

Case Report

“Sun-ray” periosteal reaction in the calvarium – uncommon presentation of a common disease

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ABSTRACT

Meningiomas usually present with vague neurological symptoms or are incidentally detected on imaging. Radiologists are aware of the classical imaging picture of this entity comprising of enhancing extra-axial mass with associated hyperostosis. However, the presence of an aggressive sunburst type of periosteal reaction in a primary intra-osseous osteolytic meningioma, along with an extra-axial soft tissue as seen in our case has not been described previously in the literature.

Keywords: Atypical meningioma, Calvarial meningioma, Intraosseous osteolytic meningioma, Sunburst periosteal reaction, Sunray periosteal reaction

INTRODUCTION

Meningiomas constitute nearly a third of all brain neoplasms presenting most commonly in the fourth–fifth decade of life. They are often asymptomatic masses, however, they may present with vague symptoms. In majority of the cases, imaging shows enhancing extra-axial mass with associated features such as dural tail and bony hyperostosis which clinch the diagnosis and are typical of a classical meningioma.^[1] However, atypical radiological features rarely complicate the imaging picture. Primary intraosseous osteolytic meningioma is a rare subtype constituting <2% of cases of meningiomas and is extradural arising within the bone thus, often being confused with primary bone tumors. These extradural meningiomas are more likely to display atypical imaging features and aggressive pathology.

CASE REPORT

A 55-year-old lady of African ethnicity presented with headache and a gradually progressive left scalp swelling for 4 months. No antecedent history of trauma or fever was present and the patient was well preserved otherwise.

On examination, a large firm-to-hard swelling was present over the left parietal region close to the vertex. The overlying skin and scalp hair were intact without any evidence of ulceration or local skin changes.

Skull radiograph showed areas of moth-eaten lytic destruction of the left parietal bone with associated sunburst type of periosteal reaction and the presence of a large soft-tissue component [Figure 1]. Contrast-enhanced magnetic resonance imaging (MRI) of the brain corroborated

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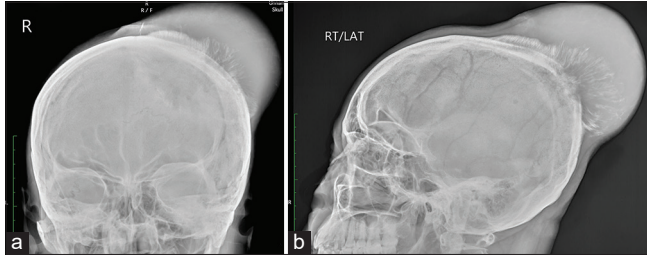


Figure 1: (a) Xray skull AP and (b) lateral shows areas of moth-eaten lytic destruction of left parietal bone with sunburst type of periosteal reaction. A large soft tissue component is seen overlying these bony changes without any foci of calcification or ossification.

the findings of the osteolytic lesion and extensive spiculated sunburst type of periosteal reaction with new bone formation along the extracranial aspect of the lesion. The mass appeared isointense on T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI) with strong diffusion restriction on diffusion-weighted images with an apparent diffusion coefficient value of $0.93 \times 10^{-3} \text{ mm}^2/\text{s}$. The periosteal reaction was particularly well seen on susceptibility-weighted images. In addition, a large, avidly enhancing extraosseous soft-tissue solid mass was present on both sides of the skull. The intracranial component measured $8.6 \times 6.5 \text{ cm}$ and showed a significant mass effect with gliosis and edema in the underlying left parietal parenchyma [Figure 2]. Differentials considered were calvarial intraosseous hemangioma, osteosarcoma, and osteoblastic metastasis. Gross primary resection of the tumor was done by a left parietal craniotomy with negative margins and the post-operative computed tomography (CT) scan revealed no residual mass. The patient did not require any adjuvant therapy and is doing well in 6-month follow-up.

Histopathology of the mass showed proliferation of spindle cells with syncytial and whorling patterns at places with few psammoma bodies with areas of increased cellularity. On immunohistochemistry, strong patchy cells were positive for EMA and PR and negative for GFAP, STAT6, and S100. Ki67 proliferative index is 20–25% in hot spot areas. The final diagnosis favored an atypical meningioma [Figure 3].

