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A Rare Case of Tubercular Cholesteatoma with TB Meningitis

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Abstract

Tubercular otitis media is an uncommon condition. Tuberculosis can affect any part of ear ranging from tympanic membrane to labyrinth. The symptoms of tubercular otitis media like painless otorrhea, multiple perforations, pale granulations, facial paralysis and severe SNHL though well described in literature, are not always present hence diagnosis is often missed. Diagnosis is usually made by clinical and histopathology examination of specimen obtained intra operatively. Here, we discuss clinical presentation, diagnosis and management of a case with cholesteatoma and TB meningitis.

Keywords

Tuberculosis, Cholesteatoma, Meningitis

1. Introduction

The incidence of tubercular otitis media ranges from 0.05% to 0.9% of all cases of chronic otitis media [1]. This is one of rarest forms of extrapulmonary tuberculosis and is often a missed diagnosis. There are no definite or pathognomic clinical signs and symptoms, making the diagnosis difficult in absence of laboratory findings [2].

Cholesteatoma in middle ear is not uncommon but presence of tubercular bacilli makes it a rare presentation. Tuberculosis should be included in the differential diagnosis of chronic middle ear infections not responding to routine management. We present an interesting case of tubercular otitis media with intracranial complication.

2. Case report

43 years old male presented to us with complaints of fever and left ear discharge for 20 days. He had history of similar discharge from left ear for which he un-
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The patient underwent surgery 10 years back. He also gave a history of right ear surgery for discharge 5 years back. This patient was receiving treatment by physician for last 20 days with no significant improvement. Patient also had hypertension for last 4 years and Diabetes Mellitus for 5 years for which he was on regular medication. On our examination, patient was febrile with PR 98/min and BP 150/100 mm of Hg. There was associated neck pain and rigidity. Kernig’s sign was positive. Ophthalmologic examination showed right lateral rectus palsy. Detailed ENT examination was done which showed bilateral inadequate conchomeatoplasty. Right ear had healthy dry mastoid cavity with mobile neotympanum while left ear showed wet cavity with purulent discharge and cholesteatoma debris. Ossicles were not visualised. Facial nerve function was normal bilaterally. Pure tone audiometry showed 63.3 dB conductive deafness on right side and 65 dB mixed deafness on left side.

Considering the history that meningitis did not respond to high dose antibiotics (I/V Ceftriaxone 2 gm 8 hourly, IV Vancomycin 1 gm 12 hourly, IV Metrogyl 100 ml TDS) we suspected the disease to be of non pyogenic cause. MRI brain with venogram was done, which showed evidence of 1.3 × 1.6 cm thick walled peripherally enhancing abscess in left cerebellum with mild surrounding oedema (Figure 1). Left sigmoid sinus was compressed, however patent. CSF examination was done which was suggestive of chronic meningitis. Hence patient was planned for urgent left ear mastoid exploration. Revision mastoidectomy done. Erosion of horizontal and posterior semicircular canal noted. Facial nerve canal was dehiscent near second genu. There was large dehiscence of Posterior fossa dural plate. We were able to drain the posterior fossa extradural abscess through Trautman’s triangle. Pus, pale granulation tissue and cholesteatoma sac obtained intra operatively was sent for culture sensitivity, AFB and histopathology examination.

Pus culture and cholesteatoma matrix showed Staph aureus and AFB in many fields. Histopathology examination also showed presence of caseating granulomas with AFB in matrix (Figure 2). Fluorescent staining of cholesteatoma matrix was done, which also demonstrated AFB (Figure 3). Hence diagnosis of cholesteatoma with TB and TB Meningitis was made. AKT was started. Patient was followed up after 15 days. Symptoms dramatically improved with marked reduction in neck rigidity and diplopia. Lateral rectus palsy recovered fully within 3 weeks post operatively.

3. Discussion

Tubercular otitis media is a very unusual cause of chronic otitis media, and is rarely considered in the differential diagnosis. Although the pathogenesis of TOM is still controversial, three mechanisms explaining middle ear tuberculosis infection have been postulated: aspiration of mucus through the auditory tube, hematogenous transmission from other tuberculosis foci and direct implantation through the external auditory canal with tympanic membrane perforation [3] [4] [5].
Figure 1. MRI brain showing 1.3 × 1.6 cm thick walled peripherally enhancing abscess in left cerebellum with mild surrounding oedema.

