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Case Report

Cerebrospinal fluid otorrhea masquerading as cerebrospinal fluid rhinorrhea: a rare presentation

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ABSTRACT

Congenital inner ear disorders are associated with cerebrospinal fluid (CSF) leaks and recurrent meningitis. Here we present a case report of a 47-year-old male patient who presented with CSF rhinorrhea and recurrent meningitis with an unidentified congenital inner disorder, which was later identified as CSF otorrhea, and had leak repair done. In this paper, we stress considering CSF otorrhea as a differential diagnosis while dealing with the presentation of recurrent meningitis.

Keywords: CSF rhinorrhea, Otorrhea, Congenital inner ear dysplasia

INTRODUCTION

Cerebrospinal fluid (CSF) leak originating from temporal bone is a result of abnormal communications that exist between subarachnoid space and temporal bone air spaces. The clinical presentation of the patient depends on the condition of tympanic membrane and eustachian tube function. In an intact tympanic membrane, the leak occurs from nose and is called as otorhinorrhea whereas in a perforated tympanic membrane patient presents with CSF leak from ear which is called as CSF otorrhea.[1] Congenital inner ear dysplasias can lead to CSF leakage and recurrent meningitis in addition to severe to profound sensorineural hearing loss.[1]

CASE REPORT

A 47 year old male patient was referred to ENT from neurosurgery department of our hospital, with complaints of clear watery discharge from nose since 1 month while sitting upright and bending forward, no history of trauma. He had 2 episodes of meningitis 1 month back. Diagnostic nasal endoscopy was done and showed frank watery discharge; however, site of lesion was not identified. NCCT paranasal sinuses and MRI brain showed CSF intensity along the anterior aspect of wall of left sphenoid sinus and posterior ethmoid sinus and tracking along the nasal cavity and left maxillary sinus in prone position. Patient was taken for Endoscopic endonasal CSF leak repair under GA, but the leak site could not be identified. Intraop fluorescein dye injected to visualise the leak site, and CSF leak visualised from left eustachian tube filling into nasal cavity. As the consent for CSF otorrhea repair was not taken, patient was reversed from GA and shifted to ward. HRCT temporal bone was performed.

On further examination, patient revealed left sided hearing loss from childhood. Pure tone audiometry was performed which showed profound hearing loss in left side. HRCT temporal bone showed common cavity anomaly in left side with fluid density opacification of middle ear cavity and mastoid air cells.

Patient was taken up for left mastoid exploration and CSF leak repair under GA. Post aural approach was done. Tympanic membrane was found intact. Fluorescein-stained CSF leak identified from oval window with a floating stapes footplate. Facial nerve was found to be intact. Eustachian tube area covered with harvested temporalis muscle and fascia, hypotympanum and round window plugged in similar fashion. Stapedectomy done.

CSF seen gushing from oval window area repaired with temporalis fascia and muscle in layers followed by gelfoam and tissue glue application as in a multilayer fashion.

Post op, patient was continued on oral antibiotics. After 1 week, dressing and sutures removed.

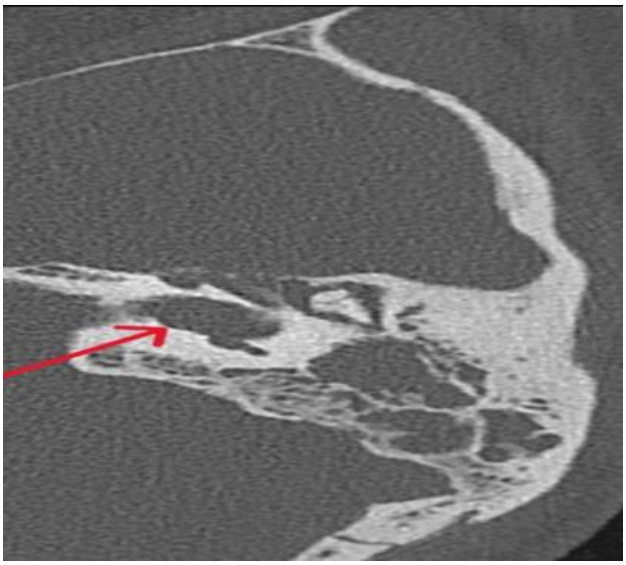

Figure 1: Axial view of HRCT left temporal bone showing common canal deformity.

