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Research Article

Variable Presentation Of CSF Otorrhoea In Chronic Otitis Media: Our Experience

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ABSTRACT

Introduction-The cases of CSF Otorrhoea in a setting of chronic otitis media (COM) are being reported since a very long time but are rare overall. Now days cases of CSF otorrhoea are not very common and that too in a setting of uncomplicated COM.Cases of CSF otorrhoea presenting within a week and after 3rd post-op week due to iatrogenic trauma are common, but uncommon in a limited choleasteatoma confined to middle ear cavity.[1]

Case Report- Here we present case series of 4 CSF Otorrhoea cases, wherein 2 cases had limited middle ear disease i.e., safe COM, while 2 cases had extensive disease in the form of choleasteatoma. i.e., unsafe COM. The presentation of all these cases was different. The unique feature of this case series is that, even a limited amount of middle ear disease can give rise to grave complication like CSF otorrhoea.

Conclusion- Despite of variable presentation of CSF Otorrhoea which we encountered in our case series, all patients had a post-operative recovery without any morbidity. This shows that meticulous history taking, accurate radiological diagnosis of defect and repair of CSF leak site with optimal surgical intervention is the key to a successful CSF otorrhoea management.

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INTRODUCTION

Incidence of COM cases presenting with complications like CSF Otorrhoea is very low. CSF Otorrhoea in unsafe COM cases was reported frequently before the advent of antibiotics; but after the discovery of effective antibiotics, incidence of CSF Otorrhoea presenting as a Complication of COM reduced drastically. CSFotorrhoea cases following injury to the head due to fall or road traffic accident are seen frequently but not common in limited middle ear disease that too in a limited cholesteatoma. To the best of our knowledge, there are only handful of cases Of CSF otorrhoea in complicated COM reported in the literature till date.[1]In our case series, we have described clinical features and management of 4cases of CSF Otorrhoea who presented to the ENT OPD of our institute which is a tertiary care referral centre. Out of 4 cases, 2 cases had Spontaneous CSF leak in limited COM, while 2 cases had CSF leak secondary to iatrogenic Trauma. Out of the 4 cases, 1 case had CSF Otorrhoea who underwent multiple revision surgeries and presented over a period of 20 years which is not reported in literature till now.

CASE REPORT

Case 1- 68 years old male patient, Kiran came with chief complaints of right earache, decreased Hearing since 15 days & right ear discharge since 2 days. Ear discharge was watery, continuous Spontaneous, non-foul smelling and non-blood stained. There was no history of trauma or similar complaints in the past. He was a known case of hypertension and was on medications.

Figure 1: Clinical image of patient

Patient was admitted in the ENT ward and all routine investigation were sent. Watery discharge
was collected in sterile test tube and sent for beta-2-transferrin which came positive.

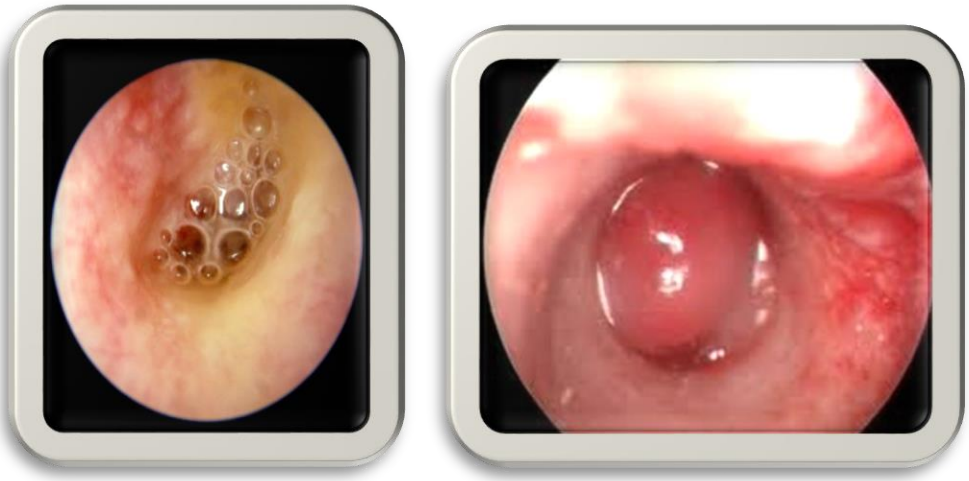


Figure 2: Otoendoscopy image of patient's External auditory canal (EAC)

