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Surgical Management of Cerebrospinal Fluid (CSF) Otorrhea Presenting as CSF Rhinorrhea: Different Approaches

Mehta Madhuri, Morwani KP and Arsiwala Zainab*

Department of Ent, N.C. Jindal Institute of Medical Sciences, India

Abstract

CSF otorrhea though a rare clinical entity, is a life threatening situation that requires rapid intervention. Though occurring through the otologic structure, actual leakage from the ear may not always be present (unless the ear drum or canal is in some way violated), but the fluid may flow down the Eustachian tube and manifests as a clear fluid through the nose. The presence of an abnormal communication of the sterile subarachnoid space with the flora of the sinonasal tract, places the patient at a great risk of meningitis. Diagnosis of such cases, need a high index of suspicion, complete clinical examination of nose and ear and relevant investigations to avoid unnecessary nasal exploration and prevent life threatening meningitis. This case report presents the diagnosis and management of two cases of CSF otorrhea presenting as CSF rhinorrhea, one having a spontaneous leak, and the other following trauma. Mention is made of how one could subject a patient to inadequate surgery, if one is not vigilant and adequate investigations are not carried out. Different methods of repair depending on the site and size of defects are outlined in the two different cases. Consent was obtained from each case in the format mentioned towards the end.

Introduction

CSF leak can occur when there is disruption in the arachnoid and dura mater coupled with an osseous defect and a CSF pressure gradient that is continuously or intermittently greater than the tensile strength of the disrupted tissue [1]. The aetiological factors for CSF leaks in mastoid include otologic or skull base surgery, trauma, tumors and spontaneous –idiopathic and congenital dehiscence [2]. Complication of Skull base surgery is the most common cause of CSF otorrhea, followed by temporal bone fractures (21%). Spontaneous leaks are extremely rare with only 500 cases being reported in literature worldwide. Traumatic leaks are commonly seen following road traffic accident associated with other multiple injuries and hence diagnosed at a later stage once the patient recovers neurologically. Most common site for dural defect is the tegmen plate. This defect in the dura mater around the temporal bone defect can transmit the CSF into the mastoid air cells which may percolate down the Eustachian tube into the nasopharynx when the ear canal and tympanic membrane are intact. Patients may complaint of unilateral nasal fluid drainage that is worse after waking up or when bending over.

Diagnosis is based on a high index of suspicion, complete clinical evaluation, and radiological imaging and/or laboratory studies. High resolution CT scan is sufficient to locate the site or sites of CSF leaks. MRI is useful in demonstrating meningocele or meningoencephalocele when associated with CSF leak, as well as for examining patients with spontaneous intracranial hypotension syndrome. MRI is also required in post-surgical cases to differentiate brain tissue from cholesteatoma or inflammatory tissue in mastoid cavity. Management consists of wide surgical exposure, amputation of necrotic, herniated part of brain or meninges with fine bipolar cautery and finally repair of dura mater and the defect in temporal bone in layers. Though various reconstruction techniques with different approaches have been described in the literature, an individualized approach should be taken for each case depending on different factors. The approaches can be classified into otological, neurological and combined. Each approach has specific advantages and final choice is made taking into consideration factors like the position and size of the defect on preoperative imaging, the aetiology of meningoencephalocele, and the preoperative audiometry.

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***Correspondence:**

Mehta Madhuri, Department of Ent, N.C. Jindal Institute of Medical Sciences, Hisar Haryana, India, Pin Code: 125005;

E-mail: madhurimehta@gmail.com

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Figure 1A: HRCT Para nasal sinuses with post-surgical changes but no anterior skull base defect.

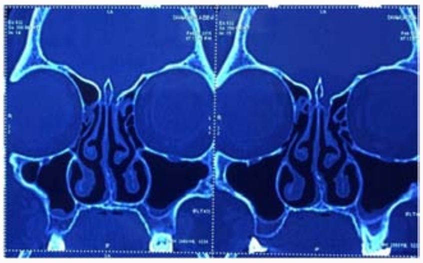

Figure 1B: HRCT Para nasal sinuses prior to endoscopic sinus surgery, coronal cuts showing normal sinuses with no anterior skull base defect.