Figure 2. H & E staining of cholesteatoma matrix showing caseating granulomas with AFB.

Figure 3. Fluorescent staining of cholesteatoma matrix showing AFB.
Like any other disease, complications of tubercular otitis media occur when there is delay in diagnosis. Complications can be intracranial or extracranial. Intracranial complications include meningitis, abscess (subdural and extradural) and otitic hydrocephalous. Intracranial extension of the disease can be direct through erosion of bony anatomical barrier that is tegmen tympani or hematogeneous along sigmoid sinus.

Here, our patient had minimal vestibular symptoms despite erosion of lateral and posterior SCC. There was no facial palsy despite significant dehiscence of Facial canal. Also intracranial symptoms were minimal in spite of a large dural defect.

Complications can be prevented only if the diagnosis is made at early stage of the disease. Tuberculosis should be suspected when there are persistent symptoms not responding to routine antibiotics. Clinicians should have strong suspicion especially in a country with high incidence of this systemic disease.

4. Conclusion

The main objective behind this article is to make ENT surgeon vigilant about this rare manifestation of very common disease in the Indian sub continent. Tuberculosis should be included in the differential diagnosis of chronic otitis media not responding to routine therapy or when presenting with intra cranial complication. Early diagnosis and timely initiation of anti tubercular therapy can prevent life threatening complications.

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Spontaneous Uterine Artery Pseudoaneurysm Rupture Following Cesarean Section: A Case Report

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Abstract
A pseudoaneurysm, also known as false aneurysm, is a collection of blood in perivascular soft tissue. Iatrogenic cause such as arterial catheterization and abdominopelvic surgery is one of the most common etiologies of pseudoaneurysm formation. Pseudoaneurysm in pelvic vessels is a complication of pelvic surgery or vessel catheterization. Cesarean section is the most common reported cause. In this article we introduced a 25 years old woman presented with abdominal pain and vaginal bleeding 1 week after Cesarean section. Imaging modalities confirmed the diagnosis of left uterine artery pseudoaneurysm. Patient was admitted to treat by endovascular intervention. Several hours after admission blood pressure decreased and abdominal pain became more sever, ultra sound study showed evidence of free fluid in abdominopelvic cavity suggestive of pseudoaneurysm rupture. Emergence laparotomy surgery and hysterectomy were done due to active bleeding secondary to pseudoaneurysm rupture. Patient was discharged from hospital 5 days after surgery without any new complication.

Keywords
Pseudoaneurysm, Ultrasound, Cesarean Section

1. Introduction
A pseudoaneurysm, also known as a false aneurysm, is a collection of blood around vessel which is confined by the adventitia or perivascular soft tissue. One of the most common etiologies of pseudoaneurysm formation is iatrogenic cause, for example arterial catheterization and abdominopelvic surgery. And blunt or
penetrating trauma, vasculitis, regional inflammatory process (for example acute pancreatitis) and fibromuscular dysplasia are other etiologies of formation a pseudoaneurysm [1] [2]. Pseudoaneurysm occurs most commonly in arteries but there are some reports of pseudoaneurysm formation in vein [3].

Pseudoaneurysm in pelvic vessels is rare but reported a complication of pelvic surgery or vessel catheterization. Cesarean section is the most common reported cause but this complication has also been reported in association with abortion, repeated curettage, myomectomy, hysterectomy, uncomplicated vaginal delivery and even oocyte retrieval for IVF [4] [5].

Uterine artery pseudoaneurysm may be asymptomatic or may thrombose spontaneously but patients usually present as delayed postpartum hemorrhage which can occur up to several weeks in postpartum period and can cause life threatening bleeding that need emergency therapy [6] [7].

Different imaging modalities include ultrasound, computed tomography, MRI and angiography can be applied to confirm diagnosis.

In color ultrasound study the “yin-yang” sign indicates turbulent blood flow within the false aneurysm sac. The pulse Doppler study shows that “to and fro” pattern, which is obvious in communicating channel or neck of pseudoaneurysm, is not the sac. The “to” component is caused by enter of blood during systole and the “fro” component is seen during diastole when the blood stored in cavity is ejected in to the artery. Color Doppler ultrasound has sensitivity of 94% and specificity of 95% in different parts of the body for diagnosis of pseudoaneurysm [8].

Pseudoaneurysm in CT is demonstrated as hypoattenuating (non-contrast) or hyperattenuating (contrast-enhanced) smooth walled sac adjacent to a vessel and contrast enhanced MRI which shows high signal sac of pseudoaneurysm [1].

