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Case Report

Primary Giant Sphenotemporal Intradiploic Meningioma

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Abstract

Intradiploic meningioma is a rare subset of meningioma accounting for 1% of all cases. Authors report a rare case of giant sphenotemporal intradiploic meningioma with orbital extension in a 27-year-old female. It was managed successfully with complete surgical excision and bony reconstruction using autologous split thickness bone graft.

Keywords: *Intradiploic, intraosseous, meningioma, primary extradural meningioma, sphenotemporal*

Introduction

The meningiomas which arise from location other than meninges are termed ectopic meningiomas.[1] Intradiploic or intraosseous meningioma is a rare type of ectopic meningioma constituting <1% of all meningiomas.[1-3] They usually arise in the first two decades of life.[2,4,5] The authors describe a rare case of a giant sphenotemporal intradiploic meningioma in a 27-year-old female patient.

Case Report

A 27-year-old female patient presented to the Outpatient Department of our hospital with complaints of progressive swelling in the left temporal and orbital region with associated intermittent mild headache for past 2 years. She also complained of visual blurring in her left eye for last 6 months. On clinical examination, a nontender bony hard swelling in the left temporal region of 6 × 7 cm size with mild nonaxial proptosis was noticed. Visual acuity in the left eye was 6/9, and there were no field defects. Rest of the neurological examination was normal. The patient was concerned because of her cosmetic disfigurement.

Neuroradiologic findings

Computerized tomography (CT) of head revealed a large, well-defined, extraaxial, expansile, lytic intradiploic lesion measuring 6 × 7 cm predominantly in the left temporal bone also involving middle cranial fossa floor and the sphenoid bone reaching up to the left orbit superiorly and sphenoid sinus medially. Bone window of CT scan further demonstrated the intradiploic nature of the lesion [Figure 1a-c]. On magnetic resonance imaging (MRI), the lesion was hypointense on T1-weighted, hyperintense on T2-weighted sequences, and demonstrated homogenous enhancement after gadolinium administration [Figure 2]. Based on radiological findings, a working diagnosis of intradiploic meningioma was made.

Surgical intervention

After detailed workup, the patient was taken for elective microsurgical excision under general anesthesia. A left frontotemporal orbitozygomatic craniotomy was performed. Intraoperatively, the tumor was limited to intradiploic space, and there was no evidence of either dural or parenchymal invasion. Both the inner and outer tables were thinned out and surrounded the tumor completely. The underlying temporal lobe and dura were seen buckled under pressure but were not involved by tumor [Figure 3]. The

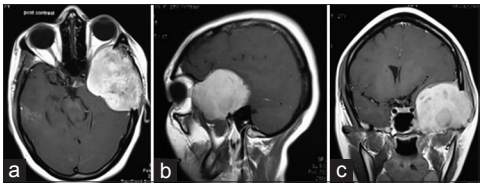

Figure 1: Contrast-enhanced computed tomography head with bone window (a-c) demonstrating well-defined, lytic, sharply marginated intradiploic lesion marked with an arrow in the left sphenotemporal region reaching up to orbit and sphenoid sinus medially. Bony shell (arrow) is seen all around the lesion

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tumor was firm in consistency, moderately vascular, and surrounded all around by a thin bony shell. The findings were consistent with an intradiploic meningioma. Complete excision of the tumor was performed. Bony defect was reconstructed esthetically using the adjacent autologous split calvarial bone graft. Postoperative course was uneventful. She was doing well at her last follow-up visit 3 months after surgery [Figures 4 and 5].

Histopathological examination

Figure 6 showed a moderately cellular tumor, arranged in sheets with interspersed collagen bundles. Tumor cells exhibited a moderate amount of clear to eosinophilic cytoplasm, fine chromatin with inconspicuous nucleoli, and occasional mitoses. There were no areas of hemorrhage, necrosis, or staghorn-like vasculature. Tumor cells were immunopositive for epithelial membrane antigen and vimentin whereas immunonegative for cytokeratin, CD99, and CD34. MIB1 labeling index was 2% (low). Based on the immunohistochemical profile, a final diagnosis of meningioma (WHO Grade I) was rendered.

