



Image Report

Intradural extension of mucocele secondary to giant frontal sinus osteoma: Diagnostic pitfalls

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ABSTRACT

Background: Paranasal sinus osteoma in association with intracranial mucocele is a rare entity while intradural extension is even rarer. Our aim of presenting this case is to highlight the diagnostic pitfalls and reiterate the importance of prompt treatment of expected complications.

Case Description: A 35-year-old known epileptic, for the past 5 years, presented with altered sensorium for the past 2 days. Computed tomography (CT) of the brain plain showed ventriculomegaly and cystic lesion in the left frontal lobe adjacent to a calvarial osteoma. A ventriculoperitoneal (VP) shunt was done which resulted in tension pneumocephalus and led us to discover the origin of osteoma from the left frontal sinus on CT functional endoscopic sinus surgery (FESS) protocol. He underwent left frontal craniotomy. The osteoma and mucocele were excised completely and watertight primary dural closure was done. Postoperative meningitis was treated with antibiotics according to the culture report.

Conclusion: Intracranial extension of mucocele led to meningitic hydrocephalus, prompting us for VP shunt. Resulting tension pneumocephalus revealed what was missed on preop CT, a small pocket of air adjacent to osteoma intracranially. Therefore, this case underscores the importance of obtaining a preoperative CT FESS to elaborate the origin of osteoma, thus planning approach differently.

Keywords: Frontal sinus, Intradural extension, Mucocele, Osteoma

INTRODUCTION

Osteomas are slow-growing benign tumors of unknown etiology, affecting population in the 2nd and 3rd decades with slight male preponderance (2:1). The proposed etiologies are developmental, posttraumatic, or postinfectious. About 3% occur in association with paranasal sinuses (PNSs) having predilection for fronto-ethmoid sinus.^[3,4] The size usually varies from 2 to 30 mm, those larger than 30 mm are categorized as large or giant osteomas.^[1] The symptoms depend on size, smaller ones are usually asymptomatic while larger ones either have intraorbital extension resulting in proptosis, epiphora, and diplopia or intracranial extension leading to serious complications such as pneumocephalus, rhinorrhea, bacterial meningitis, and mucocele.^[6,8] PNS osteoma in association with intracranial mucocele occur in <1% of cases,^[5] while intradural extension is even rarer, only 19 cases have been reported so far.

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CASE DESCRIPTION

A 35-year-old male known epileptic, presented with altered sensorium for the past 2 days. On local examination, a 5 × 3 cm irregular bony swelling noted on the left forehead. There was no focal neurological deficit and no signs of meningeal irritation. Computed tomography (CT) of the brain plain showed a calcified lesion in the left frontal region along with ventriculomegaly and a huge cystic lesion in the left frontal region [Figure 1a]. Magnetic resonance imaging (MRI) revealed a lobulated irregular lesion causing mild compression on the underlying brain parenchyma. There was marked dilation of ventricles with transependymal seepage

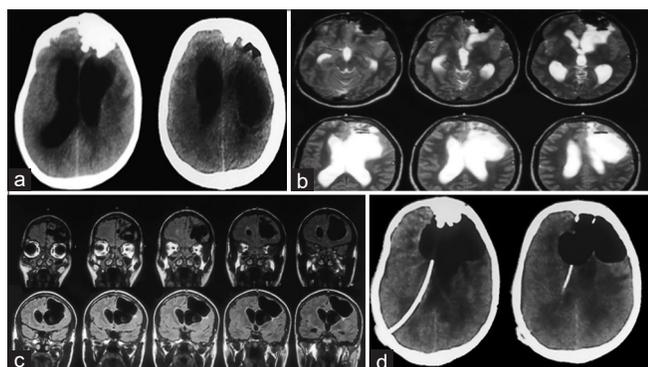


Figure 1: (a) Plain computed tomography (CT) brain showing left frontal irregular bony growth with intracranial projection, ventriculomegaly and left paraventricular cystic SOL. (b) Magnetic resonance imaging (MRI) brain plain T2 axial images showing irregular hypointense left frontal lesion encroaching on left frontal lobe, paraventricular cystic lesion of cerebrospinal fluid intensity is noted with air-fluid level in it. (c) T1 coronal MRI depicting relation of cyst to ventricle. (d) Post-op CT brain plain following ventriculoperitoneal shunt done for hydrocephalus showing enlargement of aerocoele.

suggesting high-pressure hydrocephalus [Figure 1b and c]. Patient was shunted immediately and clear cerebrospinal fluid (CSF) under high pressure was obtained. Routine analysis of CSF showed pleocytosis, with negative cultures. Patient postoperative CT scan showed tension pneumocephalus [Figure 1d].

