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Post-Traumatic Pseudomeningocele Presenting as a Pulsatile Cyst of the External Auditory Canal

Case Report

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Abstract

Pseudomeningocele is a of cerebrospinal fluid filled, extracranial cystic collection resulting from a dural defect, which may occur congenitally, postoperatively, or after trauma. Post-traumatic pseudomeningocele is rare, particularly in the temporal bone region. We report a 37-year-old woman who presented with progressive hearing loss and intermittent otorrhea, two decades after head trauma. Otoscopy revealed a pulsatile cystic lesion in the right external auditory canal. Imaging showed a ~20 mm tegmen tympani defect with herniation into the tympanomastoid area. Surgical repair involved transmastoid excision of the sac and multilayer reconstruction of the tegmen defect with mastoid obliteration. This case emphasizes the need to consider skull base defects in patients with a history of head trauma and persistent otologic symptoms.

Keywords: Ear surgery, hearing loss, pseudomeningocele, cerebrospinal fluid, temporal bone, otorrhea, case report

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Introduction

Pseudomeningocele is a false sac that does not contain brain tissue and is formed when cerebrospinal fluid (CSF) leaks from a dural defect and accumulates in the extracranial space after congenital, surgical, or traumatic events. The sac wall consists of a fibrous structure. It does not contain meninges. There is no cranial tissue inside the sac, only CSF. Its treatment is possible through a surgical repair of the defect. The causes of pseudomeningocele can be categorized into three main groups: congenital, iatrogenic, and traumatic. Congenital cases are generally associated with genetic diseases such as Marfan syndrome and neurofibromatosis (1). Common iatrogenic conditions often result from inadvertent dural tears during spinal surgery. Finally, pseudomeningocele can also occur rarely after head and spinal traumas. Post-traumatic pseudomeningocele cases in the ear are even rarer. Upon conducting a literature review on post-traumatic pseudomeningocele cases found in the external auditory canal (EAC), only two cases were identified (1,2). In conjunction with relevant literature data, we will present the case study of this particular occurrence.

Case Presentation

A 37-year-old female patient presented with complaints of hearing loss and discharge in the right ear. Her medical

history revealed that her symptoms began following a head trauma sustained approximately 20 years ago while living in her home country. The patient did not seek medical attention at the time, and therefore no medical reports or imaging studies from that period are available. The trauma reportedly involved a horizontal compression of the head from both sides, primarily affecting the superior region of the right temporal bone. The patient reported a gradual worsening of hearing loss over the years, along with intermittent episodes of discharge in the right ear. There is no history of meningitis during this period.

Otoscopic examination of the patient revealed that the right EAC was obliterated by a pulsatile, cystic lesion, and the tympanic membrane could not be visualized. No abnormal findings were observed during the Valsalva maneuver (VM). Pure-tone audiometry demonstrated a moderate conductive hearing loss in the right ear (Figure 1). Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a minimal loss of parenchyma in the right temporal lobe, which was considered to be trauma related. Decreased aeration of the mastoid air cells in the right ear was noted, and a defect measuring up to 20 mm in the tegmen tympani was observed, particularly in sagittal sections (Figure 2). CSF was seen extending into the tympanomastoid cavity and EAC. However, no brain parenchyma was identified within the cystic formation. Based on the CT findings and the homogeneous, low T1 signal intensity and continuity of the CSF on MRI, a preliminary diagnosis of pseudomeningocele was considered (Figure 3). Additionally, evaluation of the ossicular chain revealed absence of the incus.

Based on clinical and radiological findings, the pathology was considered to be a pseudomeningocele, and surgical intervention was planned. Under general anesthesia, a mastoidectomy was initiated using a retroauricular transmastoid approach. Upon reaching the antrum, a pulsatile cystic formation extending from the antrum to the tympanic cavity was observed. The cystic structure was followed, and the mastoidectomy was continued. The lesion was found to originate from a defect of approximately 20 mm in the tegmen tympani (Figure 4). Evaluation of the ossicular chain revealed that the stapes was mobile and located beneath the cystic lesion. The malleus was found anterior to the lesion, transposed and disconnected. The incus was not observed and was presumed to have undergone secondary erosion due to the pressure exerted by the lesion. The cystic structure was excised, and no brain parenchyma was identified within it.