Discussion

Meningiomas usually arise from the dura; however, rarely they may arise in neck, skin, paranasal sinus, oral cavity, salivary glands, skull, and orbit. Winkler was the first to describe the extradural meningioma.[6] Primary intraosseous or intradiploic meningioma is an extremely rare variety of meningioma accounting for about 1% of all meningiomas.[1,2,5,7] Lang *et al*. classified primary extradural meningioma (PEM) into three categories. Type 1 (purely extracalvarial), Type 2 (purely calvarial), and Type 3 (calvarial with extracalvarial extension). Type 2 and 3 can be subclassified as B (convexity) and C (skull base).[1] According to this classification system, our case is classified as Type 2C. Calvarial type of PEM account for 68% of all cases according to literature.[1,2]

Thirty-six cases of intradiploic meningioma were reviewed by Crawford *et al*.[5] Orbital and frontoparietal region are the most common sites for intradiploic meningioma, and sphenotemporal location is extremely rare. According to Cirak *et al*., psammomatous variety is the most common histological subtype.[8] The exact origin of these tumors is controversial, and many hypotheses have been proposed.[2,9-11] The most acceptable being the

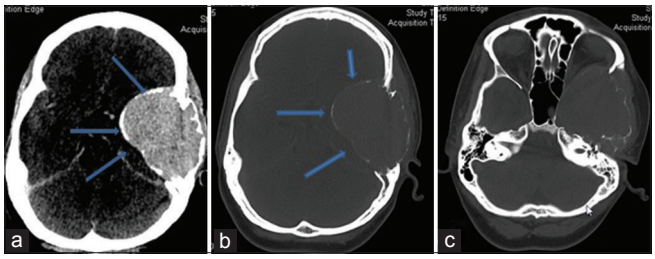

Figure 2: Contrast magnetic resonance imaging demonstrating homogenously enhancing left sphenotemporal lesion in axial (a), sagittal (b), and coronal planes (c)

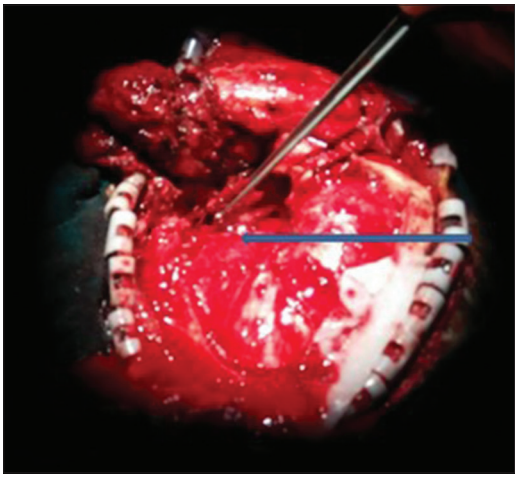

Figure 3: Intraoperative image demonstrating the pure extradural nature of lesion with intact dura

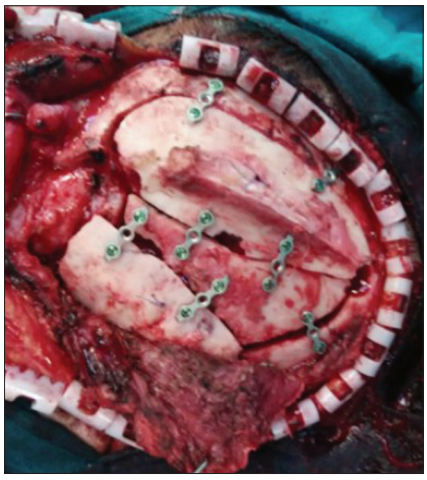

Figure 4: Post bony reconstruction using autologous split calvarial bone graft and titanium plates

