**Case report** 

# **Double Trouble of Double Fistulae**

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### ABSTRACT

The complications of attico-antral type of Chronic Suppurative Otitis Media (CSOM) are severe due to underlying bone erosion. We describe a case of a 40-year-old lady with attico-antral CSOM and mastoiditis with a postauricular fistula, who underwent modified radical mastoidectomy with excision of the postauricular cutaneous mastoid fistula. Interestingly, another fistula over the dome of lateral semicircular canal was noted intraoperatively. Few reports of occurrence of postauricular mastoid fistula with a labyrinthine fistula have been documented.

Keywords: Cholesteatoma; cutaneous fistula; labyrinth diseases; mastoiditis; suppurative otitis media.

# INTRODUCTION

hronic Suppurative Otitis Media (CSOM) is a chronic infection of the middle ear cleft that pharyngotympanic includes tube, mesotympanum, epitympanum, aditus and mastoid antrum (1). It has been categorized into safe/ tubotympanic disease (TTD) and unsafe/ attico-antral disease (AAD). AAD is associated with extracranial and intracranial complications such as mastoiditis, mastoid abscess, labyrinthitis, facial nerve paralysis, meningitis, brain abscess, and lateral sinus thrombophlebitis. We report a case of AAD with a postaural fistula complicated by a labyrinthine fistula. Double fistulae is an infrequent occurrence, with only few reports in literature.

#### CASE REPORT

A 40-year-old lady presented with chief complaints of right ear discharge for 20 years, associated with progressive, dull aching right ear pain and postural giddiness. Ear discharge was insidious in onset, intermittent and progressive, serous, scanty, foulsmelling and non-blood stained, with aggravation on water entry into the ear. There were no other aggravating or relieving factors. Giddiness was not associated with nausea or vomiting. The patient gave history of decreased hearing. There was no other significant history. On examination, there was a fistula present in the upper part of the right retro-auricular groove with its opening measuring 1cm X 0.5cm. (Fig. 1).

Mastoid tenderness was present on the right side. Tragal tenderness was absent, and fistula test was negative on both sides. Granulation tissue was present over the right posterosuperior bony external auditory canal wall (Fig. 2), due to which the right tympanic membrane could not be visualised.

The left tympanic membrane had a large central perforation. Facial nerve function was clinically normal bilaterally. Oto-endoscopy showed Grade IV

retraction of pars tensa with a posterosuperior marginal perforation. Cholesteatoma was present in the posterosuperior margin of pars tensa and posterior part of attic (Fig. 3).



Fig. 1: Clinical photograph of right postauricular cutaneous mastoid fistula (arrow)



Fig. 2: Granulation tissue (arrow) present over the right posterosuperior bony canal wall



Fig. 3: Right tympanic membrane with cholesteatoma (arrow) present in the posterosuperior quadrant of pars tensa and posterior attic.

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Scutum was partly eroded. **High-Resolution** Computed Tomography (HRCT) scan showed soft tissue within the right middle ear and mastoid air cells, with erosion of ossicles, scutum and tegmen tympani. There was also erosion of anterior and lateral wall of right mastoid cortex, communicating with the external auditory canal. Pure tone audiometry revealed mixed hearing loss. A diagnosis of bilateral CSOM, right AAD, active stage, left TTD, inactive stage with bilateral mixed hearing loss (right more severe than left) with right postauricular cutaneous mastoid fistula and right acute mastoiditis was established. Under intravenous antibiotic coverage, we performed a right modified radical mastoidectomy. The postauricular incision was modified superiorly to excise the cutaneous mastoid fistula. Intraoperatively. а labyrinthine fistula was noted at the dome of the lateral semicircular canal. The cholesteatoma was carefully dissected off the labyrinthine fistula, 2mm X 1 mm in dimension, without any perilymph leak. A piece of conchal cartilage was placed over the bony labyrinth defect to prevent pressure transmission in the future. After complete clearance of the disease, type 3B tympanoplasty was performed using conchal cartilage. Conchomeatoplasty was performed, and the wound was closed in layers without tension over the suture line. The postoperative period was uneventful. The patient was on regular follow up for the last one year with no evidence of recurrence.

# DISCUSSION

A postauricular mastoid-cutaneous fistula is an unusual condition that may arise due to chronic suppurative otitis media, tubercular otitis media, congenital cholesteatoma, mastoidectomy, extensive meatoplasty, or even natural exteriorisation of cholesteatoma from mastoid through postaural skin owing to its destructive nature (2-6). Bilateral occurrence of postaural fistula is not unknown (7). Apart from the associated postaural discharge, patients may seek medical consult for cosmetic purpose (6). Ventilating mastoid fistulas aid in preventing other lethal complications of CSOM (7).

Healing is gradual, may be spontaneous, or not at all (6). The inverted skin around the fistula may undergo necrosis, followed by epithelial migration and fusion with the mastoid cavity lining (8). Reports of closure of fistula with antibiotic therapy exist in literature (9). Surgical options include simple skin closure, temporalis muscle transplantation into the defect, temporalis fascia transposition flap, rotational skin flap technique, and postauricular myocutaneous flap (6,10). In our patient, a modified radical mastoidectomy was performed to deal with the cholesteatoma. The postaural skin incision was modified to facilitate excision of the postaural fistula.

Labyrinthine fistula occurs as a complication of CSOM in 4-12% of cases. It may affect the lateral semicircular canal (87%), promontory (8%), superior

semicircular canal (6%), and posterior semicircular canal (2%: 11). In the present case, a labyrinthine fistula was noted intraoperatively in the dome of the lateral semi-circular canal. Clinical features include giddiness (64%), positive fistula test (50%) and profound sensorineural hearing loss (15%). Hearing loss, when present, occurs due to the migration of toxins or inflammatory mediators into the inner ear (11). Our patient had giddiness and hearing loss, with a false negative fistula test, possibly resulting from the bulky cholesteatoma. Imaging studies detect fistula in 97% of cases; (11) however, HRCT could not establish this finding in our patient. The current consensus on surgical management of labyrinthine fistula is complete excision of epithelium from the fistula site and soft tissue grafting. Intraoperative steroid use has been recommended to preserve vestibular function (11). We chose to use conchal cartilage to cover the fistula site, followed by performing a type 3B tympanoplasty in our patient.

# CONCLUSION

Double fistulae in CSOM is an infrequent occurrence. A negative fistula test without evidence of fistula on HRCT is possible in the case of a labyrinthine fistula. Careful dissection of cholesteatoma from the medial wall of the attic is imperative in an unsafe ear with giddiness and mixed hearing loss. Complete excision of the postauricular cutaneous mastoid fistula is feasible, with consistent follow-up.

#### **CONFLICT OF INTEREST**

The authors declare no conflict of interest.

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