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Case report

Frontal sinus encephalocele: Case report and review of literature

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1. Introduction

Anterior encephaloceles are rare congenital abnormalities that occur most commonly in southeast Asia and are characterized by herniation of intracranial components through the cranial and facial bones due to a defect of closure of the anterior neuropore of the neural tube [1]. Congenital anterior encephaloceles are classified by their location such as fronto-ethmoidal, trans-ethmoidal nasopharyngeal, or orbital [1]. Acquired encephaloceles can also occur later in life secondary to tumor, hydrocephalus or other cause [2].

Seizures may be the presenting complaint in a wide variety of intracranial pathology; thus new onset seizures should prompt a thorough work up [3]. Seizures present as provoked or unprovoked events with recurrent unprovoked seizures defining epilepsy. New onset seizures in an adult should prompt neuroimaging to assess for structural anatomic abnormalities [3].

We report the unusual case of a 38 year-old woman who presented to medical attention with new onset seizures, was found to have an opacified right frontal sinus, which was caused by an anterior encephalocele in the right frontal sinus via a defect in the sinus's posterior wall.

2. Case report

A 38 year old previously healthy and developmentally normal female presented with new onset seizures and severe bifrontal headache of one week's duration. The patient initially experienced 5 min of generalized tonic clonic activity and a similar episode 2 days later, which also lasted about 5 min. Careful history taking elicited history of intermittent clear rhinorrhea of several weeks duration. Patient had no history of trauma, infection, previous craniofacial surgery or drug use. The patient had no evidence of neurological deficits upon presentation and was without nuchal rigidity or facial tenderness. No obvious cerebrospinal fluid (CSF) leak was observed, but the patient had a history of intermittent clear drainage. CT head demonstrated opacified right frontal sinus and patient was initially treated for complicated sinusitis. The patient was admitted, the antiepileptic medication levetiracetam was administered and an electroencephalogram was performed with was abnormal showing moderate generalized slowing without electrographic seizures. Antibiotics were given and lumbar puncture was obtained to assess for meningitis. Cerebrospinal fluid analysis was unremarkable. Although the patient had no clinical signs or symptoms of meningitis, she was treated empirically with antibiotic therapy for para-meningeal infection. Careful evaluation of a dedicated maxillofacial CT showed a defect in posterior table of right frontal sinus (Fig. 1a) and an MRI of the brain demonstrated a frontal sinus mass isointense to adjacent brain with opacification of frontal sinus and surrounding cerebritis (Fig. 1b and c), raising the possibility of an encephalocele. The patient was then referred to us for further evaluation and treatment. The patient had no further seizures after the initiation of antiepileptic mediations.