To prevent CSF leakage, approximately 2×1 cm of temporalis muscle fascia and composite muscle grafts were harvested. A muscle graft was first placed to completely cover the bony defect, followed by placement of a fascia graft. Subsequently, a superiorly based palva flap was positioned over the fascia graft and reinforced with fibrin glue. Although the materials used were sufficient to repair the tegmen tympani defect, due to the size of the defect and the risk of potential CSF leakage, mastoid obliteration was performed. The mastoid cavity was obliterated using bone cement and bone pate. After removal of the pathological mucosa in the middle ear, a tragal cartilage graft was placed beneath the residual tympanic membrane, positioned over the mobile stapes. Following this, a lumbar drain was placed by the neurosurgery team. The operation was concluded, and the lumbar drain was removed on postoperative day six.

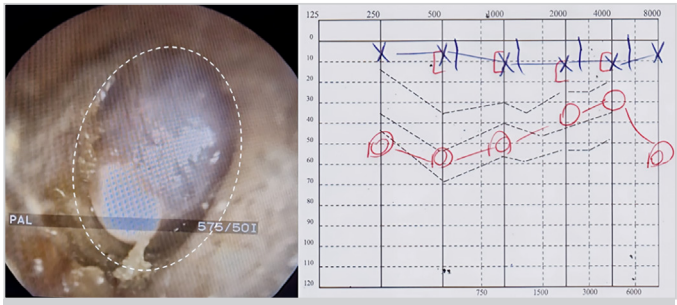

Figure 1. Preoperative endoscopic examination (left) and audiological assessment (right) of the patient

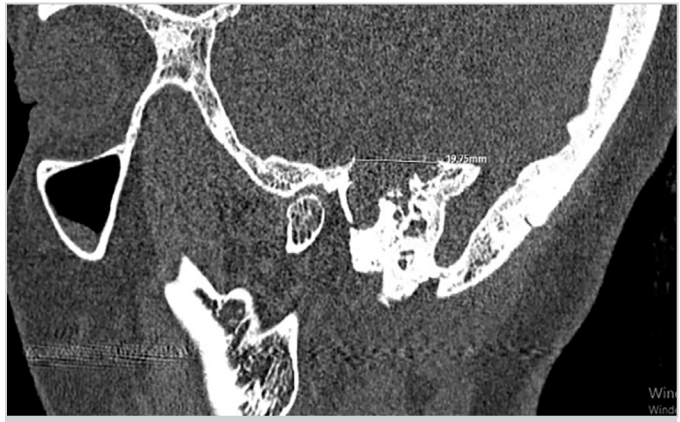

Figure 2. Sagittal section of temporal bone computed tomography showing a bony defect in the tegmen tympani measuring 19.75 mm

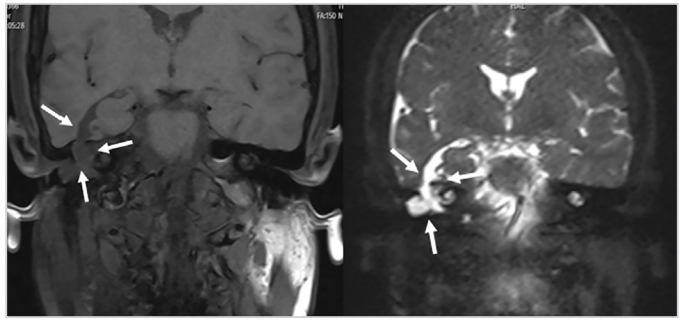

Figure 3. Coronal magnetic resonance images showing a homogeneously cystic lesion, hypointense on T1-weighted (left) and hyperintense on T2-weighted (right) sequences. The lesion extends from a tegmen defect into the middle ear and external auditory canal, suggestive of a pseudomeningocele

Histopathological examination revealed pseudocystic structures consistent with the preliminary diagnosis. No neural tissue was identified. At the patient's one-year postoperative follow-up, the graft was found to be intact. Pure-tone audiometry demonstrated a significant improvement in the air-bone gap (Figure 5). Informed consent was obtained from the patient.