DISCUSSION

Majority of meningiomas arise from the arachnoid cap cells of the dura mater and show classical imaging features, and bony changes such as hyperostosis or direct invasion of the bone are described in atypical and higher-grade tumors. Primary intraosseous meningiomas are a subtype of primary extradural meningiomas and are rare in occurrence. Even among these, only a small number of cases show both aggressive imaging features and atypical pathological features. The various bony changes described constitute osteolysis and sclerosis. However, the spiculated sunburst type of periosteal reaction seen in our case

is atypical for a meningioma. To the best of our knowledge, this is the first report of such a pattern of bony reaction in meningiomas. Due to such an extensive aggressive type of periosteal reaction, the imaging differentials considered were intraosseous hemangioma and bone-forming tumors such as metastasis, osteosarcoma, and more than a meningioma [Table 1].

Calvarial hemangiomas are characteristically intradiploic expansile lesions with thin sclerotic rims. They show a sunburst type of trabecular thickening that radiates from a central point. Often, there may be areas of cortical breach with bony trabeculae growing beyond the cortex simulating an aggressive tumor.^[2-4]

Calvarial osteosarcoma is a rare entity comprising <1% of total osteosarcoma cases. Few case reports describe this entity, which on imaging appears as a calvarial-based soft-tissue lesion with external and internal components. New bone formation and sunburst type of aggressive periosteal reaction are seen.

However, one should be vary to not confuse this “sunburst periosteal reaction” with the sunburst sign described in T2-weighted or post-contrast MRI or angiography. In this, the dural vessels are seen to diverge radially into the meningioma when viewed from the side.^[5-8]

These primary intraosseous meningiomas most commonly present as a gradually increasing scalp mass. On imaging, they are bone based rather than being “dural based” as described for classical meningiomas.^[9] There is bony erosion with areas of osteolysis and the presence of scalp and intracranial soft-tissue components. Dural and soft-tissue invasion may be coexisting. They typically appear as avidly enhancing masses having isointense signals on T1WI and T2WI. ⁶⁸Ga DOTATEC positron emission tomography/CT has been used for diagnosis, radiotherapy response assessment, and detection of recurrence of extradural meningiomas.^[10,11]

The management of choice is gross tumor excision with or without adjuvant radiotherapy.

This case emphasizes that even in the presence of a sunray type of periosteal reaction in the calvarium, meningioma should be kept as one of the differential diagnoses.

Table 1: Differential diagnosis of sunray periosteal reaction in calvarium.

Calvarial hemangioma	Intradiploic expansile lesions, thin sclerotic rim, sunburst type of trabecular thickening
Meningioma	Extradural mass with bony hyperostosis, rarely aggressive type of periosteal reaction
Osteosarcoma	<1% of total osteosarcomas. Soft-tissue mass present. New bone formation and aggressive periosteal reaction
Metastasis	Known primary, multiplicity of lesions