Otoendoscopy was done that gave impression of meningoencephalocele. Ideally MRI should have been done, but due to infrastructure constraints it was not possible. HRCT temporal bone was done which was suggestive of limited cholesteatoma tissue only in attic /anterior

epitympanum, erosion of tegmen plate with no obvious defect, rest of the mastoid was well Pneumatized with no other abnormality. Canal wall up mastoidectomy was done and CSF leak located at the tegmen plate which was repaired in 2 layers- 1st layer of temporalis fascia reinforced with second layer of tissue glue. Post-operatively CSF protocol was followed by giving IV antibiotics, Head-up position, stool softeners, avoiding straining and coughing.

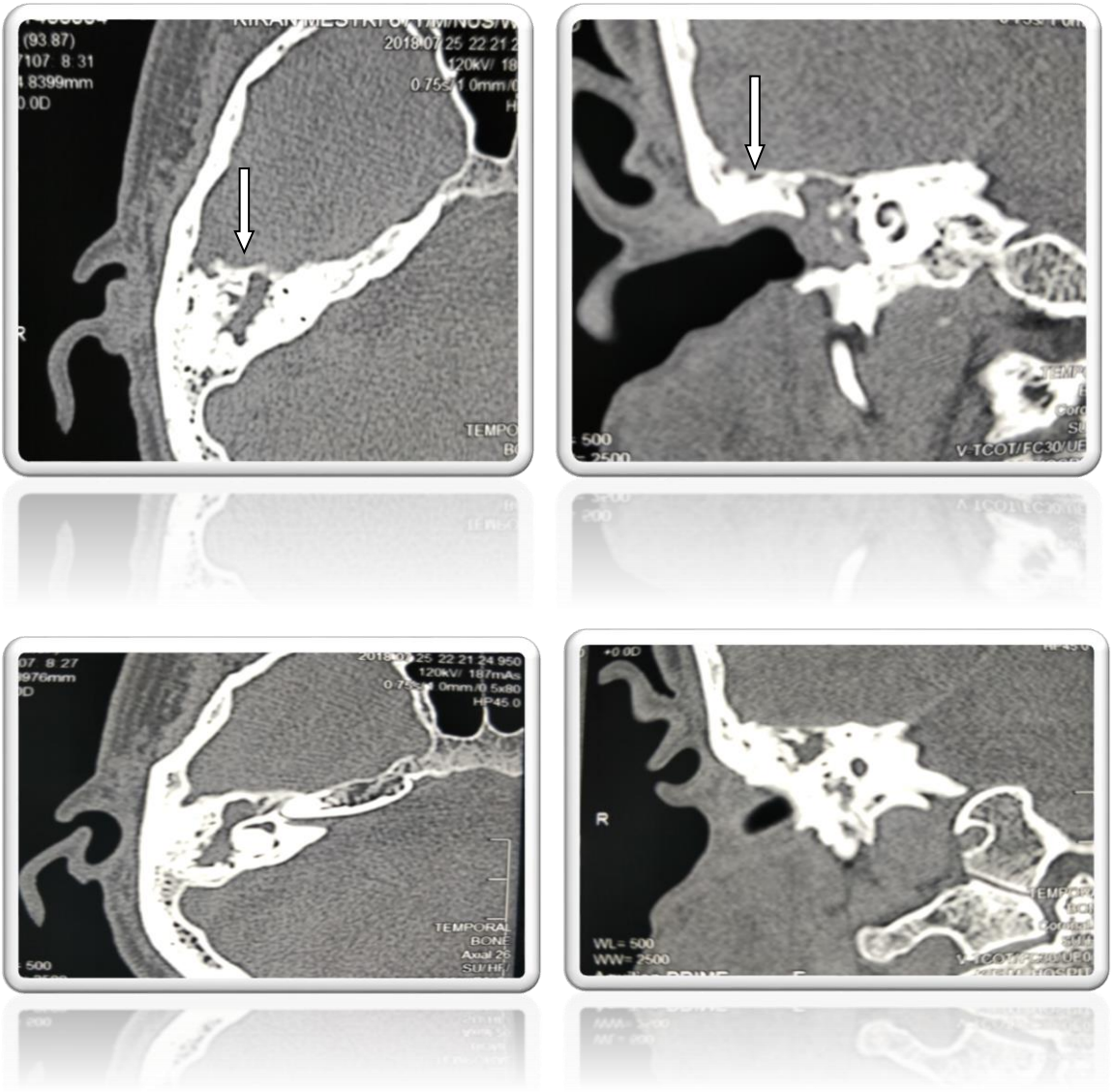


Figure 3: *Serial axial &coronal cuts on HRCT scan with arrows pointing towards soft tissue in middle ear with dehiscent tegmen plate.*

Case 2-A 25-year-oldmale patient was referred to us with history of ear surgery-Mastoidectomy done for cholesteatoma. On 3rd post-operative day patient developedCSF Otorrhoea. On examination of ear, profuse CSF leak was present. HRCT temporal bone with brain cuts was suggestive of dural defect & brain herniation. He was posted for emergency Surgery. The dural defect was identified probably in a case of iatrogenic CSF Otorrhoea and then Closed in three layers using conchal cartilage with perichondrium, temporalis fascia, surgical &Tissue glue. Patient was also started on parenteral antibiotics crossing blood brain barrier and Syrup Kesol, tablet Diamox for 10days. The patient recovered in 3weeks without any further Complications. Regular follow up was done every 3 monthly for aperiod of one year.