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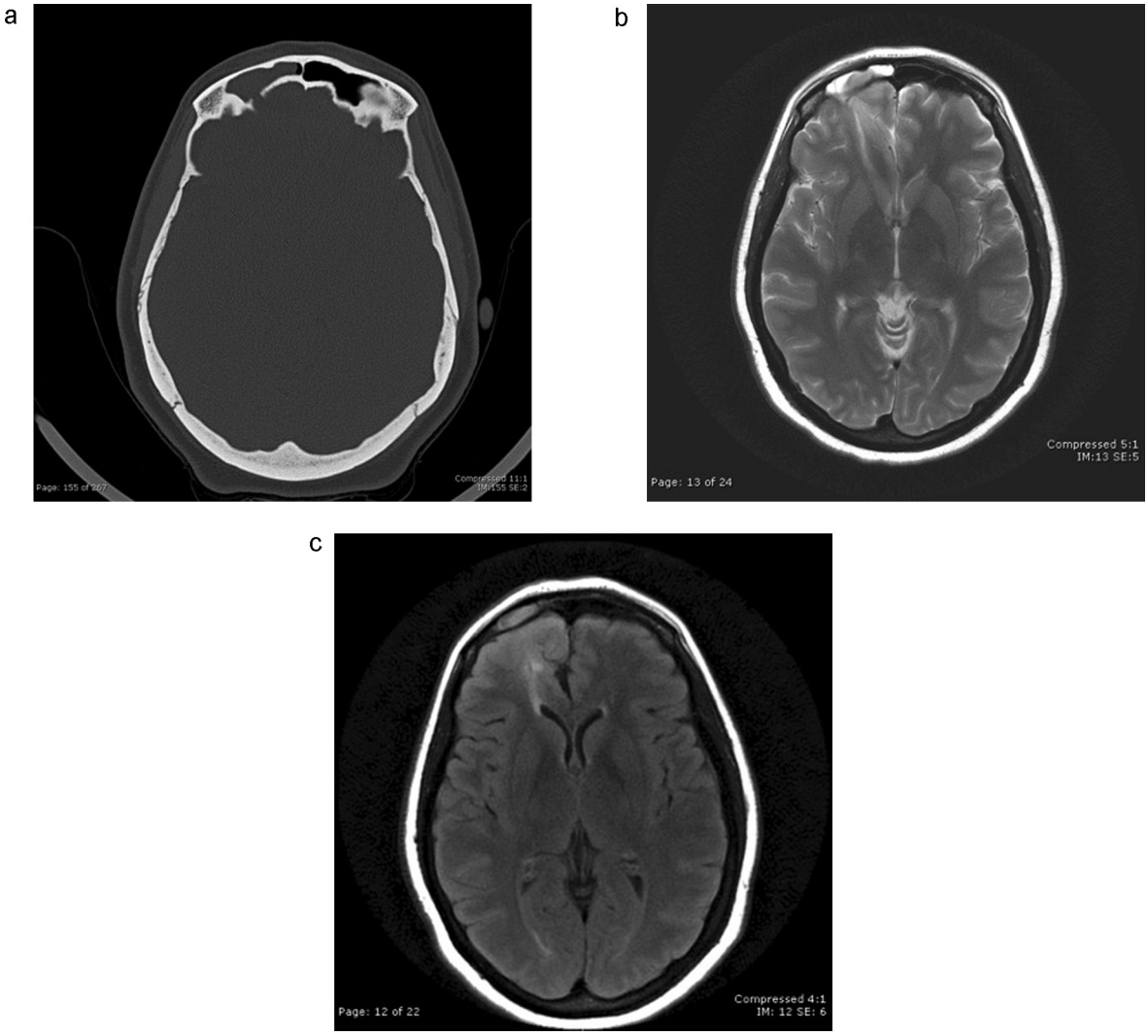

Fig. 1. (a) CT of the head bone window demonstrating opacification of right frontal sinus and defect in posterior wall of right frontal sinus, (b) MRI T2 weighted image and (c) FLAIR image showing lesion within right frontal sinus isointense to cortex with right frontal lobe FLAIR abnormality.

Once the patient had completed recommended course of antibiotics, she was taken to the operating room for resection of the encephalocele. A bicoronal incision with bifrontal craniotomy was performed with harvesting of pericranial graft. The encephalocele was encountered within the right frontal sinus (Fig. 2) protruding through the posterior table and dural defects. The lesion was completely contained within the frontal sinus without extension into the nasal cavity. The encephalocele was resected and the dural defect closed. The frontal sinus was then cranialized and the floor repaired with pericranial graft.

The pathology was consistent with an encephalocele demonstrating well-differentiated, well circumscribed neuroglial tissue surrounded by respiratory mucosa (Fig. 3).

The patient's post-operative course was unremarkable, and she was discharged home on post-operative day 4. Post-operative CT showed satisfactory cranialization of frontal sinuses without hematoma. At 11-month follow-up, the patient was doing well with no further seizures, infections or evidence of cerebrospinal fluid leak.

3. Discussion

Frontal sinus opacification is commonly due to sinusitis, mucocoele or less commonly inclusion cysts such as epidermoid cysts [4,5]. Encephalocele is rare but is part of the differential diagnosis for frontal sinus mass, especially when a CSF leak is present. Identifying and treating the encephalocele is paramount to prevent the development of meningitis. Anterior encephalocele is a rare condition that usually presents in childhood due to the external characteristics [1]. Adult presentation of an anterior encephalocele is very rare and usually presents with spontaneous CSF leak or recurrent meningitis [6].

Recurrent bacterial meningitis should prompt imaging of the neuroaxis to assess for an anatomic lesion either congenital or acquired [7]. Acute bacterial meningitis remains a life threatening infection with mortality rates reported as around 20–25%. Encephaloceles can cause recurrent meningioencephalitis due to direct communication of the central nervous system with the external environment, facilitating the entry of pathological microorganisms [7]. The bacterial organism most commonly associated with meningitis in patients with encephaloceles is *Streptococcus pneumoniae*, followed distantly by *Staphylococcus aureus* then *Neisseria meningitidis* [7].

The medical symptom that brought this patient to medical attention was new onset seizures. Though rare, encephaloceles, most commonly temporal, have been associated with epileptic seizures [8]. Temporal encephaloceles are more likely to show electroencephalogram (EEG) lateralizing features. Extratemporal