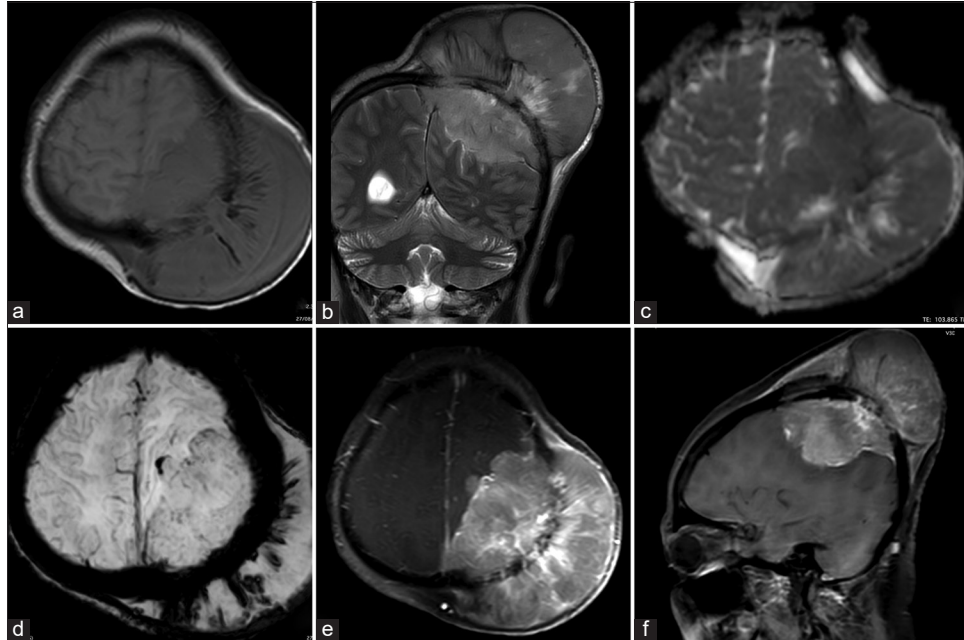


Figure 2: Magnetic resonance imaging (MRI) shows an area of marrow signal alteration in the left parietal bone with a large extraosseous soft-tissue mass having an extradural component. The patient had presented with the large scalp component of the mass. (a) The mass is isointense on T1WI, (b) hyperintense on T2WI and (c) shows diffusion restriction. Extensive spiculated sunburst type of periosteal reaction with new bone formation is seen along the extracranial aspect of the lesion. (d) Susceptibility weighted imaging (SWI) images shows the periosteal reaction very well. (e, f) On post contrast intense heterogeneous enhancement of the lesion is seen.

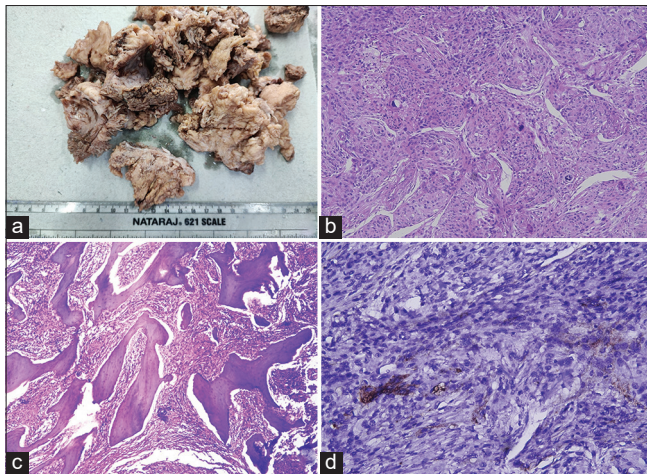


Figure 3: (a) Grossly the tumor was tan-grey and predominantly solid. (b) Sections (200x) show proliferation of spindle cells with syncytial and whorling pattern at places. Few psammoma bodies seen. 4 to 5 mitotic figures noted /10 high power fields. Focal patchy areas of increased cellularity, small cells with high nuclear-cytoplasmic (N/C) ratio, large and prominent nucleoli, patternless or sheet-like growth (loss of lobular architecture) and foci of spontaneous or geographic necrosis noted. (c) Few bone bits noted with adhered and entrapped tumor tissue (100x). (d) On immunohistochemistry (200x), strong patchy cells were positive for EMA and PR, negative for GFAP, STAT6, S100. Ki67 proliferative index is 20-25% in hot spot areas. The final histopathological diagnosis favoured an atypical meningioma (WHO grade 2).

CONCLUSION

Meningiomas are the most common brain tumors and have typical imaging features. However atypical features like sunray type of periosteal reaction and large extraaxial soft tissue components can also be seen in some cases and meningioma should be a consideration if such features are seen.

TEACHING POINTS

1. Even in the presence of a sunray type of periosteal reaction in the calvarium, meningioma should be kept as one of the differential diagnoses.
2. Other rarer differential diagnosis of the sunray type of periosteal reaction in the calvarium includes hemangioma, osteosarcoma, and metastasis.

MCQs

1. What are the types of bony changes in meningiomas? (multiple options can be correct)
 - a. Hyperostosis
 - b. Osteolysis
 - c. Sunray type of periosteal reaction
 - d. None of the above

Answer key: a, b, c

2. Which nuclear medicine study is recommended for detection of extradural meningioma?
 - a. ⁶⁸Ga DOTATOC PET/CT
 - b. Sestamibi scan
 - c. Bone scan
 - d. None of the above

Answer key: a

3. What is the treatment of choice for intraosseous meningiomas?
 - a. Gross tumor resection
 - b. Chemotherapy
 - c. Conservative
 - d. Radiotherapy only

Answer key: a

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