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Ahtesham Khizar,  
Adeel Niaz,  
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Imran Ali,  
Khalid Mahmood



# Delayed diagnosis and management of a congenital intranasal meningoencephalocele in a 24-year-old male. A case report revisiting the 'bath-plug' technique

Ahtesham Khizar<sup>1</sup>, Adeel Niaz<sup>1,2</sup>, Syed Muhammad Azeem<sup>1,3</sup>, Imran Ali<sup>1</sup>, Khalid Mahmood<sup>1</sup>

<sup>1</sup> Punjab Institute of Neurosciences, Lahore, PAKISTAN

<sup>2</sup> Lahore General Hospital, Lahore, PAKISTAN

<sup>3</sup> Sir Ganga Ram Hospital/FJMU, Lahore, PAKISTAN

## ABSTRACT

**Background:** Intranasal meningoencephalocele is a rare congenital malformation. It is defined by protrusion of cerebral tissue and meninges through a defect in the skull base. Serious complications usually occur in early childhood, however, symptoms occasionally can wait until adulthood.

**Case Presentation:** We report a case of a 24-year-old male who had a congenital intranasal meningoencephalocele with recurrent episodes of bacterial meningitis. After a delayed diagnosis, he eventually underwent Endoscopic Endonasal Repair of Meningoencephalocele via 'Bath-plug' technique.

**Conclusions:** To prevent improper management, it is crucial that ENT colleagues, to whom these cases are primarily present, are well informed about this unusual condition. For the purpose of preventing further neurological problems, thorough neuroimaging evaluations and successful surgical repair are important. The 'Bath-plug' technique for anterior skull base cerebrospinal fluid leak repair is effective for a wide range of situations.

## BACKGROUND

Intranasal meningoencephalocele is a rare condition and it is characterized by protrusion of the meninges into the nasal cavity through a defect in the cribriform plate of the ethmoid bone. Although it is typically a congenital defect, it can also occur as a result of trauma or persistently elevated intracranial pressure. Meningitis and rhinorrhea can be symptoms; however, the condition can also remain asymptomatic until adulthood. There have been reports of nasal obstruction or unilateral nasal congestion.<sup>1</sup>

## Keywords

congenital,  
intranasal,  
meningoencephalocele,  
CSF rhinorrhoea,  
endoscopy



Corresponding author:  
Ahtesham Khizar

Punjab Institute of Neurosciences,  
Lahore, Pakistan

arwain.6n2@gmail.com

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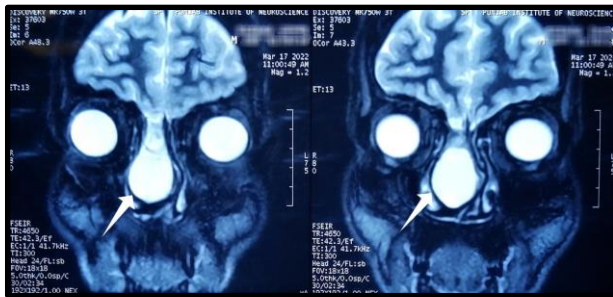
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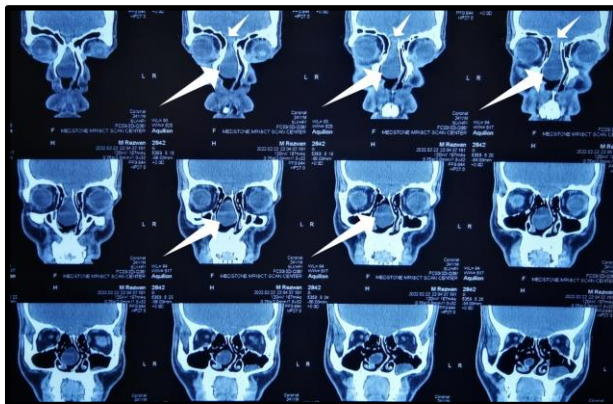
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## CASE PRESENTATION

A 24-year-old male presented to us through outpatient with complaints of right sided nasal discharge since childhood. This was a clear watery discharge and had a salty taste. There was also a complaint of right nasal obstruction and decreased sense of smell due to swelling inside the nose. There were some episodes of epistaxis as well. He had been hospitalized twice due to meningitis in the last 8 years and on the last occasion he remained on ventilatory support for 7 days. On his last visit to an ENT surgeon, he was diagnosed with an intranasal meningoencephalocele and referred to us for further management. His Magnetic Resonance Imaging (MRI) brain and paranasal sinuses revealed an intranasal meningoencephalocele (Figure.1).



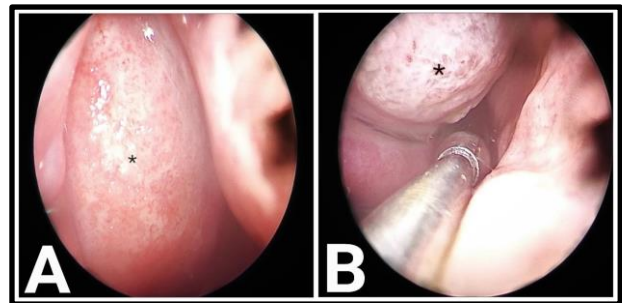
**Figure 1.** MRI Brain T2 weighted coronal image showing intranasal meningoencephalocele (pointed by white arrows).