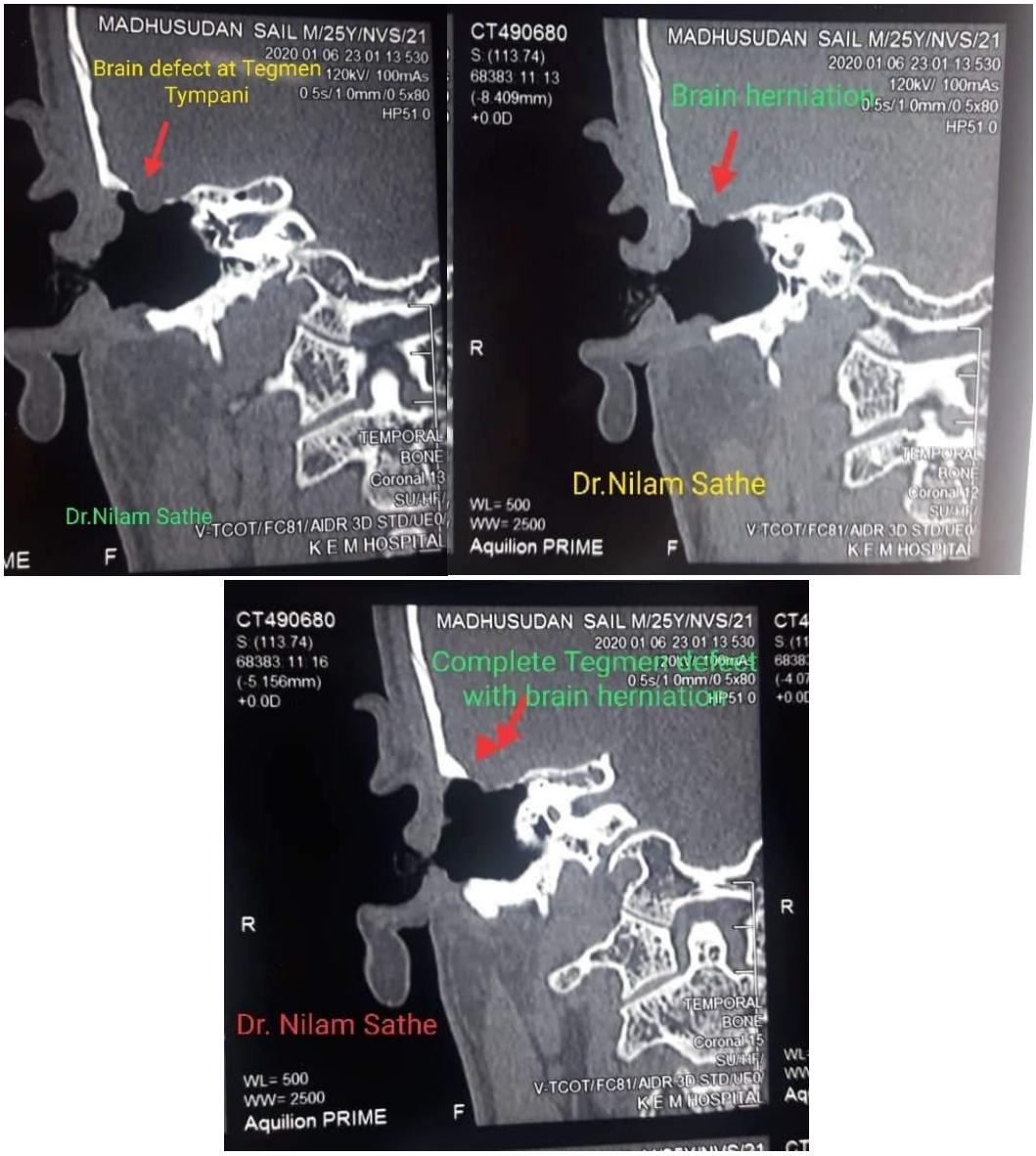


Figure 4: *Serial coronal cuts on HRCT scan showing tegmen defect & brain herniation*

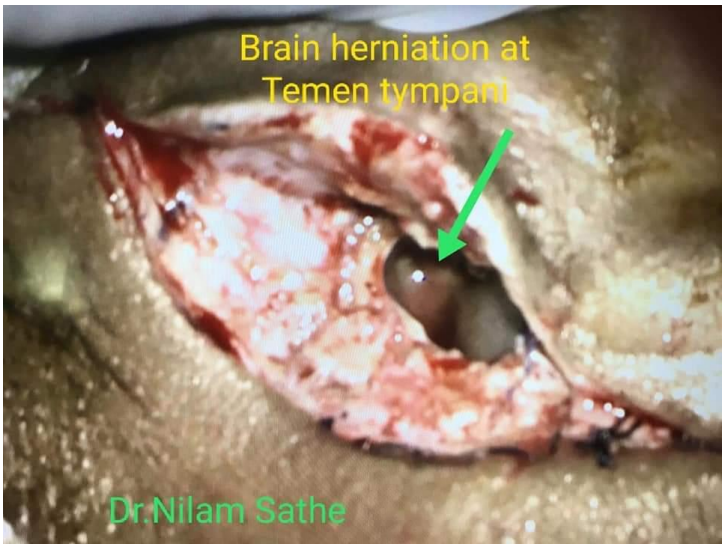


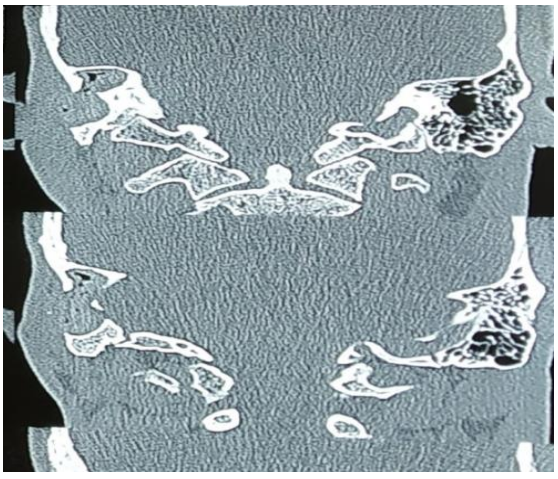
***Figure 5**: Intra-operative microscopic image showing brain herniation due to tegmen defect*

Figure 6: Picture showing post-aural healed wound after CSF leak repair

Case 3- Patient Ajmat was having CSF Otorrhoea. He was operated four times for recurrent Cholesteatoma over a period of 20 years. The anatomy was distorted due to previous surgery. HRCT temporal bone was done which helped us in knowing the anatomy, but exact site of leak was not seen. He was posted for surgery of ear. Intraoperative challenge was to find out the site of CSF leak. The bridge was still intact. Extensive cholesteatoma was present with complete attenuation of sigmoid sinus bony plate. Very gently & meticulously the whole cholesteatoma was removed; but while removing the cholesteatoma from aditus to antrum, CSF started gushing.