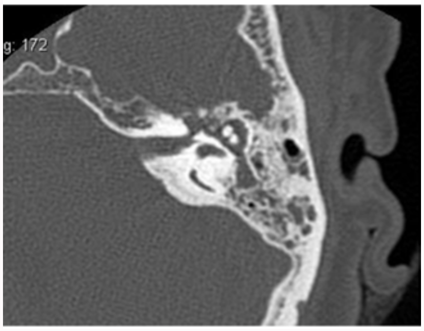

Figure 1C: HRCT temporal bone, axial cut showing defect in the tegmen tympani in the region of attic with opacification of antrum and middle ear.

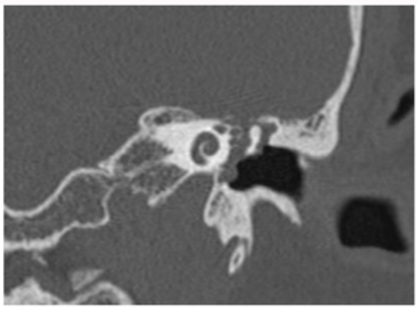

Figure 1D: HRCT temporal bone, coronal cut showing defect in tegmen antri.

Case Presentation

Case 1

A 62 years old obese female patient presented with the chief complaints of spontaneous, intermittent, unilateral, clear watery nasal discharge from left nostril since 4 months which was worse on waking up and bending forwards. She also had two episodes of fever associated with vomiting and giddiness which subsided with a course

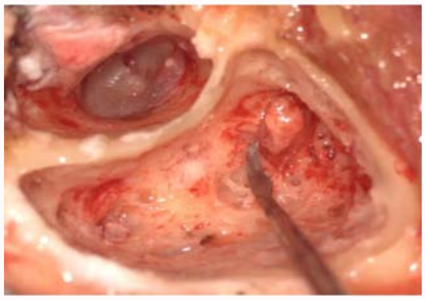

Figure 2A: Showing meningoencephlocoele in the region of attic. Tympanic membrane found intact (black arrow).

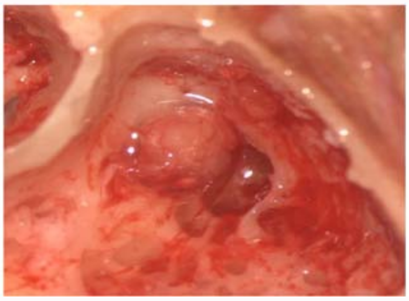

Figure 2B: Showing CSF (black arrow) and meningoencephlocoele(blue arrow).

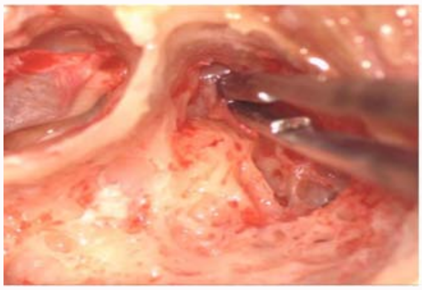

Figure 3A: Bipolar cauterization of meningoencephlocoele to reduce the size and reach margins of tegmen bone defect in all dimensions.

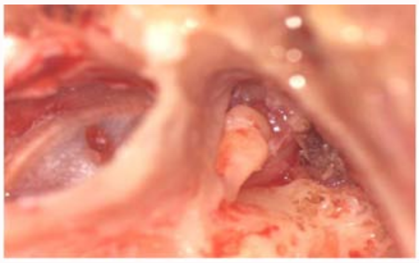

Figure 3B: The anterior extent of meningoencephlocoele which is reaching anterior and medial to head of malleus and body of incus(black arrow).

of oral antibiotics. She complained of decreased hearing in the left ear, of the same duration as that of the nasal discharge.

Fluid sample was collected and chemical analysis for glucose and beta2 transferrin was positive, consistent with CSF. Computed Tomography (CT) scan of the Para nasal sinus (PNS) showed post-surgical changes but no defect in anterior skull base (Figure 1A) which could be a potential site of leak, and audiogram showed a moderate conductive haring loss in the left ear. Patient had consulted a surgeon

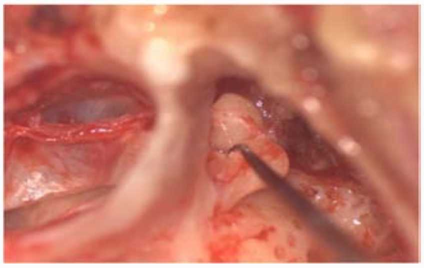

Figure 4A: Incus and head of malleus being removed , to reach anteriormost extent of the bony defect.

