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## Case Report

# Polypectomy-induced encephalocele manifested as meningitis and CSF rhinorrhea in a pregnant woman: a case report <sup>☆</sup>

Amirhossein Soltani, MD<sup>a</sup>, Fariba Zarei, MD<sup>a,b,\*</sup>, Hamid Reihani, MD<sup>c</sup>,  
Mahdi Saeedi-Moghadam, PhD<sup>a</sup>

<sup>a</sup> Medical Imaging Research Center, Shiraz University of Medical Sciences Shiraz, Iran

<sup>b</sup> Department of Radiology, Shiraz University of Medical Sciences, Shiraz, Iran

<sup>c</sup> Student Research Committee, School of Medicine, Shiraz University of Medical Sciences, Shiraz, Iran

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## ABSTRACT

Encephalocele is a protrusion of the intracranial contents through a cranium defect. Encephalocele is divided into primary (congenital) and secondary (acquired) classes. From an epidemiological point of view, primary encephalocele is much more prevalent than secondary cases. Furthermore, among the secondary causes of this condition, iatrogenic encephalocele is recognized as a rare phenomenon. In this case report, we introduce a case of secondary encephalocele in a 30-year-old pregnant female who came to our emergency center at 5 weeks of gestation with a vague headache in her forehead and a runny nose. She reported a history of nasal polypectomy 9 months ago and a 10-day hospitalization for meningitis 5 months prior to admission. MRI of the patient's brain showed evidence of cerebral parenchymal herniation to the right nasal cavity, which was suggestive of encephalocele. She was scheduled for endoscopic transnasal reconstruction, and during the operation, a significant right-sided posterior ethmoidal roof defect with CSF leak and encephalocele was revealed. Eventually, the skull defect was successfully repaired with a vascularized flap, and the patient was discharged in good general condition.

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## Introduction

Encephalocele is a protrusion of the intracranial contents through a skull defect [1]. Encephalocele is divided into primary and secondary categories. The primary causes are

congenital and present from birth. On the other hand, secondary encephalocele is caused by issues such as iatrogenic, hydrocephalus, sinusitis, and trauma [2,3]. Secondary encephalocele can occur as a simple runny nose, often confused by the patient with allergic rhinitis, to meningitis, which has devastating effects on the patient [4,5]. Primary encephalocele is classified into basal, occipital, and sincipital. Among them, the occipital class has the highest frequency. However, no

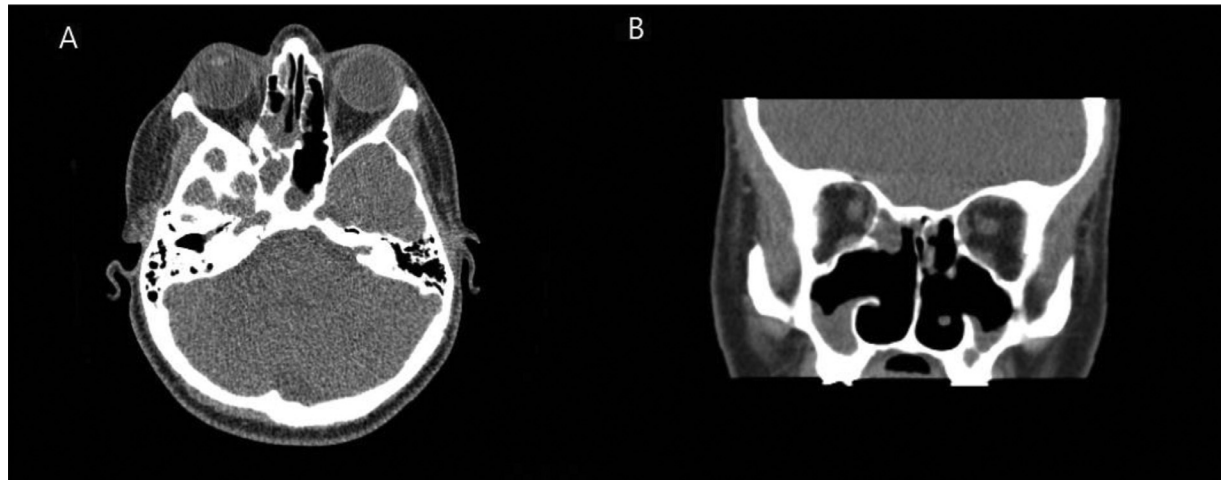
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\* Corresponding author.

E-mail address: [zareifari@sums.ac.ir](mailto:zareifari@sums.ac.ir) (F. Zarei).

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**Fig. 1 – Axial and coronal cut of paranasal sinus CT-scan shows evidence of previous surgery on nasal turbinates and medial wall of both maxillary sinuses (white arrows). Increased mucosal thickening is seen in both maxillary and ethmoid sinuses (black arrows) (A, B).**

precise classification and prevalence have been reported for secondary encephalocele [6,7]. Iatrogenic encephalocele is a rare phenomenon in this disease category, and here we present a case of a 30-year-old woman with a gestational age of 5 weeks who developed encephalocele in her right cribriform plate after nasal polypectomy.