Uterine artery embolization is preferred treatment but ligation of affected uterine artery by surgery and hysterectomy are alternative treatments.

In this case report we present a uterine artery pseudoaneurysm diagnosed one week after Cesarean section and ruptured before endovascular therapy.

2. Case Presentation

A 25 years old gravid 2 woman presented with abdominal pain and vaginal bleeding 1 week after Cesarean section surgery. She was asymptomatic during 1 week after operation. Mild tenderness in left lower quadrant on abdominal examination was detected. Laboratory data showed Hb: 10.5 and WBC count 10,000 at time of admission. Blood pressure and temperature was 110/70, 37/5 respectively.

In Ultrasound study there was a round hypoecho mass measured 4 cm with echo free center in left adnexa. Color Doppler demonstrated typical “yin-yang” sign in center of mass which is characteristic for pseudoaneurysm (Figure 1).

Pulse Doppler at the neck of mass showed “to and fro” pattern (Figure 2).

Following ultrasound study CT angiography of pelvis and MRI was requested to confirm diagnosis of pseudoaneurysm.
Figure 1. Saggital ultrasound image of uterus use concurrent color Doppler demonstrates typical “ying-yang” sign at left adnexa characteristic for pseudoaneurysm (arrow), (U) uterus.

Figure 2. Pulse Doppler at the neck of pseudoaneurysm shows typical “to and fro” pattern, to component (small arrow), fro component (large arrow).

CT angiography showed a collection of contrast adjacent to left internal iliac artery suggestive of pseudoaneurysm (Figure 3). Contrast enhanced MRI demonstrated enhanced center of pseudoaneurysm and peripheral hematoma (Figure 4).
Figure 3. CT angiography of pelvic arteries using volume rendering technique demonstrates contrast pooling at the pseudoaneurysm site (arrow).

Figure 4. Axial T1 weighted fat suppressed contrast enhanced MRI demonstrates enhanced center of pseudoaneurysm and peripheral hematoma (arrow), (U) uterus.

Patient was admitted to treat by endovascular intervention. Several hours after admission blood pressure decreased and abdominal pain and vaginal bleeding became more severe. Ultrasound study showed evidence of free fluid in abdominopelvic cavity which was suggestive of pseudoaneurysm rupture. Although hysterectomy is the last choice in treatment of uterine artery pseudoaneurysm, due to rupture of pseudoaneurysm and because ligation of internal iliac artery was not effective, hysterectomy was done to control bleeding. Patient was dis-
charged from hospital 5 days after surgery without any new complication.

3. Discussion

Pseudoaneurysm in pelvic vessels is a rare complication of pelvic surgery. Cesarean section is the most common cause but this complication has also been reported in association with abortion, repeated curettage, myomectomy, hysterectomy and even uncomplicated vaginal delivery [4].

Ultrasound, CT angiography, MRI and angiography are modalities applied to diagnosis of this complication [1].

Pseudoaneurysm of uterine artery is a rare cause of delayed postpartum hemorrhage. Delayed postpartum hemorrhage is defined as bleeding from 24 hours after delivery up to 6 weeks postpartum and most commonly occurring between 8 and 14 days postpartum. Retained products of conception, subinvolution of the placental bed and endometritis are more common causes of delayed postpartum hemorrhage [9].

Treatment includes ligation of uterine artery by surgery or endovascular embolization but endovascular embolization of involved artery is preferred treatment. Hysterectomy is the last choice if uterine artery ligation by surgery or embolization is not effective to control bleeding [4]. The first case of selective arterial embolization that used successfully to treat uncontrollable postpartum bleeding was reported by Brown et al. in 1979 [10]. Angiographic embolization has the advantages of decreased morbidity, ability to localize the bleeding site and provide a more distal occlusion than surgical ligation, and preservation of future fertility compared to hysterectomy [9].

4. Conclusion

Uterine artery pseudoaneurysm should be considered in the differential diagnosis of delayed postpartum hemorrhage. It occurs most commonly after Cesarean section but is also associated with abortion, repeated curettage, myomectomy, hysterectomy and vaginal delivery. Ultrasound, CT angiography and MRI are modalities applied to diagnosis of this complication. Uterine artery embolization is preferred treatment but ligation of uterine artery by surgery and hysterectomy are alternative treatments.

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