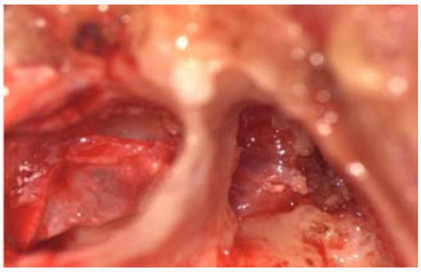

Figure 4B: Complete exposure of meningoencephlocoele (black arrows).

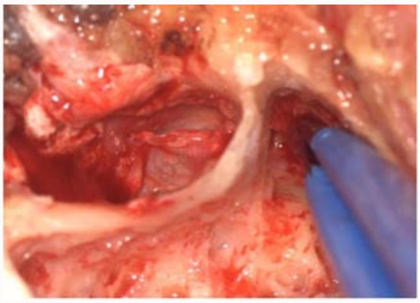

Figure 5A: Bipolar cautery of meningoencephlocoele.

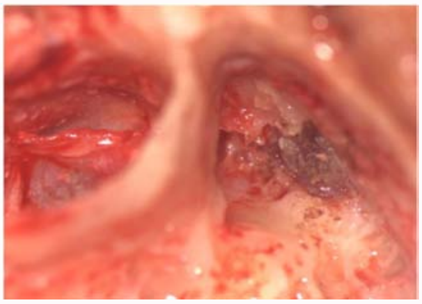

Figure 5B: After reduction of Meningoencephlocoele extent of bone defect exposed in all dimensions.

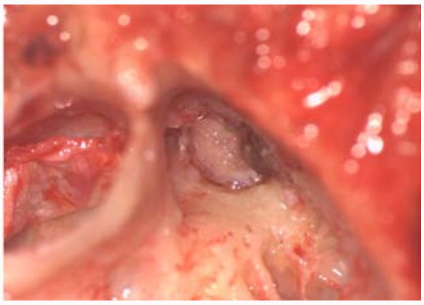

Figure 6A: Defect sealed with muscle layer.

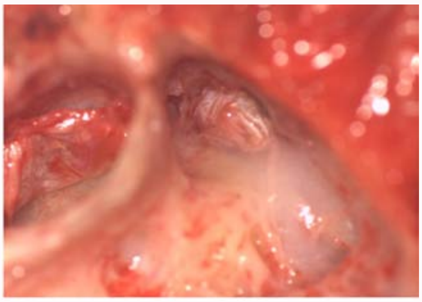

Figure 6B: Followed by temporalis fascia graft and tissue glue.

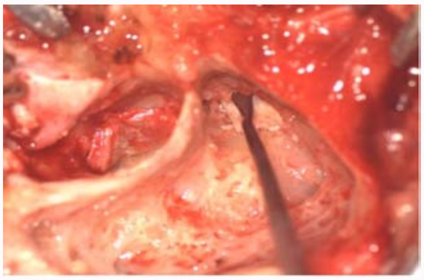

Figure 6C: Finally by cartilage and then covered with fascia and glue once again.

in the past, who had carried out left sided functional endoscopic sinus surgery in order to look for the site of leak, in spite of normal high resolution CT scan of Para nasal sinuses (Figure 1B), but as no defect was found, and patient had persistent CSF leak even post nasal exploration, the patient consulted us. In view of persistent CSF leak through the nose, a normal CT PNS (prior to nasal exploration), and hearing loss, we were directed for radiological assessment of the temporal region. CT scan of the temporal bone revealed a defect in the tegmen tympani with opacification of the left middle ear by fluid and soft tissue (?Meningoencephlocoele) (Figure1C and D). Endoscopic examination of left nostril revealed evidence of post endoscopic sinus surgery changes with CSF leak from nasopharynx area. Microscopic examination of left ear revealed intact tympanic membrane. Decision was taken to explore the left middle ear and mastoid via a transmastoid approach.

