

## Ruptured Intradiploic Epidermoid Cyst Presenting as Acute Bacterial Meningitis in an Young Infant: An Intriguing Case Report

Rajniti Prasad\*, Priyanka Agrawal\*\*, Ankur Singh\*\*\*, Mishra O.P.\*\*\*\*

### Abstract

Intradiploic epidermoid cysts, an uncommon benign lesions that variably affect the two tables of calvaria. The clinical manifestations are usually related to mass effect. Here we report a case of intradiploic epidermoid cyst in right parietal bone with intracranial rupture causing acute bacterial meningitis as per cerebrospinal fluid analysis and cranial tomograph. To the best of our knowledge, acute bacterial meningitis as complication has never been described in the existing literature.

**Keywords:** Epidermoid Cyst; Intradiploic; Meningitis.

### Introduction

Intradiploic epidermoid cysts are benign tumours that are predominantly congenital in origin [1]. Calvaria is the most common site of involvement of osseous epidermoid, although the lesions can also involve mandible, maxilla, tibia, femur, phalanges and temporomandibular joints [2]. The lesions are usually asymptomatic, show insinuating pattern and variable rate of growth. Symptoms, when present, are mostly related to the mass effect of these lesions. Calvarial epidermoid cyst usually presents as palpable lump but may present as headache, focal neurological deficit and features of raised intracranial pressure. We report an unusual complication of intradiploic epidermoid with rupture of the capsule leading to acute bacterial meningitis in a 45 days old infant with a brief review of pertinent literature.

### Case Report

A-45-days male child presented to OPD with complaints of multiple episodes of seizures(tonic

posturing, staring gaze and frothing from mouth) since birth with each episodes lasting for 15 to 20 seconds, followed by unconsciousness for 5 minutes. There was also history of fever, lethargy and poor feeding for 5 days. Seizure was controlled with intravenous lorazepam and phenytoin sodium (loading: 20 mg/kg followed by 5mg/kg/day 12hrly as maintenance). There was no history of respiratory distress. The child was born as full term, appropriate for gestational age, by spontaneous vaginal delivery in hospital and cried immediately after birth. There was no history of birth trauma.

On examination, the infant was lethargic with head circumference non-concordant with the age (between +2 SD and +3SD). The anterior fontanelle was flat and open. A swelling of size approximately 5x2 cm was present in right parietal region, which was firm, non compressible, non mobile, non translucent, fixed to underlying structures. Local temperature over the swelling was not raised. All primitive reflexes were absent.

His complete blood counts(Hb: 14 g/dL, TLC: 12400/mm<sup>3</sup>, Neutrophils: 57%, Lymphocytes 40%, Monocyte 3% and Platelet count: 1,87,000/mm<sup>3</sup>), renal function test, C- reactive protein, thyroid function tests were within reference range. Aerobic blood culture was sterile. Cerebrospinal fluid examination showed total count 252 cells/mm<sup>3</sup> with neutrophilic predominance (85%), glucose 36 mg/dL against blood glucose of 100 mg/dL, and protein; 85 mg/dL.

On CECT, there was evidence of a well defined,

**Author Affiliation:** \*Professor and Incharge, \*\*Senior Resident, \*\*\* Assistant Professor, \*\*\*\* Professor, Department of Pediatrics, Institute of Medical Sciences, Banaras Hindu University, Varanasi-221005.

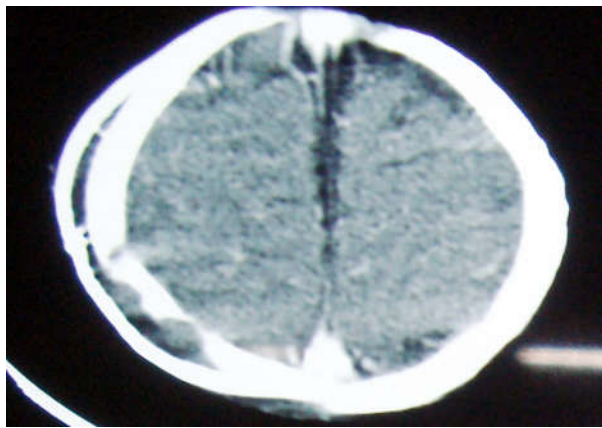
**Reprint Request:** Rajniti Prasad, Professor and Incharge, Division of Pediatric Neurology, Department of Pediatrics, Institute of Medical Sciences, Banaras Hindu University, Varanasi-221005, U.P.

