Cribriform plate dehiscence and encephalo-meningocele may not be the only cause of recurrent bacterial meningitis

Dear Editor,

We read with interest the article by Soyah et al. about a 9-year-old male with cribriform plate dehiscence and encephalo-meningocele, who presented clinically twice with bacterial meningitis due to Streptococcus pneumoniae [1]. Recurrence of bacterial meningitis could be prevented by endoscopic repair of the defect [1]. It was concluded that recurrent bacterial meningitis requires examination by computed tomography (CT) or magnetic resonance imaging (MRI) and that, if due to a congenital skull base defect, surgical repair may prevent recurrence of the infection [1]. The study is impressive, but some points require further discussion.

The first point is that the assumption that bacterial meningitis arose from migration of bacteria via a fistula from the epipharynx into the subarachnoid space has not been confirmed [1]. Rhinorrhoea can be expected in cerebrospinal fluid (CSF) fistulas from the subarachnoid space to the epipharynx [2], which was not reported by the index patient. In addition, the patient did not undergo a flourescine test to document CSF leakage into the epipharynx.

The second point is that the neurological exam was reported as normal, but at least hyposmia or anosmia could be expected in a patient with congenital dehiscence of the cribriform plate and encephalo-meningocele [3]. In addition, impairment of the hypothalamus or the pituitary gland may occur. Have the pituitary hormones been analysed and were they all within normal limits?

The third point is that tuberculosis has not been ruled out as an underlying pathology causing bacterial meningitis [4]. We should know whether the Quantiferon test was positive and whether the family history was positive for tuberculosis or not.

The fourth point is the discrepancy between the cerebral CT and the cerebral MRI findings. It is reasonable to assume that an encephalo-meningocele of the size shown in figure 1 should also be visible on cerebral CT. How do the authors explain this discrepancy?

The fifth point is that no long-term follow-up was carried out. It should be specified in detail until when the patient actually remained free of meningitis recurrence. Has the meningitis recurred since the case was published?

The sixth point is that it is not only the skull or the meninges that prevent the infection of the brain, but also the blood brain barrier and the cerebral immune system. Therefore, it is conceivable that recurrent meningitis was not due to a structural skull base defect, but rather due to impaired immune system or blood brain barrier impairment. Was there evidence for immunodeficiency in the index patient?

In summary, the excellent study has limitations that should be addressed before drawing final conclusions. Clarifying the weaknesses would strengthen the conclusions and could improve the study. Before attributing recurrent bacterial meningitis with streptococcus pneumonia to encephalo-meningocele, alternative routes of infection should be considered and thoroughly ruled out.

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AUTHOR'S ANSWER

Recurrent pneumococcal meningitis in children: Diagnostic dilemma

Dear Editor,

We want to express our sincere gratitude for the interest in our recent article on the 9-year-old male with cribriform plate dehiscence and encephalo-meningocele, who experienced recurrent bacterial meningitis due to Streptococcus pneumoniae. We welcome the opportunity to address the points raised by Finsterer and Mehri [1] to

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further clarify our findings and conclusions.

Firstly, regarding the assumption that bacterial meningitis arose from the migration of bacteria via a fistula from the epipharynx into the subarachnoid space, we agree that direct evidence of such a pathway was not documented. While rhinorrhea is a common sign of cerebrospinal fluid (CSF) fistulas, its absence does not preclude the presence of a fistula [2]. While a fluorescein test could have provided more definitive evidence of CSF leakage, clinical and radiological findings were strongly suggestive of the pathophysiological mechanism proposed.

Regarding the second point about the neurological exam, it is indeed true that hyposmia or anosmia would typically be expected in a patient with congenital dehiscence of the cribriform plate and encephalo-meningocele. Assessing hyposmia or anosmia in young children can be difficult. However, our patient's clinical evaluation did not reveal any olfactory impairment. We did not assess pituitary function due to the absence of clinical signs indicative of hypothalamic or pituitary dysfunction.

In this case, the patient's presentation did not align with the typical course of tuberculous meningitis (TBM), which often involves a subacute to chronic onset, cranial nerve involvement, and basilar meningitis features [3]. Also, characteristic CSF findings of TBM include a lymphocytic-predominant pleiocytosis. The acute symptoms and initial tests were more consistent with common bacterial meningitis, making tuberculous meningitis unlikely. Furthermore, the patient responded well to standard empirical antibiotic therapy for bacterial meningitis, which supports the initial diagnosis.

The fourth point raises a valid question regarding the apparent discrepancy between the cerebral computed tomography (CT) and Magnetic Resonance Imaging (MRI) findings. Several factors may contribute to this discrepancy. First, MRI is generally more sensitive and provides higher resolution images of soft tissues compared to CT. MRI can detect subtle abnormalities in brain tissue and cerebrospinal fluid spaces that may not be as apparent on CT [4]. Second, axial cranial computed tomography may fail to identify defects in the basal ethmoidal area and cribriform plate, whereas coronal thin sections show detailed anatomy of the anterior cranial fossa and identify most skull defects. In our case, an urgent cerebral CT scan was performed to rule out contraindications for lumbar puncture, possibly explaining the absence of visualization of this congenital defect.

As for the follow-up, the patient has been monitored for two years post-surgery and has remained free of meningitis recurrence during this period. We agree that longer follow-up would be beneficial to definitively assess the success of the surgical intervention.

Lastly, you are absolutely correct in noting that the protection of the brain against infections is not solely dependent on the structural integrity of the skull and meninges. The blood-brain barrier and the cerebral immune system play crucial roles in safeguarding the central nervous system from pathogens. We initiated investigations to search for a skull base defect because the organisms commonly associated with meningitis due to CSF leak include S. pneumoniae [5]. Furthermore, the patient showed no clinical signs of immunodeficiency, and basic immunological tests, such as complete blood counts, serum immunoglobulin levels, and complement pathway tests, were all normal. However, a comprehensive immunological assessment was not conducted

In summary, we appreciate the constructive feedback which highlights important considerations and limitations of our study. We hope our response addresses the concerns raised and contributes to a deeper understanding of the case.

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