**Figure 2.** CT paranasal sinuses with FESS protocol coronal image showing defect in the anterior skull base (pointed by small white arrows) and intranasal meningoencephalocele (pointed by large white arrows)/

We performed a Computed Tomography (CT) paranasal sinuses with FESS (Functional Endoscopic Sinus Surgery) protocol for detailed study of meningoencephalocele (Figure. 2). After baseline

investigations and anaesthetic fitness, he underwent Endoscopic Endonasal Repair of Meningoencephalocele via 'Bath-plug' technique. The defect was approximately 13 mm in size. Per-operative endoscopic views of intranasal meningoencephalocele can be seen in Figure. 3 A&B. The 'Bath-plug' technique consists of introducing a fat plug with a specially secured vicryl suture into the intradural space and then placing some traction on the suture to seal the defect similarly as a bath plug seals a bath. This repair was superimposed by dural sealant. Lumbar drain placed preoperatively was removed on the second postoperative day. Postoperative period was uneventful.



**Figure 3. A & B:** showing per-operative endoscopic views of intranasal meningoencephalocele (marked by black asterisks).

## DISCUSSION

Congenital malformations like intranasal meningoencephalocele are extremely uncommon. The signs and symptoms of an intranasal meningoencephalocele can be mistaken for nasal polyps. To prepare for the right surgical approach, it is crucial to identify the type of basal meningoencephalocele that is now present. CT is helpful to delineate the encephalocele sac. It also helps for the precise position and extent of the cranial bone defect. 3D CT is particularly useful in planning an operative technique.<sup>1</sup> To avoid fatal neurological complications from intranasal meningoencephalocele, a thorough neuroradiological assessment and appropriate surgical technique are crucial.<sup>2</sup>

Recurrent meningitis is a rare occurrence. Giunta et al<sup>3</sup> reported a young woman with an intranasal meningoencephalocele who experienced 18 episodes of meningitis. Recurrent meningitis following a number of nasal polypectomy procedures has also been reported due to erroneous diagnosis.<sup>4</sup> The significance of MRI in diagnosis and

treatment is emphasised by these authors as there have been cases of intranasal meningoencephalocele that were initially diagnosed as nasal polyps and resulted in cerebrospinal fluid (CSF) rhinorrhea.<sup>4</sup> Meningoencephaloceles confined to the nose are uncommon in adults. Khan and Salahuddin<sup>5</sup> described the treatment of an adult with a large intranasal meningoencephalocele, and how removal of this lesion caused a gap in the cribriform plate, which was afterwards repaired with tissue and fibrin glue. Bykova *et al*<sup>6</sup> also reported an intranasal meningoencephalocele in a 64 year old female, which clinically resembled a nasal polyp but histological analysis revealed an aberrant glial tissue.

Meningoencephaloceles are thought to be caused by a developmental defect in the closure of the neural tube.<sup>7</sup> These may manifest as a single deformity, but complex malformations of the face, skull, and brain are also possible. Meningoencephaloceles are classified based on the location of the defect in the cranial bones.<sup>7</sup> Suwanwela *et al*<sup>8</sup> classified anterior basal meningoencephaloceles into four subgroups: 1) transethmoidal, 2) sphenothmoidal, 3) transsphenoidal and 4) frontosphenoidal or sphenoorbital. The fronto-ethmoidal region of the anterior skull has been identified in prior studies as being the area most commonly impacted. According to a theory, the rostral neuropore, which is the last portion of the neural tube to seal, is prone to aberrations throughout cranial development.<sup>7</sup>

Intranasal meningoencephaloceles can only be treated surgically. Transmaxillary route was used to execute the first successful intranasal meningoencephalocele surgery. An endoscopic transnasal technique can be considered to reduce surgical risks if the defect in the skull base is minimal.<sup>2</sup> For Endoscopic Endonasal Repair of Meningoencephalocele, a 'Bath-plug' technique has been reported in literature<sup>9</sup> and it entails inserting a fat plug with a specially secured vicryl suture into the intradural space and applying traction on the suture to seal the defect similarly to how a bath plug seals a bath. The 'Bath-plug' technique for closure of anterior skull base CSF leaks is a reliable technique for a large variety of cases.<sup>10</sup>

## CONCLUSION

A congenital defect of the skull base, including meningoencephalocele, should be taken into

consideration when a patient has recurrent intracranial infection but no history of immunodeficiency, cranial trauma, or neurosurgical operation. To prevent improper management, it is crucial that ENT colleagues, to whom these cases primarily present, are well informed about this unusual condition. For the purpose of preventing further neurological problems, thorough neuroimaging evaluations and successful surgical repair are important. The "Bath-plug" technique for anterior skull base CSF leak repair is effective for a wide range of situations.

## Abbreviations:

CT: Computed Tomography;

MRI: Magnetic Resonance Imaging;

FESS: Functional Endoscopic Sinus Surgery;

CSF: Cerebrospinal fluid.

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