Surgical steps (left ear): Cortical mastoidectomy was performed. CSF leak was found from the defect which was in the medial portion of tegmen tympani and antri in the region just superior and anterior to the head of malleus and body of incus. Through the defect herniated necrotic brain along with meninges could be visualized. Hypertrophied mucosa and granulation tissue was seen around the ossicles and tegmen, along with hyper cellularity in that region (Figure 2A and B). Lateral wall of antrum and posterior attic was drilled away to expose the meningoencephlocoele. Gentle bipolar cauterization of the herniated brain was done (Figure 3A) to reduce its size and reach the anterior extent of meningoencephlocoele which was reaching anterior and medial to head of malleus and body of incus (Figure 3B). The incus and head of malleus removed gently and meningoencephlocoele was exposed in its anterior most extent (Figure 4A and B). Reduction of rest of the meningoencephlocoele towards the middle cranial fossa was performed with fine bipolar

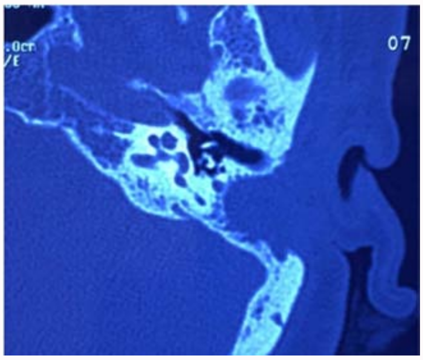

Figure 7A: Post-operative HRCT temporal bone **A:** axial cut showing repair of defect in tegmen and ossiculoplasty with head of malleus over head of stapes (yellow arrow).

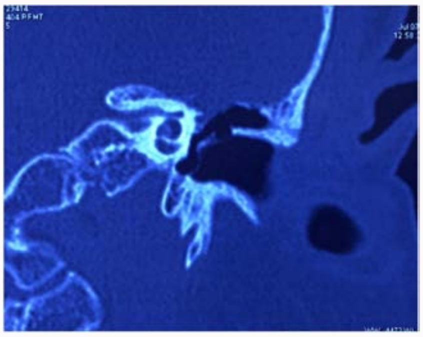

Figure 7B: Showing repair of tegmen with cartilage (black arrow) which cannot be differentiated from surrounding soft tissue.

Figure 7C: Post operative otoendoscopy examination showing intact tympanic membrane.

cautery (Figure 5A) and the defect in tegmen exposed completely (Figure 5B). As the size of the defect was <2 cm x 2 cm and located in medial part of tegmen plate, cartilage graft was chosen for repairing the bony defect which was in two dimensions. It's worth mentioning here that bone graft being stiff and non-malleable could not be used in this case.

The complete defect was repaired in 4 layers. As it was a meningoencephlocoele, an extra layer in the form of free muscle graft was used. A piece of muscle was harvested from the post auricular region which was about one and a half times the size of the defect and was carefully tucked into tegmen defect. The muscle layer was then covered with the temporalis fascia graft which was also gently tucked underneath the tegmen (used to replace the defect in the dura) and sealed with tissue glue. These layers were then secured in place by a large piece of tragal cartilage (bigger than the size of the defect), carefully tucked in the defect of the tegmen in all directions. This was then finally covered with temporalis fascia again and then covered with tissue glue (Figure 6A, B and C).

The patient was followed postoperatively on day 7 and 21 after discharge, then 3 monthly for the first year, 6 monthly in the second year and has been asked to stay on long term follow up at least once yearly. There is no recurrence of CSF leak either through the nose or ear, and hearing of the patient has returned to near normal. The post-operative HRCT temporal bone showed repair tissue in place (Figure 7A and B) and otoendoscopy showed intact tympanic membrane (Figure 7C). After a 15-months follow-up the patient remains symptom free and no recurrence is noted.

Case 2

A 27 year old boy came with complaints of clear, unilateral watery nasal discharge on left side, 2 months following head trauma. Patient had no neurological deficit following trauma. Patient also had no complaints of giddiness though he was having mild hearing loss on the left side. Computed tomography scan of the paranasal sinuses was done which was normal. In view of a normal CT PNS and clear watery nasal discharge with an intact tympanic membrane left side, decision was taken to do a CT scan of the temporal bone. CT revealed a defect in the region of tegmen antri left side (Figure 8A and B), and hence a decision of doing an exploratory mastoidectomy was taken.

Surgical steps (left side): The tympanic membrane was found intact and cortical mastoidectomy showed active CSF leak from defect in tegmen antri and tegmen mastoideum near its lateral portion superior and posterior to the ossicles (Figure 9A and B). So ossicles were not disturbed. Tympanomeatal flap had to be lifted to

Figure 8A: HRCT temporal bone showing tegmen defect in coronal cuts.

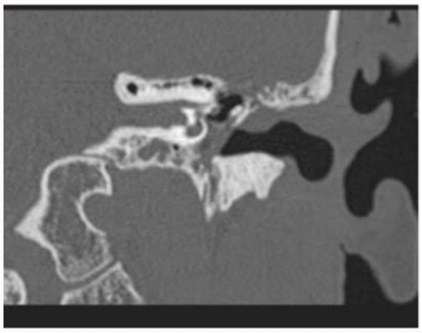

Figure 8B: HRCT temporal bone showing tegmen defect (yellow arrow) in axial cuts with opacification middle ear.