Discussion

Pseudomeningocele is a persistent condition defined by the extradurally accumulated CSF, resulting from leakage through a dural discontinuity. It can be incidentally detected. Pseudomeningocele is typically iatrogenic, but traumatic and congenital causes can also occur. Examples of iatrogenic causes include direct trauma to the dura, excessive traction, or lacerations from bone fragments during extradural surgeries such as laminectomy (3). Among the congenital causes of pseudomeningocele are diseases such as neurofibromatosis and Marfan syndrome. Individuals with neurofibromatosis and Marfan syndrome exhibit a more elastic dural layer. Consequently, they are more susceptible to pseudomeningocele formation as a result of increased vulnerability to damage in the event of any injury. Additionally, congenital pseudomeningocele is often associated with defects in vertebral bodies (3).

Taveras and Ransohoff (4) reported that the development mechanism of pseudomeningocele involves the compression of the dura mater by pulsatile cerebral pressure following a dural opening caused by head trauma, leading to the leakage of CSF into the subcutaneous space through the damaged dura mater. In cases of dura mater damage, a spherical valve-like mechanism inhibits the physiological outflow of the CSF, resulting in progressive cyst enlargement and the development of a fibrous capsule (5). Traumatic pseudomeningocele can occur in various ways. In the case presentation by Scott and Merrell (5), a pseudomeningocele developing over time in the EAC following a skull fracture was described. In this presentation, they noted that the patient's symptoms and signs of chronic otitis media began following a previous head trauma.

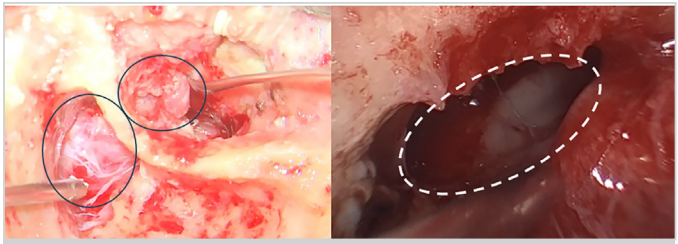

Figure 4. Intraoperative images showing a lesion extending from the mastoid cavity into the external auditory canal (left), and a tegmen defect (right)

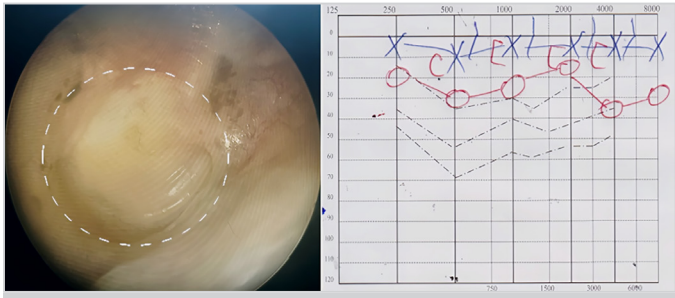

Figure 5. Postoperative one-year endoscopic examination (left) and audiological assessment (right) of the patient

Diagnosis can be established through a detailed medical history, physical examination, and radiological imaging. In the medical history, suspicion for pseudomeningocele or meningocele should be raised in patients with a history of head trauma or traffic accidents, those presenting with spontaneous CSF otorrhea, individuals with conductive hearing loss confirmed by audiologic evaluation particularly when accompanied by pulsatile tinnitus as well as patients exhibiting symptoms of meningitis or presenting with a cystic lesion that changes in size with the VM (6). In the literature, two similar cases have been reported in which physical examination revealed non-pulsatile lesions and no changes with the VM. In contrast, the lesion in our case was pulsatile. One of the previously reported cases demonstrated conductive hearing loss, while the other had sensorineural hearing loss. Radiological imaging in both cases revealed a defect in the tegmen with a continuity extending into the mastoid and middle ear cavities (1,2).