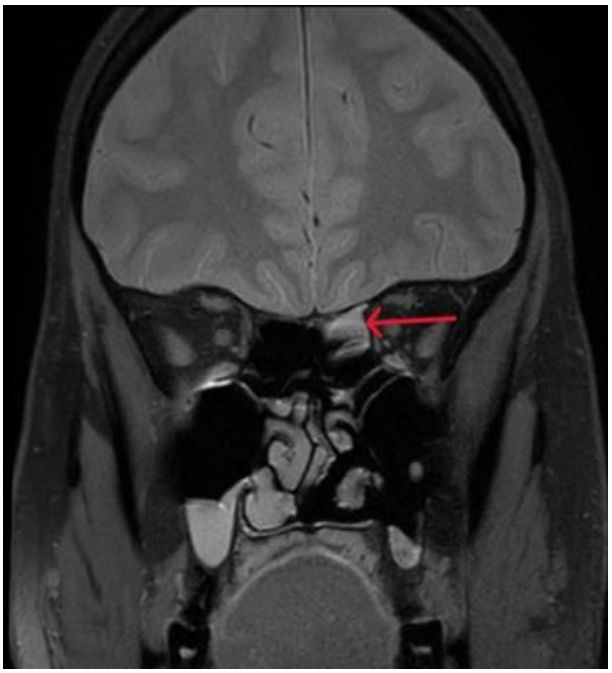

Figure 2: Coronal view of MRI brain showing CSF in left sphenoid sinus.

DISCUSSION

The CSF otorrhea associated with inner ear dysplasias are commonly seen in paediatric age group, mostly before the age of 10. The presentation is very rare in adults, however few cases have been reported world-wide.[6] Our case becomes significant in this background, as the patient is an elderly male with no prior hearing abnormality presented with recurrent meningitis and watery discharge from nose. The CSF otorrhea was not identified in the first visit considering patients age and the unavailability of a history of hearing loss. The chance of developing bacterial or viral meningitis in those with a congenital inner ear disorder is 93% in children and only 38% in adults.[7]

There have been many classifications for congenital ear malformations, but the most followed one is by Sennaroglu.[2] According to the literature, these inner ear malformations can be divided into eight groups.[3,4] Our patient had a common cavity malformation which accounts for about 0.7-26% of cochlear malformations.[5] CSF leak associated with congenital inner ear malformations could be due to the presence of abnormal communications between subarachnoid space and inner ear.

The leak site was identified to be oval window in our patient, which including the stapes footplates and the annular ligament is usually the most common site of leakage in these cases. Inner ear malformations are usually associated with otic capsule abnormalities which can lead to stapes footplate or annular ligament weakness. Also, the continuous pressure variations of CSF can lead to stapes footplate or annular ligament thinning.[8]

HRCT temporal bone is an ideal investigation tool for CSF otorrhea as it identifies CSF accumulation in tympanic or mastoid cavity as a soft tissue shadow. In those patients, who are presenting with hearing abnormalities with CSF leak, HRCT can be advised as a relevant investigation tool.[8] Audiological tests should also be done hand in hand to identify hearing loss.

The gold standard treatment for CSF otorrhea in congenital ear disorders remains as surgery. The choice of surgery depends on many factors like site of leakage, intraoperative findings and hearing levels of patient.[9] Different surgical approaches have been utilised by surgeons all over which includes transmastoid approach, middle cranial fossa approach or a combinatin of both.[8] We have done a postaural transmastoid approach for the leak repair. For the leaks arising due to oval window fistula, it is a practice to remove stapes footplate and surrounding mucosa and covering the area with temporalis muscle and fascia.[8] In our case, as we encountered a floating footplate, stapedectomy was performed and the oval window area was closed with temporalis muscle and fascia. It is better to do a multilayer repair to avoid recurrences.[8] We have done a multilayer repair which includes temporalis muscle and fascia followed by surgical and tissue glue. In addition,

eustachian tube opening also was plugged with temporalis fascia.

Early diagnosis of congenital inner ear disorder becomes a necessity as it can lead to complications like CSF otorrhea which when undiagnosed can in turn lead to recurrent meningitis.[8] In our patient, congenital inner ear disorder itself was not diagnosed, hence CSF otorrhea was unidentified which leads to development of recurrent meningitis.

CONCLUSION

Congenital inner ear dysplasia are associated with CSF otorrhea and recurrent meningitis. An adult patient with unidentified congenital inner ear dysplasia when presented with CSF rhinorrhea, CSF otorrhea was missed. Hence in all patients presenting with CSF leak or recurrent meningitis, hearing evaluation should also be advised to rule out the possibility of CSF otorrhea. Early diagnosis is also necessary to prevent recurrent episodes of meningitis.

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