CT bone window was done, which elaborated details of osteoma [Figure 2a-c]. A left frontal craniotomy with free bone flap was done [Figure 2d]. About 5 × 2.5 cm irregular hard bony growth found disrupting the underlying dura [Figure 2e]. The cystic lesion was accessed through dural defect, thin clear mucoid substance drained and excised completely. There was no communication between cyst and ventricle at any point. Remaining osteoma was excised and sinus ostia was plugged with muscle piece. The dural defect was repaired and further reinforced with Dietz pericranial flap. Postoperatively patient did well, scans were satisfactory [Figure 2f] and no CSF leakage was found. On the 5th day, the patient developed fever and neck rigidity. A moderate growth of Colistin-sensitive *Klebsiella* species and *Acinetobacter* were cultured from CSF. A 2 week course of Colistin resulted in clinical improvement and patient was discharged home. Histopathology report showed a tumor with typical features of osteoma. Currently the patient is in follow-up, fits are controlled with oral sodium valproate.

DISCUSSION

In neurosurgical emergency, cases of hydrocephalus presenting with altered sensorium are routinely shunted. Here, we attempt to highlight a simple but rare cause of hydrocephalus, in which the underlying cause was overlooked being rare as well as lack of detailed 3D imaging in ER.

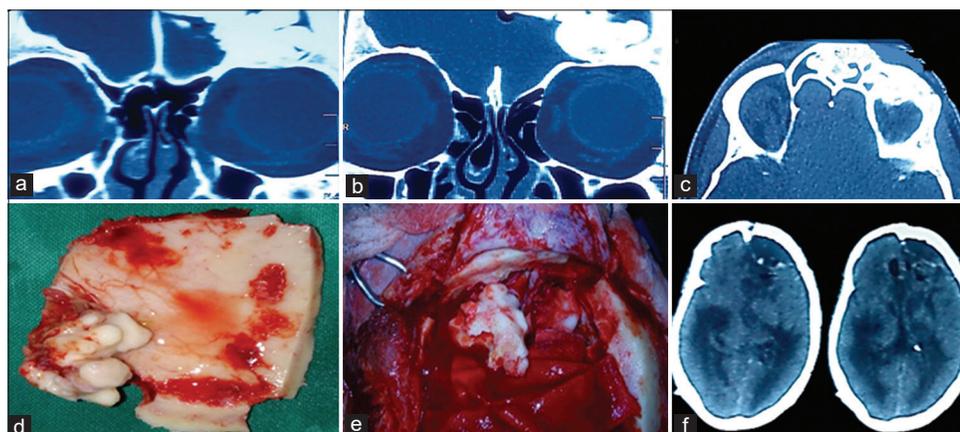


Figure 2: Computed tomography (CT) bone window (a and b) coronal view showing attachment of osteoma to the roof of orbit, lateral to the lamina papyrea signifying no ethmoidal involvement (c) axial view showing AP diameter of lesion filling >75% of frontal sinus respectively. (d) Inner aspect of free bone flap incorporating part of osteoma. (e) Remaining osteoma projecting from left frontal sinus (f) postop scan showing complete excision of osteoma but delayed extradural collection.

In this case, osteoma was assumed to be of calvarial origin as no apparent association with cyst or any sinus was appreciated on routinely done CT. More than 1 cm slice thickness, bony lesion overlapping orbital roof and acquisition only in axial view are the potential pitfalls of routine scan which can be resolved by CT functional endoscopic sinus surgery (FESS). It was the enlargement of aerocele after shunt which raised the suspicion of relation of osteoma with PNS. To confirm, CT FESS was done, which revealed the exact anatomical details of osteoma in 3D. Retrospective review of imaging revealed small foci of air entrapped between osteoma and adjacent brain on plain CT and air-fluid level within cyst on MRI brain.

We speculate that the meningitis due to intradural extension of mucocele was the cause of altered sensorium as well as hydrocephalus. This is supported by CSF pleocytosis found after shunting. The absence of signs of meningitis at presentation and negative culture after shunt cannot reliably exclude meningitis. The existing guidelines for surgical management are based on the grading system proposed by Chiu,^[2] with endoscopy reserved for low grade (I, II) and osteoplastic flap for high-grade osteomas (III, IV). Ledderose *et al.* and Seiberling *et al.* present series in which a proportion of high-grade osteomas have been treated using an endoscopic approach. However, a higher rate of residual tumor, neostium stenosis, extended surgical time, and occasional use of additional brow incision is reported in these series.^[7,9]

Our case belonged to grade III (high grade) so we chose transcranial route to achieve near-total removal of bony lesion and effective dural repair. We speculate that dural disruption due to mucocele was culprit for bacterial meningitis, and it was there long before cultures were positive.

CONCLUSION

We speculate that the meningitis was the cause of altered sensorium as well as hydrocephalus. The absence of signs of meningitis at presentation cannot reliably exclude meningitis. Small foci of air on CT and fluid level on MRI also provide clue to the underlying pathology. Through evaluation of apparently calvarial bony lesions adjacent to PNS, via CT FESS should be the rule, and if implemented earlier in this case, not only a single procedure dealing with osteoma and CSF diversion would suffice, but later could be temporary thus avoiding permanent shunt hardware.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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