E-mail: rajnitip@gmail.com

© Red Flower Publication Pvt. Ltd.

non enhancing, near CSF density lesion of approximate size 55x 13 x 11 mm. in intradiploic space of right parietal bone with few thin enhancing septae and septal calcifications. Thinning and focal erosion of inner and outer cortex was seen, however there was no periosteal reaction or extraosseous soft tissue component suggestive of intradiploic epidermoid cyst( Figure 1). Ventricular system, posterior fossa, basal cistern, bilateral cerebral and cerebellar hemispheres and deep gray matter nuclei were normal.

Patient received IV antibiotics (vancomycin and ceftriaxone) for 14 days. Patient was discharged on oral phenytoin and oral pyridoxine. Neurosurgery



**Fig. 1:** CT scan show well defined, lesion in intradiploic space of right parietal bone with few thin enhancing septae and septal calcifications

opinion was taken for excision of cyst.

### Discussion

In our case, the CSF examination pointed towards the presence acute bacterial meningitis, while CT scan revealed no evidence of hydrocephalus, parenchymal lesion or abnormal meningeal enhancement. There was presence of an epidermoid cyst in the calvaria with the erosion of the inner calvarial table. Possible rupture of the cyst capsule with discharge of the presumably infected cyst contents into cisternal space was postulated as the etiological factor [3].

Intracranial epidermoid cysts are benign extraparenchymal inclusion cyst that are derived from incoherent ectodermal remnants sequestered due to defects in neuroectodermal separation during 3<sup>rd</sup> to 5<sup>th</sup> week of embryogenesis [1]. It is the most common congenital intracranial tumor and accounts for 0.2-1.8% of intracranial tumors [4]. 75-90% of these tumors are intradural lesions present in cisternal spaces involving cerebellopontine angle, fourth ventricle,

parasellar cisterns, intraventricular with 10-25 % tumours in extradural location involving cranial vault and spine [4,5]. Rarely these lesions can also be present in cerebral hemispheres and brainstem [6,7].

Peak age of these tumours is between 3<sup>rd</sup> and 4<sup>th</sup> decade. Majority of epidermoid cysts are clinically silent for a long time or present with focal swelling over the scalp. The most common symptom is occasional headache. If the tumours attain large size, they can cause neuropathy of V, VII and VIII<sup>th</sup> cranial nerves. Other described symptoms such as tinnitus, hearing loss, intracranial hypertension are rare and reported in literature as isolated case reports [5,6].

Complicated cephalhematomas, eosinophilic granulomas, metastatic lesions should be considered as a differential diagnosis in pediatric age group. MRI is the imaging modality of choice with these lesions depicted as T1 hypointense T2 hyperintense cystic lesions with incomplete nulling on FLAIR sequences. However the most reliable diagnostic is the presence of restricted diffusion on Diffusion weighted sequences with the lesions showing signal comparable to brain parenchyma on the apparent diffusion constant (ADC) maps [8]. In our case, CT scan is highly suggestive of epidermoid cyst and parent could not afford for MRI.

Rupture of the epidermoid cyst wall can result into contained leakage of the cholesterol and debris rich cyst content into the subarachnoid space leading to aseptic meningitis or chronic granulomatous arachnoiditis [4]. To the best of our knowledge, rupture of an infected intradiploic epidermoid cyst, causing acute bacterial meningitis has never been reported.

The management of these lesions is surgical excision of the cysts along with the capsule which are the only growing part of these tumors.

### References

1. Toglia JU, Netsky MG, Alexander EJr. Epithelial (epidermoid) tumors of the cranium. Their common nature and pathogenesis. *J Neurosurg.* 1965; 23: 384-93.
2. Narlawar RS, Nagar A, Hira P, Raut AA. Intradiploic epidermoid cyst . *J Postgrad Med.* 2002; 48(3): 213-4.
3. Velamati R, Hageman JR, Bartlett A. Meningitis secondary to ruptured epidermoid cyst: case-based review. *Pediatr Annals.* 2013; 42: 248-51.
4. Kalgutkar A, Kini S, Jambhekar N, et al. Intradiploic primary epithelial inclusion cyst of the skull. *Ann Diag Pathol.* 2006; 10: 20-23.
5. Moreira-Holguin JC, Medelez-Borbonio R,

- Quintero-Lopez E, Garcia-Gonzalez U, Gomez-Amador JL. Intradiploic epidermoid cyst with intracranial hypertension syndrome: Report of two cases and literature review International Journal of Surgery Case Reports. 2015; 16: 81-6.
6. Iaconetta G, Carvalho GA, Vorkapic P, et al. Intracerebral epidermoid tumor: a case report and review of the literature. Surg Neurol. 2001;55: 218-22.
  7. Kachhara R et al: Epidermoid cyst involving the brain stem. Acta Neurochir(Wien). 2000; 42: 97-100.
  8. Katzman GL. Epidermoid cyst. In: Diagnostic imaging: brain. Salt Lake City, Utah: Amirsys. 2004; 1-7-16.
-