ISSN 1220-8841 (Print)
ISSN 2344-4959 (Online)

ROMANIAN
NEUROSURGERY

Vol. XXXVI | No. 3 September 2022

Intradiploic meningioma. A series of two cases and literature review

Neha Singh,
Deepak Kumar Singh,
Rakesh Kumar

DOI: 10.33962/roneuro-2022-057

Romanian Neurosurgery (2022) XXXVI (3): pp. 342-345
DOI: 10.33962/roneuro-2022-057
www.journals.lapub.co.uk/index.php/roneurosurgery

Intradiploic meningioma. A series of two cases and literature review

Neha Singh[1], Deepak Kumar Singh[2], Rakesh Kumar[2]

[1] Department of Radiodiagnosis and Imaging, Dr Ram Manohar Lohia Institute of Medical Sciences, Lucknow, INDIA
[2] Department of Neurosurgery, Dr Ram Manohar Lohia Institute of Medical sciences, lucknow, INDIA

ABSTRACT

Intradiploic meningioma is a rare subtype of primary intraosseous meningiomas. The convexity and the skull base are the two primary locations for intraosseous meningiomas. They usually present as a painless expansile mass without any neurological findings. Intradipoic meningiomas are seldom correctly diagnosed preoperatively and are usually mistaken for primary bone tumours. CT scan head with bone windows and contrast-enhanced MRI brain should be done for diagnosis and evaluation. Intraosseous meningioma should be considered in the differential diagnosis of patients presenting with osteoblastic or osteolytic skull lesions as the treatment of choice is resection, which is potentially curative.

Keywords
primary intraosseous meningioma, intraosseous meningioma, intradiploic meningioma

INTRODUCTION

Primary intraosseous meningioma is the most uncommon manifestation of meningiomas.1 The extradural or ectopic meningiomas form 1-2 % of all meningiomas, which was first described by Winkler, in 1904 [5,8]. Most of the intraosseous meningiomas arise from cranial bones, although a few cases in the mandible have been reported [4]. Intradiploic meningioma is a term to describe the purely calvarial meningioma. Preoperative diagnosis of Intradiploic meningioma is very difficult and often diagnosed as bone tumours or metastasis. Here we are reporting two cases of intradiploic meningioma presented with painless scalp swelling and surgically treated with good outcome.

CASE REPORT

Case 1

A 45-year-old man presented with a painless scalp swelling in right fronto-parietal region. The scalp swelling was present for last 6 years and had gradually increased in size. There was no history of trauma. Physical examination revealed a swelling in the right fronto-parietal region about 3 cm in diameter. Swelling was non tender, immobile and not adhered to the overlying skin. The patient had no neurologic deficit.

Corresponding author:
Neha Singh

Dr Ram Manohar Lohia Institute of Medical Sciences, Lucknow, India

neha.singh.dr@gmail.com

ISSN online 2344-4959
© Romanian Society of Neurosurgery

First published
September 2022 by
London Academic Publishing
www.lapub.co.uk

The laboratory studies were unremarkable. CT head revealed a right-sided fronto-parietal intradiploic mass expanding the calvaria with marked thinning of both inner and outer tables with areas of cortical breach. The lesion was protruding extra cranially causing focal bulge over the scalp. The intracranial component was causing compression of underlying brain parenchyma. MR imaging showed a solitary calvarial mass lesion of the frontoparietal region that was hyper intense on T2-weighted images, hypo intense on T1-weighted and was showing marked post contrast enhancement. MR imaging also demonstrated intracranial but extradural extension of the lesion. (Figure 1) Preoperative diagnosis of primary or secondary malignant osteolytic lesion was made. Complete workup was done to rule out calvarial metastasis but no primary could be found. Per-operatively scalp was easily elevated from tumour and there was destruction of both the inner and outer tables of the claverium with extradural extention. Total tumour removal with a wide surgical resection followed by cranial reconstruction was done. Final histo- pathological diagnosis of Intra-diploic meningioma was made. At one year follow up, patient was doing well without any evidence of recurrence.