We removed the bridge & the CSF leak site was noted at the junction of bridge & tegmen plate. This CSF leak was closed using conchal cartilage with perichondrium, temporalis fascia andsurgicel. The patient recovered without any complications after 4weeks. Regular follow up was done at interval of 1 month and 3 months for a period of one year.

***Figure 7**: Serial coronal cuts on HRCT scan showing attenuation of right sigmoid sinus plate*



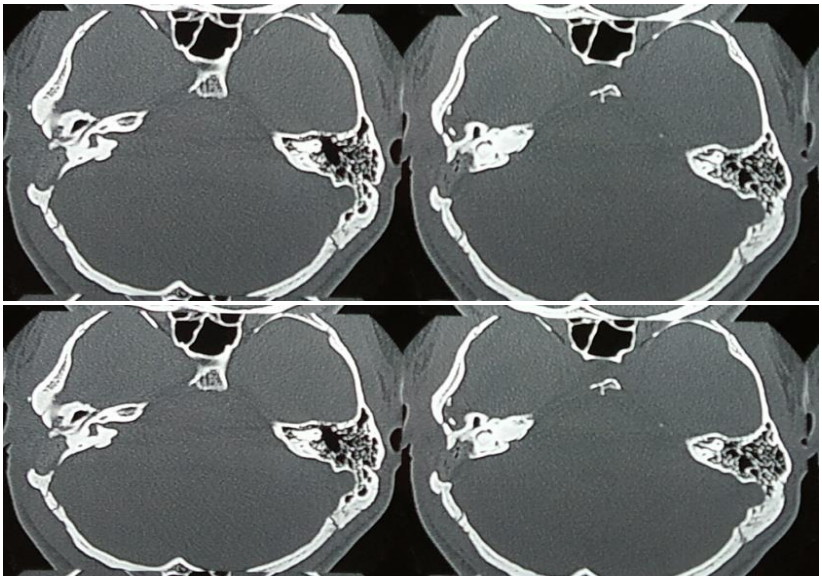


Figure 8: Serial axial cuts on HRCT scan showing attenuation of right sigmoid sinus plate

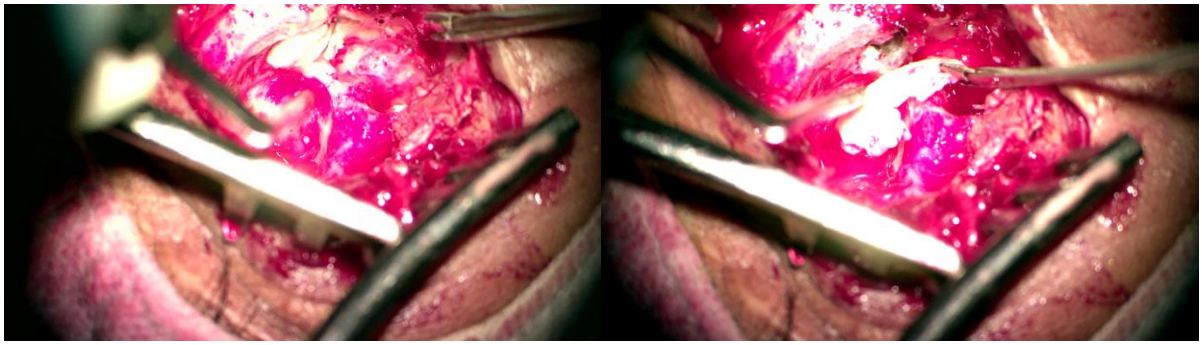
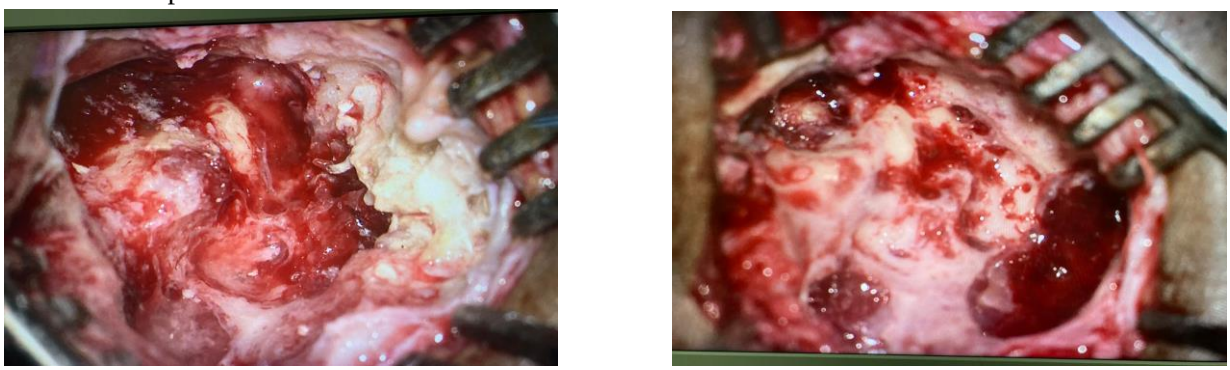


