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CASE REPORT

Pure Primary Intraosseous Meningioma: A Case Report And Review Of Literature

SUSHILA JAISWAL ,AWADESH KUMAR JAISWAL*, SANJAY BEHARI *, RAKESH PANDEY

ABSTRACT

A 60 year old lady presented with progressive increasing swelling over the scalp for 1 year and mild headache for 2 months. Examination revealed a non tender bony hard mass over the left parietal region. CT scan showed localized hyperostosis and thickening of the left parietal bone, with normal underlying brain parenchyma. The mass was surgically excised. Histopathology was suggestive of meningothelial meningioma. The case is discussed in the light of relevant literature.

Key words: Calvarial, extradural, intraosseous, meningioma

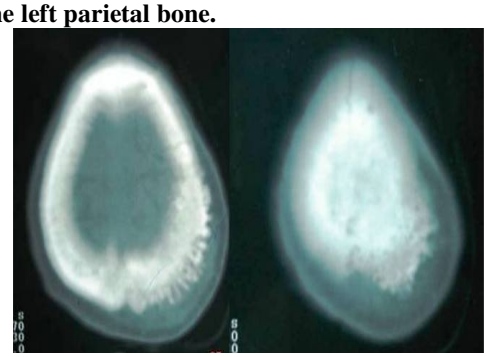
*Department of Pathology and Neurosurgery, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Raebareli Road, Lucknow, INDIA-226014.
Corresponding author:
Dr. Sushila Jaiswal, MD,
Assistant Professor, Department of Pathology, Sanjay Gandhi Postgraduate Institute of Medical Sciences, Rae Bareli Road, Lucknow 226014 UP, India
FAX: 91-522-2668078,2668017
Telephone: 91-522-2668700 extension 2107, 2741, Mobile: +919450904700
Email: sushilapath@yahoo.com

Primary intraosseous meningioma is a term which is used for extradural meningiomas that arises in the bones [1]. It accounts for approximately fewer than 2% of mening over all and two thirds of all extr meningiomas [2]. The authors present h case of rare primary calvarial meningioma and review of the relevant literature.

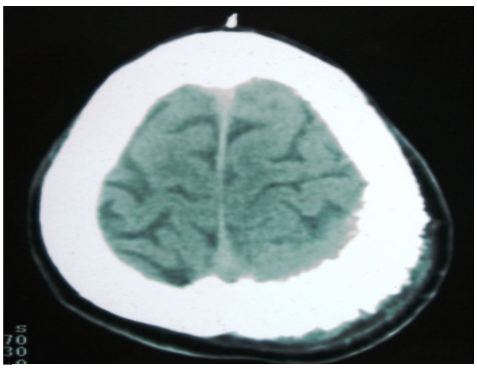
Case report

A 60 year old lady presented to us with the history of progressive increasing swelling over the scalp for 1 year and mild headache for 2 months. There was no history of seizure, severe headache or vomiting. Her neurological examination was unremarkable. Local exami revealed a non tender bony hard mas the left parietal region. The overlyin was freely mobile. CT scan showed localized hyperostosis and thickening of the left parietal bone, with normal underlying brain parenchyma ([Table/Fig 1] and [Table/Fig 2]).

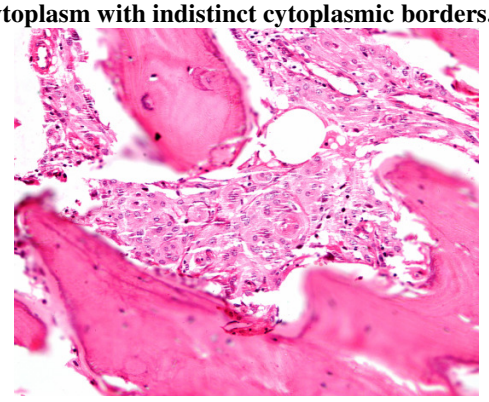
[Table/Fig 1] NCCT (head) bone window showing localized hyperostosis and thickening of the left parietal bone.