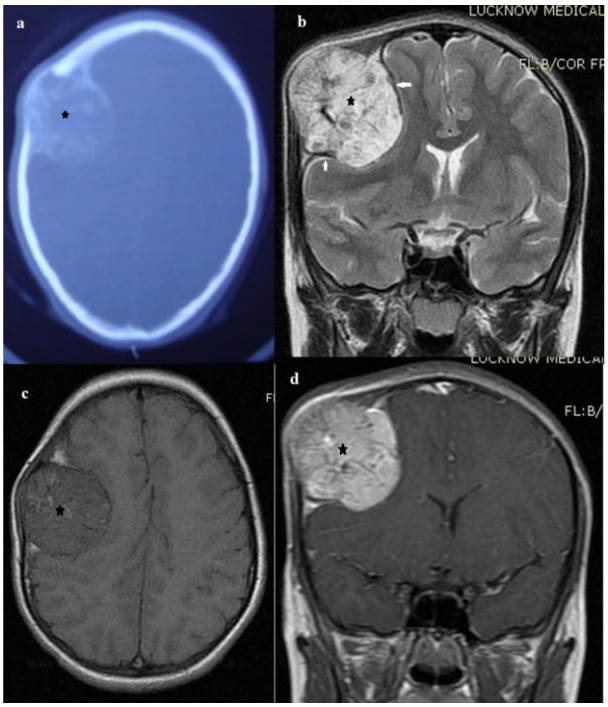

Figure 1. (a) axial image of NCCT Brain in bone window shows right-sided fronto-parietal intra-diploic mass (*) expanding the calvarium with large intra and extra cranial components causing marked thinning of both inner and outer tables with areas of cortical breach. MRI brain shows large intra-diploic expansile lesion (b) hyper-intense on coronal T2WI, (c) hypo-intense on axial T1WI, and (c) marked enhancement on coronal fat supressed T1WI. Displacement of duramater and compression of underlying brain parenchyma by the intracranial component of the lesion is clearly visible (arrow).

Case 2

A 50-year-old man presented with a painless progressive swelling of scalp the right frontal region and headache for eight years. There was no history of trauma. Physical examination revealed a swelling of about four cm diameter in the right frontal region. Swelling was non tender, nonmobile and not adhering to the overlying skin.

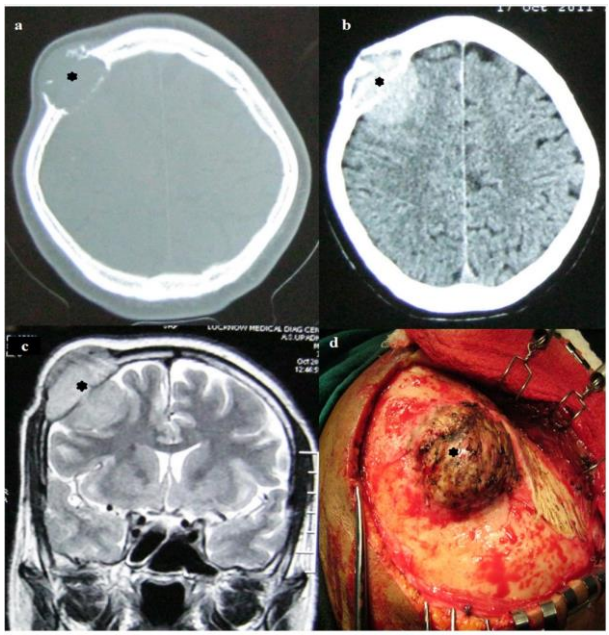

Figure 2. (a) axial image of NCCT Brain in bone window and (b) soft tissue window shows right-sided frontal intra-diploic expansile lesion (*) with intra and extra cranial components causing destruction of both inner and outer tables. MRI brain shows large intra-diploic expansile lesion (c) T2WI of MRI brain in coronal plane shows hyper-intense lesion causing compression of underlying brain. (d) Per- operative image showing extra- calvarial component of the lesion.

The patient had no signs of neurologic deficit. The laboratory investigations were within normal limits. CT Scan of head revealed a right-sided frontal intradiploic mass expanding the calvaria with destruction of both inner and outer tables. The lesion was extending both intra- and extra cranially.

MR imaging showed a solitary calvarial mass

lesion of the frontal region that was hypo intense on T1-weighted and hyper intense on T2-weighted images with post contrast enhancement. The lesion showed intracranial but extradural extension of the lesion. (Figure 2) Preoperative diagnosis of primary or secondary malignant osteolytic lesion was made. Complete workup was done to find out primary lesion anywhere else in the body, however tests were negative. Per-operatively scalp was easily elevated from tumour and there was destruction of both the inner and outer tables of the calvarium with extradural extension of the lesion. Total tumour removal with a wide surgical resection followed by cranial reconstruction was done. Histo-pathological features were suggestive of Intra- diploic meningioma. On six months follow up patient was well without any evidence of recurrence.

Discussion

Primary intraosseous meningioma is a subtype of extradural meningiomas that arise in bone and does not involve the underlying dura [7]. Approximately 68% of the primary extradural meningiomas involved the calvaria. Intraosseous meningiomas most commonly involve the fronto-parietal and orbital regions. Primary extradural meningiomas occur approximately with equal frequency in both sexes. Primary intradural and extradural meningiomas occur predominantly during later decades of life with a second peak incidence in younger patients (especially during the second decade) in primary extradural meningiomas [3]. Lang et al. classified interosseous meningiomas as type I- purely extracalvarial, type II- purely calvarial and type III- calvarial with extracalvarial extension. The type II and III are further divided as convexity (C) or skull base (B) forms [3]. Thus, intraosseous meningiomas could be considered Type II or III primary extradural meningiomas based on whether extracalvarial extension is observed or not, while intradiploic meningiomas are type II only [3].

