

Interictal ^{18}F - FDG PET/computed tomography brain in a case of frontoethmoidal encephalocele

ABSTRACT

Encephaloceles, also known as meningoencephaloceles, are a group of neurological disorders characterized by herniation of the cerebral parenchyma along with overlying meninges through openings in the skull bone. They can be seen following congenital defect in neural tube closure or acquired defect in the skull bone. We report a case of acquired frontoethmoidal encephalocele presenting with drug-refractory epilepsy. Interictal FDG PET-computed tomography done for lateralization and localization of seizure focus showed right-sided frontoethmoidal encephalocele associated with hypometabolism in the adjacent right frontopolar cortex, concordant with ictal onset on electroencephalogram.

Keywords: Electroencephalogram, FDG PET brain, frontoethmoidal encephalocele, interictal

CASE REPORT

A 21-year-old male patient with a history of head trauma 5 years back came for the evaluation of drug-refractory focal epilepsy for the past 3 years. His seizures were characterized by sudden-onset behavioral change with oral and right upper limb automatisms lasting for 1–2 min associated with amnesia for the event and no significant postictal deficit. Coronal and axial computed tomographic (CT) images [Figure 1a and b] showed a break in the inner table of the right frontal sinus (solid arrows) likely due to head trauma. Interictal ^{18}F -FDG PET and magnetic resonance imaging (MRI) were done for lateralization and localization of seizure focus. 185 MBq of ^{18}F -FDG was intravenously administered to the patient. The patient was positioned comfortably in a quiet, dimly-lit room for 45 min, and static brain acquisition was done for 10 min under Siemens Healthineers, Biograph Horizon PET-CT scanner. Coronal FDG PET [Figure 2a] and MRI [Figure 2b] images show herniation of the right frontal cortex (solid arrow) into the ethmoidal labyrinth through a defect in the right frontal bone. Coronal and axial FDG PET and MRI images also show hypometabolism with no morphological abnormality in the dorsolateral (Figure 2c-f: dotted arrows) and mesial frontal (Figure 2c-f: dashed arrows) aspect of the right frontopolar cortex. No other area of

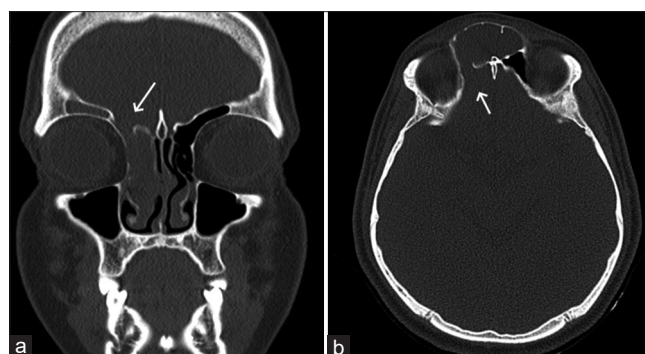


Figure 1: Coronal (a) and axial (b) computed tomography head images in bone window showing break (solid arrow) in the inner table of right frontal sinus as a consequence of head trauma

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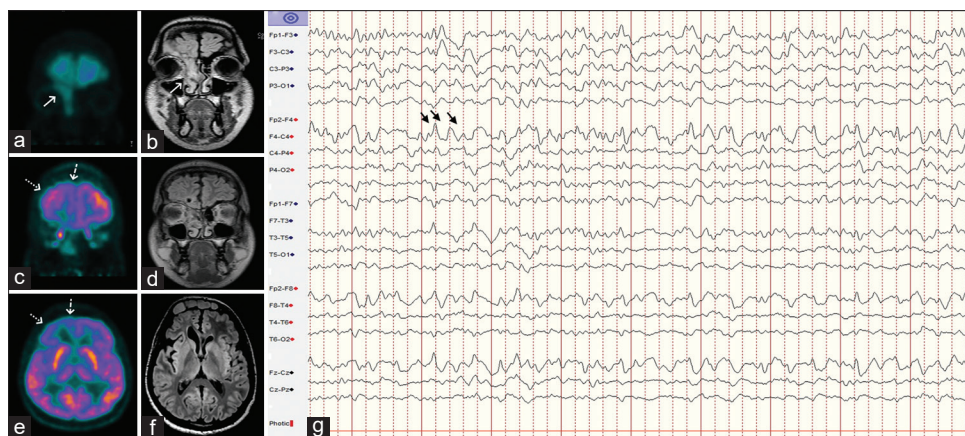