A 30-year-old pregnant female, gravid three para one ab one, came to our emergency department during the fifth week of her pregnancy. She complained of a vague headache in her forehead and a runny nose that the usage of over-the-counter antihistamine pills had not stopped it. The patient's headache started that morning, but her runny nose started four days prior to admission. The character of the patient's pain was compressing and vague, which was worsened with sitting and standing positions. In addition, the patient's nasal discharge was watery and came out of her right nostril, which was intensified by the Valsalva maneuver.

By exploring the patient's archived records, we obtained a history of a 10-day hospitalization of the patient that was related to 5 months ago. In the previous admission, she was presented to the emergency department with a severe headache, nausea, vomiting, stiff neck, and was hospitalized with a diagnosis of meningitis. The patient gradually developed bilateral hearing loss during her previous hospitalization, which eventually led to complete deafness. She also mentioned asthma and sinusitis in her past medical history. The patient reported a cesarean section ten years ago and a nasal polypectomy 9 months ago in terms of surgical history.

Her pre-admission medications included Levetiracetam 500 mg PO BID, Pantoprazole 40 mg PO every morning, Ceterizine 10 mg PO PRN, Folic acid 400 mcg PO QD.

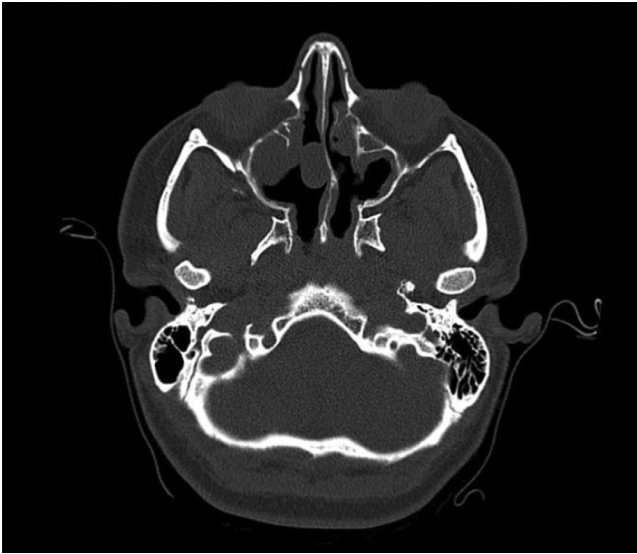
On physical examination, she had a blood pressure of 120/70 mm Hg with a pulse rate of 78 beats per minute (bpm). No abnormalities were observed in her neurological examinations. The external ear was normal, and the tympanic membrane was intact on both sides. There was also no discharge from any of the ears. The nasal mucosa was slightly erythe-

matous, and she complained of occasional obstruction. In addition, a slight discharge came out of the patient's right nostril after the Valsalva maneuver. Furthermore, no signs of scar, trauma, or deformity were observed in the patient's head. The patient's laboratory test results were also in the standard range.

The patient's history of nasal polypectomy increased clinical suspicions of CSF rhinorrhea, so Spiral CT Scanning of the Paranasal Sinuses, Spiral CT scan of Temporal bone, and MRI of the brain without contrast were requested for further evaluation.

CT scan of the paranasal sinuses revealed evidence of middle turbinectomy as well as resection of the medial wall of both maxillary sinuses, demonstrating the patient's previous polypectomy. Para-nasal CT also suggested pan-sinusitis due to the mucosal thickening in the maxillary, sphenoid, ethmoid, and frontal sinuses but showed no trace of CSF rhinorrhea origin (Fig. 1). Temporal CT scan revealed unremarkable internal and external auditory canals and an intact tympanic cavity. In addition, CT scan showed a soft tissue density in the right nasal cavity suggestive of a nasal polyp (Fig. 2). Finally, MRI was able to detect a defect in the cribriform plate with evidence of cerebral parenchymal herniation, which indicated encephalocele (Fig. 3).

According to the diagnosis of encephalocele, Neurosurgery and ENT consultation were requested for her, which according to their discretion, she was scheduled for the endoscopic transnasal reconstruction of encephalocele. During the endoscopic operation and after fluorescein injection, a large right-sided posterior ethmoidal roof defect with CSF leak and encephalocele was revealed. The skull defect was packed with a vascularized flap which was harvested from her right thigh; then, she was transferred to the intensive care unit with a lumbar drain and nasal pack. She was finally discharged from the hospital on the 6th postoperative day with a satisfactory situation. In her subsequent hospitalization, she came to our center 8 months later for a cesarean section and gave birth to



**Fig. 2 – Soft tissue density structure adjacent to right side of nasal bone, suggestive of nasal polyp (white arrow).**

a healthy boy. The patient had no complaints of headache or CSF rhinorrhea in follow-ups performed 6 months and 3 years later, respectively.