There are three explanations for the origin of primary intraosseous meningioma. First being the trapping of arachnoid cap cells in the cranial sutures during birth and head molding suggested by Azar-Kia, et al. It is thought that the formation of small meningoceles containing arachnoid cells during that period which got entrapped within the cranial sutures and give rise to intraosseous meningiomas later in life [1]. Second postulated theory states that lesions are secondary to previous trauma which was first described by Cushing and Eisenhardt [2]. Thirdly it has been postulated that meningiomas may arise from mesenchymal precursors, in reaction to a yet unknown stimulus [9].

Usually, the Neurological signs and symptoms are absent in the patients. A painless expansile mass without any neurological finding is usually the initial symptom. The symptoms are dependent on tumor location, size, and involvement of the neighbouring structures. Meningiomas presenting with scalp swelling and extracranial soft-tissue masses are more aggressive in nature than others [10]. Osteolytic meningiomas associated with a soft-tissue component must be considered malignant until proven otherwise [6].

Although calvarial meningiomas have been observed to be benign and slow-growing, but they are more prone to develop malignant changes (11%) compared with intracranial meningiomas (2%) [3]. Bone remodelling and calvarial thickening at the site of origin of the meningioma are frequent with these tumors. In calvarial meningiomas osteolytic skull lesions are also known to occur. Hyperostosis is the most common radiographic finding (reported in 59% of the cases) but osteolysis, as in our case is also reported in 35% of the cases. Mixed picture of hyperostosis and osteolysis is reported in 6% of the cases in the literature [2]. Scalp swelling with osteolytic skull lesions and extracranial soft-tissue masses suggest the aggressiveness of meningioma.

CT scan head with bone windows is necessary to detect the tumor, cortical destruction, and both intra- and extraosseous extension. MRI brain provides a better anatomic delineation in the evaluation of the soft tissue component and extradural extension of the lesion. Although rare, intraosseous malignant meningioma should be excluded in an osteolytic skull lesion associated with soft-tissue component.

Complete tumour excision with a wide surgical resection followed by cranial reconstruction is the treatment of choice in symptomatic primary intra-diploic meningioma. In subtotal resection due to involvement of critical structures within the orbit, paranasal sinuses, or skull base radiological follow up is mandatory [2]. In symptomatic residual lesions and lesion showing evidence of progression adjuvant radiation therapy is recommended [2].

Conclusions

In the patient having osteolytic calvarial lesions associated with a soft-tissue component, a primary intraosseous meningioma should be considered. Osteolytic meningiomas associated with a soft-tissue component must be considered malignant until proven otherwise. CT scan head with bone windows and MRI brain should be done for diagnosis and evaluation. By keeping the defferential diagnosis in mind while evaluation, this can be treated with good outcome. Complete excision with a wide surgical resection followed by cranial reconstruction should be done.

References

1. Azar-Kia B, Sarwar M, Marc JA, Schechter MM: Intraosseous meningioma. Neuroradiology 6:246–253, 1974.
2. Crawford T S, Ette K. Kleinschmidt. DeMasters and Kevin. O. Lillehei: Primary intraosseous meningioma. J Neurosurg .83:912–915, 1995.
3. Lang FF, Macdonald OK, Fuller GN, DeMonte F: Primary extradural meningiomas: a report on nine cases and review of literature from the era of computerized tomography scanning. J Neurosurg. 93:940–950, 2000.
4. Lell M, Tudor C, Aigner T, Kessler P: Primary intraosseous meningioma of the mandible: CT and MR imaging features. Am J Neuroradiol. 28:129–131, 2007.
5. Muzumdar DP, Vengsarkar US, Bhatjiwale MG, Goel A: Diffuse calvarial meningioma: a case report. J Postgrad Med. 47:116–118, 2001.
6. Partington MD, Scheithauer BW, Piepgras DG:Carcinoembryonic antigen production associated with an osteolytic meningioma. Case report. J Neurosurg 82:489-492,1995.
7. Politi M:Intraosseous hemangioma of the skull with dural tail sign: Radiologic features with pathologic correlation.Am J Neuroradiol. 26:2049-2052, 2005.
8. Pompili A, Caroli F, Cattani F, Iachetti M:Intradiploic meningioma of the orbital roof. Neurosurgery.12:565-568, 1983.
9. Willis RA: Pathology of Tumors, ed 4. London: Butterworths, pp 658–662, 736–746, 1967.
10. Younis G, Sawaya R: Intracranial osteolytic malignant meningiomas appearing as soft-tissue masses. Neurosurgery .30:932–935,1992.