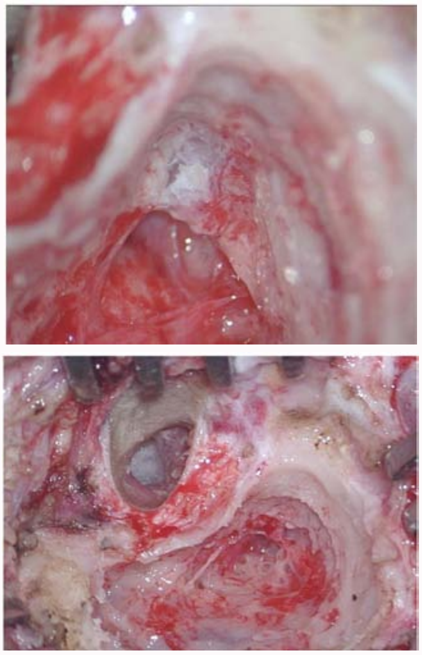

Figure 9A and B: Defect in the tegmen antri and tegmen mastoideum with active CSF leak (black arrow). Intact tympanic membrane can be visualised (white arrow)

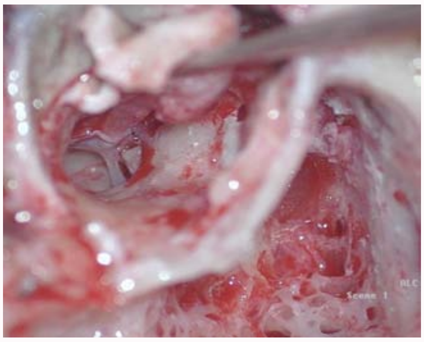

Figure 10A: Tympanomeatal flap (TMF) is lifted. Fracture line running from tegmen plate to posterior bony canal wall can be visualised (white arrows).

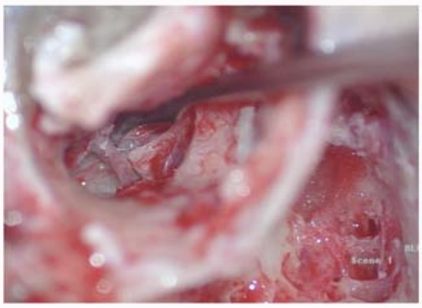

Figure 10B: The adhesions around long process of incus and incudostapedial joint (black arrow) being cleared.

look for cause of mild conductive hearing loss (Figure 10A). Fracture line could be seen extending from tegmen bone to posterior bony canal wall (Figure 10A). There were adhesion bands around intact ossicular chain which were released (Figure 10B).

The meningoceole was gently bipolarised and defect in tegmen antri was localised in its lateral part (Figure 11A). The linear fracture line was extending from lateral to medial part of tegmen antri. The dura was separated from around the margins of defect so as to assess the exact size and extent of defect (Figure 11B). The assessment of the size of the defect was done, which was around 2 cms x 2.5 cms. As the defect was laterally placed and size was more than 1cm [2], the transmastoid approach was combined with mini middle cranial fossa approach and an autologous bone graft from the temporal craniotomy was used in place of cartilage graft to cover the tegmen defect.

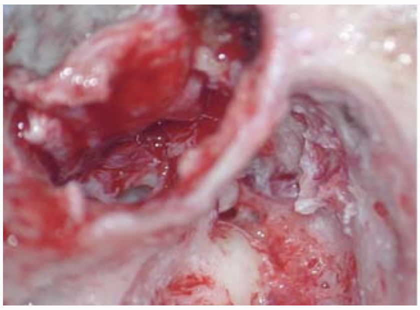

Figure 11A: Meningocoele was gently bipolarised. Linear fracture line with defect in tegmen antri in lateral part is visible (black arrows).

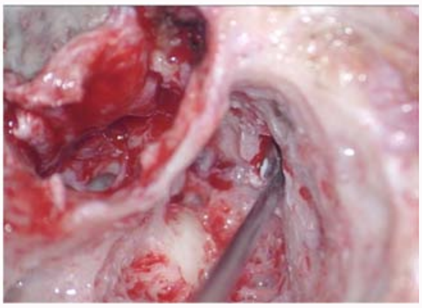

Figure 11B: Dura was lifted from the margins of defect to assess the exact size and extent of defect.