Figure 9: Intra-operative images- Left side showing site of CSF leak and right-side showing repair of the leak with piece of conchal cartilage

Case 4 - A 54-year-old female presented with CSF leak on 3rd post-operative week after Mastoidectomy surgery. Patient had recurrent extensive cholesteatoma with extradural abscess accompanied by huge defect of tegmen plate &sigmoid sinus. Findings were confirmed on HRCT temporal bone with CT scan of brain. Revision canal wall down mastoidectomy was done. CSF leak was repaired by sealing tegmen & sigmoid plate defect with conchal cartilage, temporalis fascia, surgical & tissue glue. Patient recovered in a month without any complications.

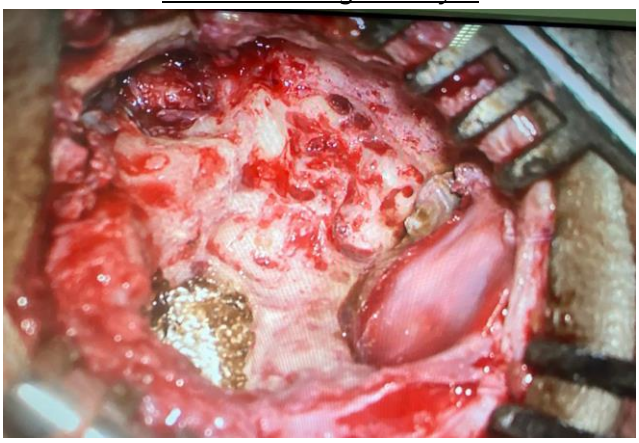


M1 **M2**

***Figure 10**: M1- showing defect at the tegmen plate M2- showing defect at the sigmoid plate*

M3 **M4**

Figure 11*: M3- showing covergae of leak with surgicel-1st layer, M4: showing repair with conchal cartilage-2nd layer*



M5

Figure 12*: M5- showing repair with tissue glue(3rd layer) put on top of the assembly ofsurgicel, conchal cartilage and temporalis fascia*

DISCUSSION

Chronic otitis media is still widespread in India and other developing countries despite availability of newer antibiotics. Lack of hygiene, low socio-economic status, crowding, lack of education and unawareness about the need to seek help of a doctor are some of the factors responsible for patients presenting late to a hospital with complications related to COM.

Extracranial complications like mastoiditis are very common and can be managed with IV antibiotics and simple surgery like mastoidectomy without any post-operative morbidity. Intracranial complications are rare these days but are still prevalent in many parts of India.Out of the various intracranial complications, CSF Otorrhoea is such a complication which is very rare but requires urgent medical attention and surgical intervention. To the best of our Knowledge there are only 20 cases of CSF Otorrhoea in complicated COM described in the Literature till date.[1]Our study described 2 cases that had spontaneous leak in a limited COM without any other pre-existing complication and without history of trauma, which is rare in the literature. Albertiand Dawes reported a series of cases of CSF Otorrhoea associated with COM.[1] Hicks et al . reported cases of unsafe COM, of which some cases intracranialabscesses.[2] Adkins and Osguthorpe describedthemini-craniotomy procedure forthe management of CSF otorrhoea from tegmen plate defects caused by COM.In our case series, case no. 2& 4 had had tegmen plate defect while case no.1 had only dehiscence of tegmen plate in a case of limited

COM and without any history of trauma/ previous surgery.