Radiological diagnosis can be achieved through CT, which may reveal a defect in the tegmen, and MRI, which can demonstrate continuity of brain tissue extending into the tympanic cavity or mastoid antrum (7). Additionally, CSF typically exhibits a homogeneous, fluid-like appearance on MRI, characterized by low signal intensity on T1-weighted images. Previous studies have reported that most pseudomeningocele do not exert a mass effect on the thecal (dura/dural) sac, as they are generally continuous and isobaric in nature. The combined use of CT and MRI has been shown to yield an accurate initial diagnosis in approximately 89% of the cases (8-10). In the differential diagnosis, true meningocele or meningoencephalocele should be considered. Evaluation should include determining whether the lesion is pulsatile or changes in size with the VM. Furthermore, MRI is essential for assessing the location and extent of the cystic lesion (3).

The defect responsible for pseudomeningocele can be repaired using either a transcranial or a transmastoid approach. The transcranial (craniotomy) approach offers advantages such as superior visualization of the defect and easier manipulation of the temporal lobe. However, its main drawback is its invasiveness and the associated risk of cranial

complications. As a less invasive alternative, the extracranial transmastoid approach enables access to the defect through a retroauricular incision and mastoidectomy (4). In the first of the two previously reported cases, a similar transmastoid approach was employed; however, mastoid obliteration was not performed due to the small size of the defect and the absence of pulsation. In the second case, surgical intervention was not undertaken as the patient declined surgery (1,2). Various grafting materials may be used in both surgical approaches, including temporalis muscle, temporalis fascia, autologous cartilage, bone tissue, and fat. To date, no definitive superiority among these materials has been established in the literature. The key factor in achieving successful surgical outcomes is the implementation of a multilayer repair technique. Single-layer repairs have been associated with poor long-term outcomes. In contrast, the use of at least two graft layers whether autologous, synthetic, or a combination of both has significantly improved the success rate of primary closure. Moreover, to prevent CSF otorrhea, it is advisable to seal any exposed intracranial air cells or small bony dehiscence in the tegmen during the initial surgical procedure. In cases where CSF leakage is present, obliteration of the mastoid air cells may be beneficial to ensure closure of multiple or unidentified leak sites (11,12).

In our case, we opted for the less invasive extracranial transmastoid approach. Due to the extensive size of the defect, multilayer repair was performed using a temporalis muscle graft, temporalis fascia graft, superiorly based Palva flap, and fibrin glue, followed by mastoid obliteration.

Conclusion

The detection of a pulsatile, cystic mass in the EAC associated with hearing loss, particularly in patients with a history of previous otologic surgery or head trauma, should raise clinical suspicion of pseudomeningocele, a rare pathological condition.

Ethics

Informed Consent: Informed consent was obtained from the patient.

Footnotes

Authorship Contributions

Surgical and Medical Practices: O.Ö., A.Ç., K.H., Ö.Y., Concept: O.Ö., A.Ç., K.H., Design: O.Ö., K.H., Data Collection and/or Processing: O.Ö., A.Ç., Ö.Y., Analysis and/or Interpretation: O.Ö., K.H., Literature Search: E.C.Ö., E.D., Writing: O.Ö., A.Ç.

Conflict of Interest: The authors declare that they have no conflict of interest.

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Main Points

- Post-traumatic pseudomeningocele is an extremely rare condition.
- It is a sac deformity in which cerebrospinal fluid leaks through a dural defect and collects in the extradural space. The collection is not a true arachnoid-lined sac and does not contain brain tissue.
- The patient may present to the clinic with a pulsatile cyst in the external auditory canal and conductive hearing loss. Definitive treatment involves surgical repair of the defect.

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