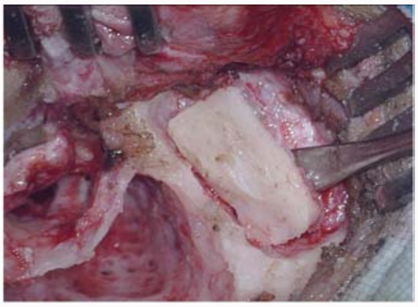

Figure 12: Bone graft harvested from squamous temporal bone.

The defect was repaired in three layers. As it was only meningocoele with no brain tissue, the muscle layer was omitted. The reinforcement of dura was done with temporalis fascia and bony defect was repaired with squamous temporal bone graft. For harvesting bone graft, the squamous temporal bone was exposed by elevating the superficial temporal muscle. A craniotomy of size 3 cms x 3.5 cms was drilled in the squamous temporal bone. Inferiorly the craniotomy reached few mms above the tegmen margin so as to minimise the degree of temporal lobe retraction. Anterior extent was up to base of zygoma and posterior extent was up to sinodural angle so as to cover and reach beyond the defect in tegmen plate in all dimensions. The craniotomy

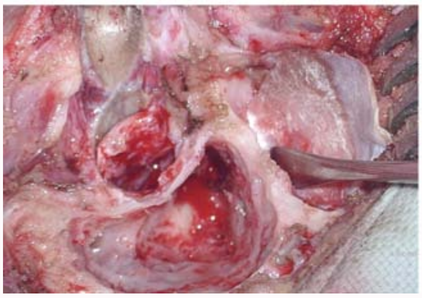

Figure 13A: First layer of temporalis fascia.

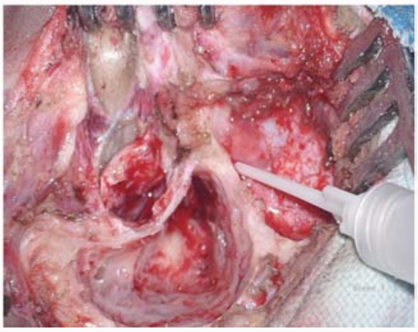

Figure 13B: Use of fibrin glue in-between the layers.

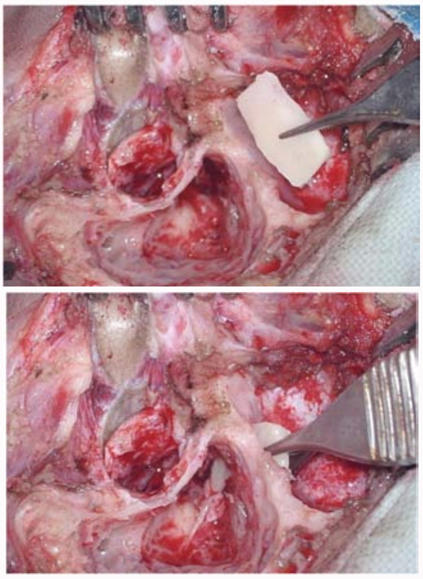

Figure 14A and B: Second layer of squamous temporal bone being placed via the middle cranial fossa approach.

was performed with a medium sized cutting burr first and small sized diamond burr in final stages, when we reached close to dura, so as to avoid any injury to dura. The bone graft was slowly separated from the underlying dura after completing drilling the margins.

The middle cranial fossa dura was then carefully separated from the tegmen bone from middle cranial fossa side, around and beyond the complete circumference of defect so as to create space for placing the fascia and bone graft. The first layer which was temporalis fascia, harvested at the start of the operation, was placed from middle cranial fossa side to repair the dura (Figure 13A). The fascia layer was reaching beyond the defect in all dimensions. Fibrin glue was used in

Figure14C: Graft reached beyond the defect in all directions.

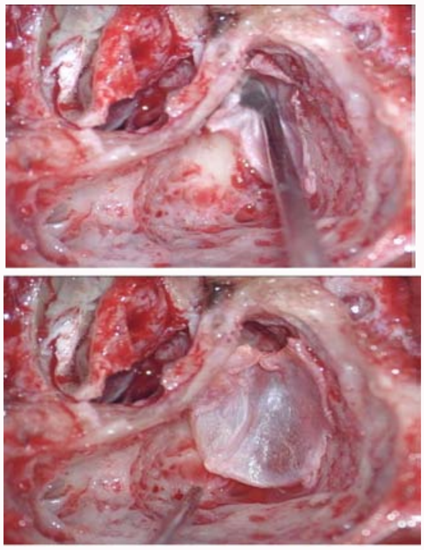

Figure 15A and B: The third layer of repair with temporalis fascia tucked into and all around the defect from mastoid side.