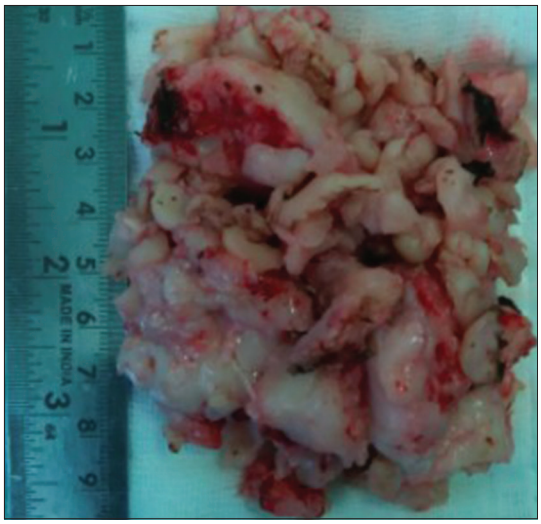

Figure 5: Resected tumor tissue

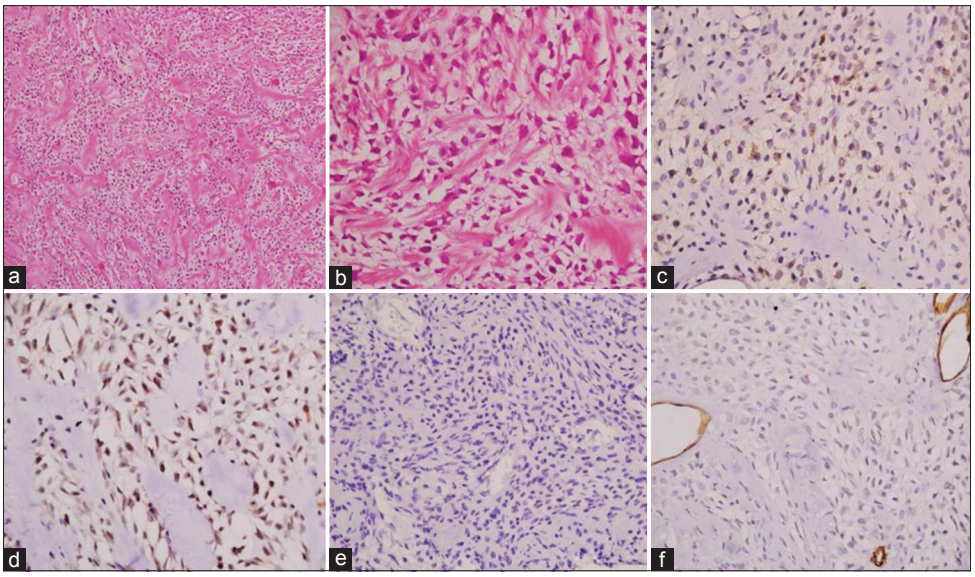

Figure 6: Photomicrographs showing sheets of tumor cells with interspersed collagen bundles (a: H and E, ×40); polygonal cells with clear to eosinophilic cytoplasm (b: H and E, ×400); immunoreactivity for epithelial membrane antigen (c: Immunohistochemistry, ×400) and vimentin (d: Immunohistochemistry, ×400); immunonegative for cytokeratin (e: Immunohistochemistry, ×400) and CD34 (f: Immunohistochemistry: ×400)

entrapment of meningocytes or arachnoid cap cells in cranial sutures during head molding at the time of birth and neural embryogenesis.[1,2] Implantation of arachnoid cap cells during mechanical trauma or dural tear has also been hypothesized.[4] There was no definite history of significant trauma or old skull fracture in our case. Direct origin from multipotential mesenchymal or metaplasia of mesenchymal cells has also been proposed. Another well-accepted theory is the cellular dedifferentiation of abnormal cells in the diploic space.[2] These tumors may be osteolytic, osteoblastic, or mixed type on imaging.[5,10,11] Aggressive lesions tend to involve soft tissue and are osteolytic in nature.[5,9-11] PEM grows slowly over time and produces signs and symptoms due to compression of adjacent neural structures. Tumor excision with wide surgical resection and meticulous bony reconstruction is the treatment of choice in symptomatic patients.[2,5,7]

Surgical resection is further warranted to confirm the diagnosis and to exclude other differentials such as plasmacytoma, metastasis, and fibrous dysplasia, and decide upon adjuvant therapy.

Complete surgical excision is possible as these tumors are usually encased in a bony shell.[1,2,7,10] In the present case, autologous split bone graft was used for bony reconstruction. Intradiploic meningioma usually does not cause dural breach, and intradural exploration is not advised if there is no dural infiltration.[1,2,5,8]

Conclusion

Intraosseous or intradiploic meningioma is a rare type of meningioma, and excellent surgical results can be achieved in view of the extradural nature of the lesion. Authors recommend autologous split calvarial bone graft for reconstruction of the bony defect to achieve desired cosmetic result. CT with bone window supplemented by contrast-enhanced MRI helps to make a preoperative diagnosis. Intradiploic meningioma should be considered as a differential in patients presenting with expansile bony lesions.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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