There is only handful of cases of COM with or without cholesteatoma described in literature, in whom there was a dural defect and resultant herniation of brain.[4,5] In our study, 3 cases of COM with cholesteatoma had dural defect with brain herniation which were subsequently repaired in layers using autologous tissue and synthetic material. Cases of recurrent CSF Otorrhoea leading to meningitis have been described in the literature.[6] We had case no. 3 in our series which presented with CSF Otorrhoea over a span of 20 years and underwent 4 revision surgeries.

Recurrent infection and inflammation of the meninges most probably could have been the cause for CSF Otorrhoea in this case as brain tissue was exposed due to attenuation of sigmoid sinus plate. Potential sites of CSF leak such as oval window, round window, stapes footplate, Eustachian tube and promontory have been described in the literature,[7] but in our case series there was no such site of CSF leak found on CT scan and intra-operatively.

According to the literature, the aetiopathogenesis of CSF Otorrhoea in COM is due to 2 main mechanisms mainly- 1. Pre-existing infection like choleasteatoma which has the property to erode the bone and thus expose the middle ear to intra-cranial contents. 2. Iatrogenic trauma to the already thinned and dehiscent structures like tegmen and sigmoid sinus due to previous operations and pre-existing COM[8,9]

Role of congenital dehiscence of the tegmen plate is attributed to the pathogenesis of CSF otorrhoea which is very rare.[10] We had case no. 1 in the series who had dehiscence of tegmen plate on CT scan and history of CSF Otorrhoea in a pre-existing COM without any cholesteatoma. Case no. 2 in the series had tegmen defect and subsequent brain herniation which was probably secondary to iatrogenic trauma sustained in previous surgery. Case no. 3 in the serieshad sigmoid sinus attenuation with breach at the junction of bridge and tegmen plate. The pathology is this case was due to the combination of COM and most probably iatrogenic trauma due to 4 revision surgeries over a period of 20 years. Case no. 4 in the series had extra-dural abscess on the presentation with large defect of tegmen plate and sigmoid sinus plate.CSF leak in this could be explained by the fact that erosion of bony plate over the tegmen and sigmoid sinus leads to breach of dura. Dural breach could have been subsequently responsible for the passage of extra-dural abscess tracking into the middle ear cavity.

The most common surgery for CSF Otorrhoea cases is mastoidectomy followed by drainage of abscess if any and subsequent closure of the leak; but each and every case is different and final management depends on the specific individual surgical findings intra-operatively.[2] In case no. 1 canal wall up mastoidectomy was done to remove the limited cholesteatoma and CSF leak at the tegmen repaired with a layer of temporalis fascia and tissue glue. In case no 2, 3 & 4 in our series, canal wall down mastoidectomy was done to remove the disease and site of CSF leak was located which was subsequently repaired using conchal cartilage with perichondrium (autograft), temporalis fascia (autograft), surgicel (oxidised cellulose as a synthetic haemostatic agent) and tissue glue (synthetic sealing agent). Reports of synthetic material used for brain herniation and CSF leak repair are mentioned in the literature.[2] Craniotomy approach which is used for middle fossa defects with CSF leak & brain herniation was not used in any of our cases. Radical mastoidectomy which is one of the non-neoplastic indications for CSF Otorrhoea in COM were not required in this case series.

CONCLUSION

- Out of the various intracranial complications, CSF otorrhoea is such a complication which is very rare but requires urgent medical attention and surgical intervention.
- There are very few cases of CSF otorrhoea in complicated COM described in the Literature till date that had spontaneous

leak in a limited COM without any other pre-existing complication and without history of trauma, which is rare in the literature.

- Despite of variable presentation of CSF otorrhoea which we encountered in our case series, all Patients had a post-operative recovery without any morbidity.
- This shows that meticulous history taking, accurate radiological diagnosis of defect and repair of CSF leak site with optimal surgical intervention is the key to a successful CSF otorrhoea management.
- Extensive & morbid Procedures like radical mastoidectomy, craniotomy is not always required even if the case is of a complicated COM with intra-cranial complications.

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