Figure 15C: Final layer of fibrin glue to seal all layers.

between the layers (Figure 13B).

After spreading the fascia graft, for second layer, the already harvested autologous bone graft is placed from middle cranial fossa side between the temporalis fascia graft and tegmen bone (Figure 14A and B). It covered the defect in bone and reached beyond the defect in all dimensions (Figure 14C). The third layer was with temporalis fascia which was again tucked in around the defect from mastoid side (Figure 15A and B). Fibrin glue was again used to seal the defect with all layers (Figure15C). Post operatively patient was examined on 7th and 21st post-operative day. After that regular follow up was carried out every three months for one year. The HRCT scan temporal bone was carried out after one year. Post-operative CT scan showed bone

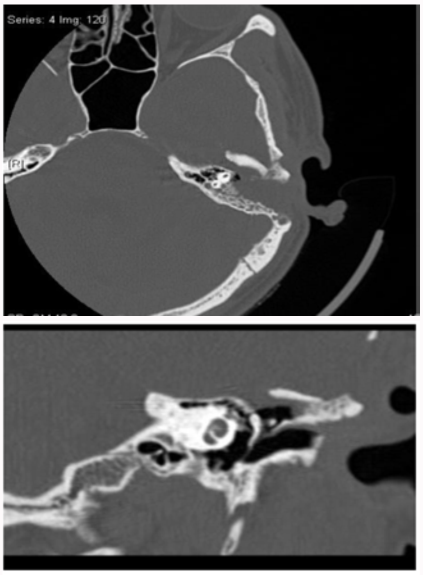

Figure 16A and B: Post-operative scan showing graft in situ in A: axial and B: coronal cuts (yellow arrow)

Figure 16C: Otoendoscopy showing intact tympanic membrane.

graft in place (Figure 16A and B) and post-operative otoendoscopy showed intact tympanic membrane (Figure 16C). One year post surgery, the patient is completely symptom free with no recurrence of CSF leak through nose or ear and his hearing has returned to near normal.

Discussion

CSF leakage otorrhea can be either acquired or spontaneous. Acquired causes which are more frequent include skull base surgery, trauma and tumors, whereas spontaneous which are very rare can be either due to congenital defects in the tegmen or dehiscence of a congenitally thin tegmen either due to raised intracranial pressure or arachnoid granulations.

Children are subject to spontaneous CSF otorrhea, and it can be detected with congenital disorders, such as Mondini anomaly, patent cochlear aqueduct, patent Hyrtl fissure, patent petromastoid canal, and patulous facial canal [3]. Spontaneous CSF otorrhea can be also detected in adults, and it occurs when the bone matrix, which is between the middle ear and the intracranial space, is abnormal. As temporal bone develops there may be congenital deficiency within the skull bones which over the time keeps persisting. With every heart beat there is thrust of brain on these weak areas. The pressure keeps on building over the years leading to remodelling of weak bones finally presenting with herniation of brain and meninges with or without CSF leak, in to mastoid air cells. These leaks can be single or multiple depending on sites of weak areas. Other causes for spontaneous leak can be, a defect in the annular ring of the stapes footplate resulting in drainage of CSF into the middle ear. Similar presentation may be observed in a patient with widely patent cochlear or vestibular aqueduct, abnormal patency of petro mastoid canal or patent Hyrtl fissure. CSF may propagate along the fallopian canal and can leak adjacent to the facial nerve [4].

In such situations, as the tympanic membrane is intact(unless patient is suffering from otitis media with perforation of membrane) patient usually presents with CSF rhinorrhea .When we get a normal CT Scan paranasal sinuses, in case of CSF rhinorrhea, a high resolution CT Temporal bone is required to look for defect in tegmen bone with or without meningoencephalocele.

In post-traumatic CSF otorrhea presenting as CSF rhinorrhea, we may find additional fracture lines on high resolution CT scan temporal bone. Localization of an otogenic CSF leak is accomplished using high resolution CT scan with axial and coronal sections, and unless an otologic source is certain, the scan should cover all 3 cranial fossae [4]. MRI is usually required in post-surgical cases to differentiate brain tissue from cholesteatoma or inflammatory tissue in mastoid cavity.