Figure 2: Interictal coronal ^{18}F -FDG PET and magnetic resonance imaging images showing herniation of right frontal cortex into the ethmoid labyrinth (solid arrow) through a defect in the right frontal bone (a and b) along with hypometabolism in the adjacent dorsolateral (dotted arrows) and mesial frontal aspect (dashed arrows) of the right frontopolar cortex (c-f). Video electroencephalogram (g) localized the seizure focus to right frontopolar region corresponding to area of hypometabolism

hypometabolism was seen in rest of the cerebral cortex. Video electroencephalogram done as part of presurgical evaluation recorded two habitual seizures with right frontopolar and right frontal lobe ictal onset [Figure 2g: solid filled arrows]. Based on clinical, electrical, radiological, and functional imaging concordance, a diagnosis of right frontopolar epilepsy secondary to encephalocele was considered. Detailed neuropsychological evaluation with Montreal Cognitive Assessment, Addenbrooke's cognitive assessment, and Frontal Assessment Battery was essentially normal. Following extensive presurgical workup, electrocorticography-guided tailored resection of epileptogenic neural tissue along with protruded neural parenchyma was done. Latter surgical reconstruction of bony and meningeal defect was performed. Neuropathological examination of the operated specimen shows areas of gliosis and abnormal neuronal arrangement in the epileptogenic zone. The patient was seizure-free at 1-year follow-up.

INTRODUCTION

Encephalocele refers to sac-like protrusion of intracranial contents through acquired or congenital defects in the skull bone.^[1] They most commonly occur following congenital failure in neural tube closure. Less commonly, they can also be seen following trauma, surgery, neoplasia, and infectious damage to the skull bone.^[2,3] Depending on the location of the cranial defect, they can be temporal, occipital, frontal, sphenoidal, and rarely parietal.

DISCUSSION

Frontoethmoidal encephalocele represents 15% of all encephaloceles with a herniation of intracranial contents

through the defect in the anterior cranial fossa. Based on location of defect in the frontal bone, there are three subtypes, namely nasoethmoidal (sincipital type), nasofrontal, and naso-orbital. Among these, nasoethmoidal was the most common and naso-orbital was the least common subtype.^[4] Encephaloceles can present as cerebrospinal fluid rhinorrhea, otorrhea, recurrent meningitis, and drug-resistant epilepsy.^[5,6] Long-standing mechanical traction from the herniated cerebral cortex causes chronic inflammation and gliosis in the surrounding neural parenchyma, leading to sensorimotor disturbances, as well as partial epilepsy from the involved cortex as seen in our case.^[7] Further, histopathological examination of epileptogenic zone in few reported cases with acquired encephalocele showed areas of gliosis rather than frank dysplasia, likely due to irritative traction from prolapsed brain tissue.^[1,8] Few cases in the literature reported the possible association of cortical micro dysgenesis, such as focal cortical dysplasia, band heterotopia, and nodular heterotopia with congenital encephalocele.^[9,10] The role of interictal FDG PET in lateralizing partial epilepsy caused due to focal cortical malformations and mesial temporal sclerosis had been reported extensively in the literature. This index case depicts the image findings of less commonly seen acquired frontoethmoidal encephalocele, leading to focal epilepsy.

CONCLUSION

Based on the findings, in this case, interictal PET can play a role in better estimation of extent of epileptogenic zone adjacent to prolapsed brain tissue in case of acquired encephalocele. This helps in tailored surgical procedures for optimal resection of epileptogenic zone, avoiding major neuropsychological deficit.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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