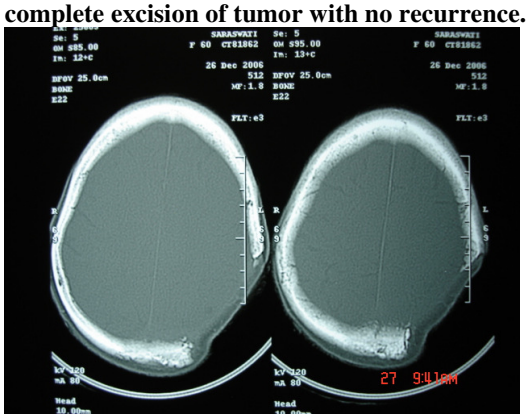
[Table/Fig 2] NCCT (Head) soft tissue window showing localized hyperostosis and thickening of the left parietal bone with normal underlying brain parenchyma.



[Table/Fig 3]: Photomicrograph showing bony tissue infiltrated by tumor composed of whorls of polygonal cells displaying mildly anisomorphic nuclei with conspicuous nucleoli in places and moderate amount of eosinophilic cytoplasm with indistinct cytoplasmic borders.



[Table/Fig 4] - Follow up NCCT (head) showing complete excision of tumor with no recurrence.



The patient was operated and excision of the involved bone was performed. Intra-operatively, the tumour was completely confined in the involved bone and was soft and vascular. It was neither attached with the underlying dura nor with the overlying scalp tissue. Histopathology showed fibrocollagenous and bony tissue which was infiltrated by a tumour and composed of sheets, nests and whorls of polygonal cells, displaying mildly anisomorphic nuclei with clumped chromatin, conspicuous nucleoli in places and moderate amount of eosinophilic cytoplasm with indistinct cytoplasmic borders. No glial tissue or features of malignancy were noted. These features were suggestive of intraosseous meningothelial meningioma [Table/Fig 3]. The patient was discharged on the 7th post operative day and at a follow up after 19 months, she was found to be symptom free and did not have any radiological recurrence [Table/Fig 4]. During follow up, she was advised cranioplasty, but she refused any further procedure.

Discussion

Meningiomas are of two types depending on the site of involvement. Primary intradural lesions which are located in the subdural, intracranial or intraspinal space and primary extradural meningiomas which arise in sites other than the subdura e.g. nasal cavity, paranasal sinuses, skin, orbit, calvaria [3],[4], neck [5],[6], parotid gland [7], mediastinum [8], lung [9] and even in the fingers [10]. These are also known as ectopic, secondary,

[Table/Fig 5]: Summary of differences in intradural and extradural meningiomas

Parameters	Intradural meningiomas	Extradural meningiomas
Location	Intradural	Extradural
Age	Later decades	Bimodal peaks, one in 2nd and another in peak during the fifth through seventh decades of life.
Sex	Twice as frequently in females as in males.	Approximately the same frequency in each sex

Numbers	Usually solitary	Uniformly solitary, rarely with two separate lesions.
Radiological features of adjacent bone	Usually osteoblastic if bone is involved.	Mostly osteoblastic, rarely osteolytic.
Histological incidence of malignant features	2%	11%
Origin	Meningeal cells	From ectopic meningocytes or arachnoid cap cells

extracalvarial (if located outside the calvaria), cutaneous, extracranial, primary extraneuraxial and extradural meningiomas [5],[11],[12]. Extradural meningiomas that arise in the skull have been referred to as calvarial, intradiploic and intraosseous [13]. Primary extradural meningiomas are different from primary intradural meningiomas in that they may have secondary extracranial extensions and/or they may have metastasized. Yilmaz et al reported a case of frontal primary osteolytic introsseous meningioma in a 41 year old male [14]. Sheikhrezaie et al described a case of primary introsseous osteolytic meningioma in the fronto-parietal region in a 62 year old male [15]. Pure primary intraosseous localization without underlying dural involvement is very rare [16],[17]. Our case is an example of pure primary intraosseous meningioma, which neither had any infiltration or attachment with the underlying dura nor with the scalp tissue.

A comparison between extradural and intradural meningiomas is summerised in [Table/Fig 5] [5].