An individualized approach should be taken for repair of temporal bone cerebrospinal fluid leaks. Various reconstruction techniques with different approaches have been described in the literature. The approaches can be classified into five varieties naming: transmastoid, transmastoid with mini middle cranial fossa approach, transcranial intradural, combined transcranial/transmastoid and finally, obliteration of mastoid cavity and middle ear with cul-de-sac closure of external auditory canal.

Each approach has specific advantages and final choice is made taking into consideration factors like the position and size of the defect on preoperative imaging, the etiology of meningoencephalocele, and the preoperative audiometry. The transmastoid approach provides information about the precise size and location of the dural defect and is alone sufficient in case leak is small (<1 cm^2) and closer to medial part of tegmen plate. Here tragal cartilage graft is used to repair bone defect and temporalis fascia is used to reinforce dura mater.

It's combined with mini middle cranial fossa approach [5] to repair the defects which are bigger than 1 cm^2 but less than 3 cm^2. In this approach the squamous temporal bone graft is used to repair the bone defect and temporalis fascia to repair the dural defect. In mini middle cranial fossa approach, as the name suggests, a craniotomy is performed to harvest squamous temporal bone graft, though its dimensions are smaller and more posteriorly based than those of the conventional middle cranial fossa approach. The principle is that the size of craniotomy can vary according to the site and size of defect.

If the size of defect is even bigger and there is associated profound hearing loss (as these cases are usually post traumatic with multiple injuries), the mastoid may need to be obliterated with fat. Obliteration of the middle ear and Eustachian tube may also be required, especially if the leakage is not limited to the mastoid [6]. If diffuse leak is observed from multiple mastoid air cell tracts the approach remains complete obliteration of mastoid cavity and middle ear with cul de sac closure of external auditory canal.

A primary transcranial approach is needed for defects that are very big sized, multiple, located in the petrous apex, and in revision cases. In Trans cranial intra dural approach, two extra layers of artificial dural graft are placed. First layer of artificial dural graft is with Duragen (size 5.0 cm × 5.0 cm) placed extradurally between dura and cartilage graft and second layer of artificial dural graft (Duragen size 2.5 cm × 2.5 cm) is used to cover the dural gap intra durally between inner surface of dura and brain. Intradural graft placement is generally preferred in such cases, as it guarantees repair of meninges after cauterising the protruding part in mastoid cavity. And possibly in revision cases, it ensures, that the infected parts of cortical contents are not pushed intra cranially. Such a graft placement also facilitates better resistant to intracranial pressure [7].

Conclusion

CSF rhinorrhoea may not always be due to anterior cranial fossa defect. High index of suspicion with thorough history and relevant investigations will lead to correct diagnosis and management of CSF leaks that present as nasal leaks but having an otologic origin.

References

1. Yerkes SA, Thompson DH, Fisher WS 3rd. Spontaneous cerebrospinal fluid rhinorrhea. Ear Nose Throat J. 1992; 71: 318-320.
2. Kutz JW Jr, Husain IA, Isaacson B, Roland PS. Management of spontaneous cerebrospinal fluid otorrhea. Laryngoscope. 2008; 118: 2195-2199.
3. Cueva RA, Danner CJ. Prevention and management of cerebrospinal fluid leaks. In: Gulya AJ, editor. Glasscock-Shambaug Surgery of the Ear, 6th edition. Shelton, Connecticut: People's Medical Publishing House. USA. 2010; 765-770.
4. Gacek RR, Leipzig B. Congenital cerebrospinal otorrhea. Ann Otol Rhinol Laryngol. 1979; 88: 358-365.
5. Lindstrom DR, Toohill RJ, Loehrl TA, Smith TL. Management of cerebrospinal fluid rhinorrhea: the Medical College of Wisconsin experience. Laryngoscope. 2004; 114: 969-974.
6. Hafsa C, Kriaa S, Jerbi Ommezine S, Ben Achour N, Salem R, Golli M, et al. A cause of recurrent meningitis in a child: cochlear dysplasia. J Radiol. 2008; 89: 60-63.
7. Blioskas S, Magras I, Polyzoidis S, Kouskouras K, Psillas G, Dova S, et al. Repair of Temporal Bone Encephalocele following Canal Wall Down Mastoidectomy. Case Reports in Otolaryngology. 2014.