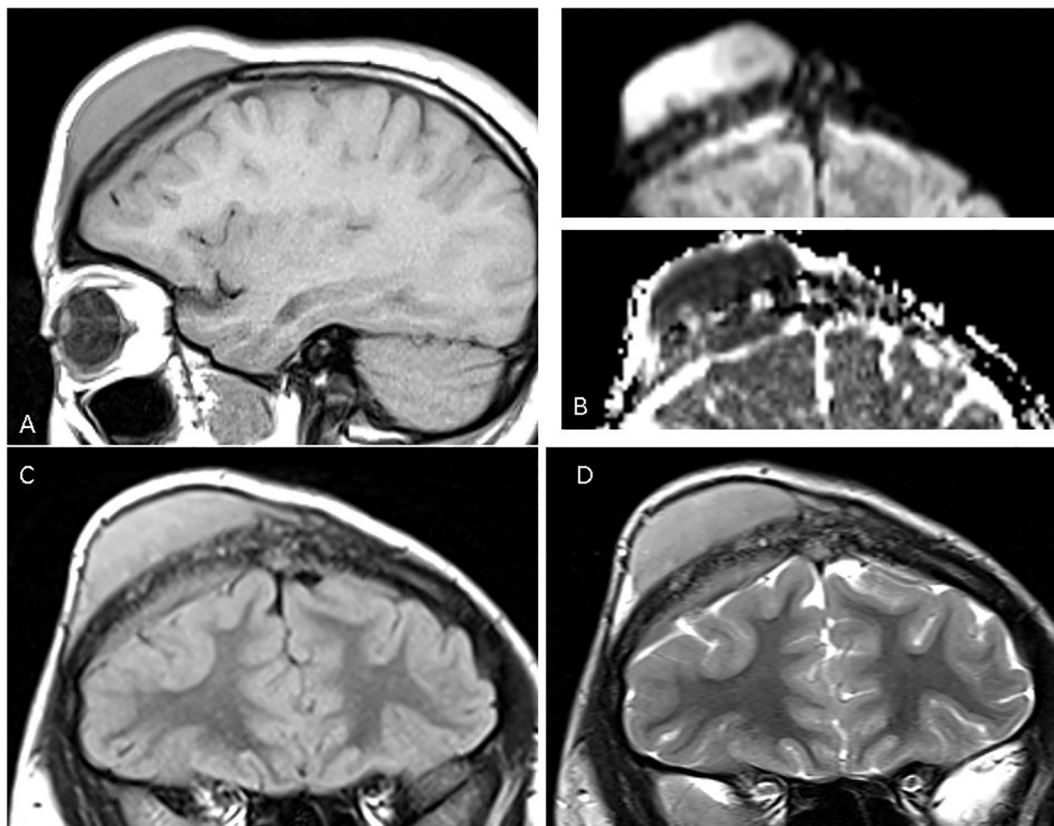
MRI showed an infiltrating lesion involving the frontal calvarium and the adjacent intracranial and extracranial soft tissues. Diffusion weighted imaging demonstrated restricted diffusion with diffuse high DWI and low ADC signal in the lesion.

The brain and subdural space appeared unaffected (Fig. 2). Differential diagnosis at this time was sarcoma, histiocytosis, primary bone lymphoma, infection or metastasis. She was offered surgical resection

and exploration via a right craniotomy for removal the mass and possible infection.

*2.2. Surgical procedure*

General anesthesia was induced and was prepped and draped in a sterile fashion. A semi-curvilinear incision was made above the hairline and dissected into the subgaleal space, maintaining the capsule of the mass. The mass was sharply incised and was found to be a solid tumor. The periphery of the tumor was further dissected and the associated periosteum was incised circumferentially down to bone. The extra-cranial portion of the tumor was freed from underlying bone. The tumor



**Fig. 2.** T1 sagittal (a), Diffusion DWI/ADC axial (b), FLAIR coronal (c), and T2 coronal (d) MRI: Frontal calvarial lesion, isointense to grey matter on all sequences with restricted diffusion, measuring 1.4 × 4.5 × 5.6 cm with bone marrow replacement, cortical destruction of the inner and outer table and intracranial and extracranial soft tissue component with moderate dural thickening.

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