Lang et al classifed extradural meningiomas, based on their position with respect to the cranium. Type I tumours included lesions that were purely extracalvarial, with no attachment to the bone. Type II tumours were purely calvarial, being located entirely within the bone of the skull. Type III tumours corresponded to the calvarial tumours with extracalvarial extension; in other words, a tumour that is located within the skull but also having a soft-tissue component that is extended extracranially. Type II and type III tumours were subdivided into the skull base (B) or convexity (C) tumours [5]. Our case was an example of type II tumour.

The convexity and the skull base are the two primary locations for intraosseous meningiomas. Like many intracranial lesions, the clinical presentation and differential diagnosis depend largely on the size and location of the lesion. Convexity intraosseous meningiomas most commonly present as slowly growing scalp masses, with a possible relationship to a cranial suture [19]. The common locations include the periorbital region and the frontoparietal region [20]. These are typically firm and painless, with normal overlying skin and may bedetected incidentally [12]. The neurological signs and symptoms are usually absent in the patients; however, the presenting symptoms such as neurological deficit, seizures, vomiting, dizziness, hearing loss, tinnitus, headache and vague sensations in the head have also been reported [22],[23],[24]. These symptoms may be present for months or years prior to the diagnosis. Like their convexity counterparts, skull base intraosseous meningiomas are usually slow- growing and painless. Their location, however, may cause different symptoms than the convexity lesions for months to years prior to the diagnosis [25]. These symptoms may include cranial nerve deficits such as ophthalmoplegia or visual field problems, or the signs and symptoms which are related to mass effects such as proptosis or

deformity [25]. The associated findings on ophthalmological examination may include conjunctival oedema and optic atrophy. The lesions involving the nasal cavity or the sinuses may present with nasal obstruction or epistaxis [5]. Azar-Kia, et al. postulated that calvarial meningiomas arise from meningocytes that are trapped in the cranial sutures during head molding at birth [18].

The microscopic features of intraosseous meningiomas are similar to that of convential meningiomas i.e arranged in sheets and forming whorls and psammoma bodies. The cells are polygonal with central nuclei, often containing empty looking vacuoles and homogenous cytoplasm [17]. The bone appears normal, with replacement of the marrow by fat, fibrosis and tumour cells. Few case reports on intraosseous meningiomas revealed meningotheliomatous meningiomas [22],[24],[26],[27] and also the microcystic [27], psammomatous [28], transitional [25], chordoid, atypical, malignant and the fibroblastic types [29]. The authors of one report reviewed 48 cases in the literature and found that the majority (30 cases, 62.5%) were of the meningothelial type; transitional (12 cases, 25%) was the next most common, followed by fibroblastic (4 cases, 8.3%), psammomatous (1 case, 2.1%) and malignant (1 case, 2.1%) [24]. Like intradural meningiomas, the tumour cells of intraosseous meningiomas are usually positive for vimentin and epithelial membrane antigen, but are negative for desmin, cytokeratin, and glial fibrillary acidic protein [24],[30]. The tumour cells may show positive staining for the S100 protein [30]. In another report, the (MIB-1) staining index was found to be 2.4%, suggesting a low proliferative potential [24].

Recent studies indicate that intraosseous meningiomas have a higher incidence of malignant features than the intradural meningiomas [24],[30]. These malignant features may be indicated by microscopic tumour invasion of the underlying dura or the overlying soft tissue structures [26]. Other features which are possibly consistent with a more malignant subtype include increased mitotic activity, increased cellularity, atypia, papillary features, giant cells and focal areas of necrosis. Metastasis, although rare, also indicates a more malignant subtype. One report reviewed 65 published cases of intraosseous meningiomas which were assessed during the CT era and found that 17 (26%) had atypical or malignant histological features [5]. Osteolytic lesions, although they are a more rare form of intraosseous meningiomas, have a higher likelihood of atypical or malignant features as compared to the osteoblastic tumours. In our case, there was no evidence of malignancy and also, the patient was disease free after 19 months of treatment.

The treatment for intraosseous meningiomas is the total excision of the involved bones. Cranioplasty may be performed simultaneously or at a later date. Our patient underwent complete surgical excision of the tumour. Cranioplasty was advised during follow up, which she refused.

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