## Discussion

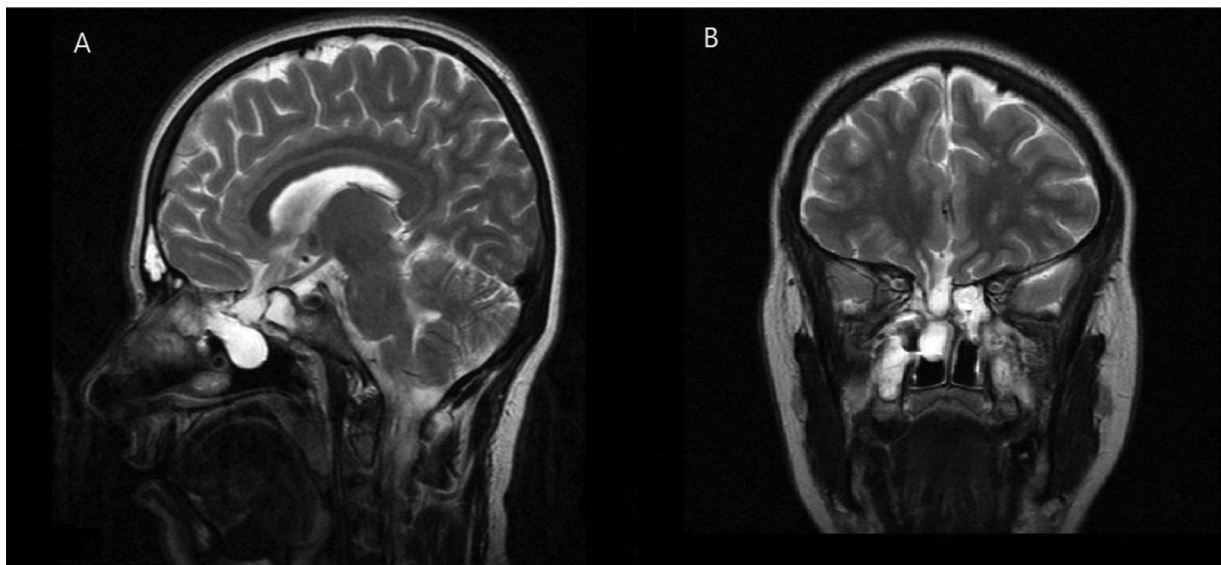
As mentioned above, trauma has the largest share among secondary encephalocele causes, and iatrogenic encephalocele is a rare phenomenon in this category [3]. Our data on iatrogenic encephalocele are mainly limited to case report studies, which procedures such as septoplasty, vacuum extraction deliv-

ery, craniotomy, mastoidectomy, and nasal polypectomy have been reported as the potential causes of acquired encephalocele [8–12]. In our case, polypectomy in the right nasal cavity of the patient was the underlying cause of encephalocele.

Clinical manifestations of encephalocele can include mild to severe headaches, neurological symptoms, ataxia, meningitis, CSF rhinorrhea, etc. There is also a possibility of misdiagnosis of the encephalocele symptoms with situations such as allergic rhinitis, nasal polyps, migraines, or benign lid swelling [13–16]. The most important signs that can bring physicians closer to the diagnosis of encephalocele are spontaneous CSF rhinorrhea and meningitis. In a study by Harada et al [17], of 7 cases examined, 6 had CSF rhinorrhea, and 4 had meningitis. The impressive point about our case was that she was hospitalized four months after the polyp operation with a diagnosis of meningitis, and she was not diagnosed with encephalocele until her second hospitalization, which had a chief complaint of runny nose and headache. Another noteworthy point about our patient is the concurrence of encephalocele diagnosis during pregnancy, which can have many risks for mother and fetus. According to the authors' knowledge, the presence of secondary encephalocele in a pregnant woman has been reported rarely [18].

In the field of diagnostic imaging, our Choice modality is MRI, and CT scan is used to diagnose bone problems, especially in the sincipital types of encephalocele [19]. Like our case, CT scans were more helpful in conditions such as sinusitis and polyps, while MRI was able to identify a brain tissue herniation into the nasal cavity clearly.

Treatment of encephalocele cases is surgery. This surgery can be performed in the form of transnasal or transcranial. Most studies consider the transcranial approach superior to the transnasal method due to its higher sterility, increased field of view of the surgeon, and greater success rate [20]. However, due to the size of the defect and the unpleasant scar that the transcranial operation leaves for the patient, according to



**Fig. 3 – Evidence of small defect is seen in cribriform plate in midline with evidence of brain parenchyma herniation through it (white arrows), which can be suggestive for encephalocele (A, B).**

the consultation with the neurosurgeon, the transnasal procedure was performed for our patient.

In conclusion, encephalocele is most often present congenitally, but this should not distract physicians from secondary cases of encephalocele, which can be caused by various causes such as trauma, iatrogenic and chronic sinusitis. Clinical complaints such as headache, meningitis, and unexplained runny nose in patients with a history of operation in the nasal and head areas should be carefully evaluated to find the underlying cause. Accurate diagnosis of encephalocele is a rare but essential diagnosis for the patient's health, and any delay has irreparable consequences for the patient.

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### Patient consent

The authors have obtained written informed consent from the patient to publish his case (including the publication of images). Written consent was obtained from the patient.

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