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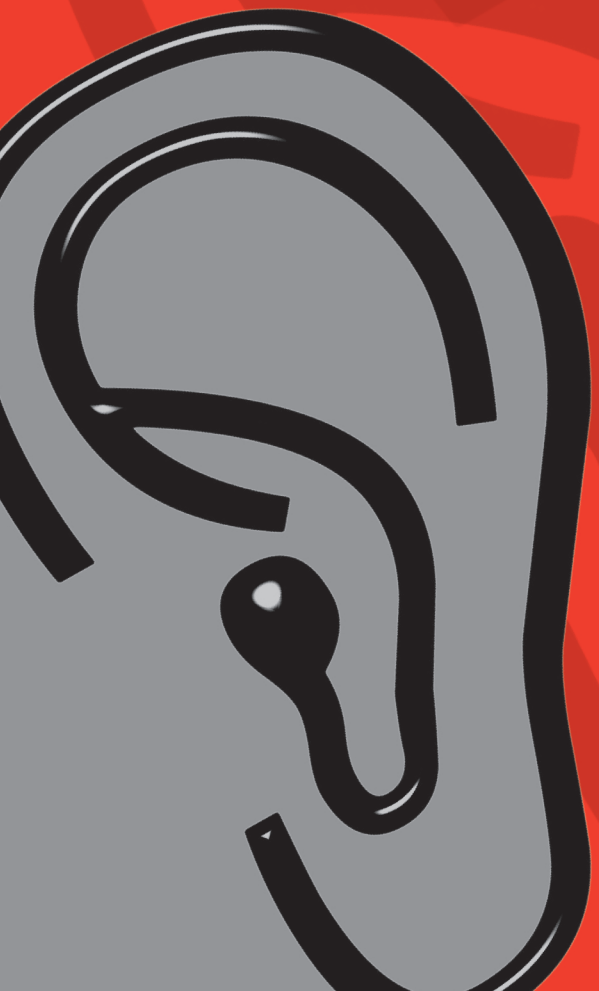

**Dr. M. K. Taneja**
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Case Report

# A Therapeutic Approach of Isolated Suppurative Labyrinthitis Complicated by Meningitis

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## Abstract

Suppurative labyrinthitis (SL), a major complication of otitis media, results from a bacterial infection spread through the round window to the inner ear. The most frequent symptoms are hearing loss, vertigo, nystagmus, nausea, and vomiting. This infection poses a significant risk of meningitis due to direct communication of the perilymph to the cerebrospinal fluid through the cochlear aqueduct. There are scarce data in the literature documenting isolated SL as the only cause of meningitis. There are no data describing which treatment is more effective, how often this complication occurs, or when to expect this fatal outcome. We report a case that developed from a labyrinthine suppurative infection to a late-onset meningeal infective disease. This case highlights the importance of correct and timely treatment of SL with early mastoidectomy. The indications of mastoidectomy in these cases are discussed, with a brief review of the literature.

**Keywords:** Mastoidectomy, meningitis, suppurative labyrinthitis

## Introduction

Suppurative labyrinthitis (SL) is a bacterial infection of the inner ear with the potential risk of meningitis. Symptoms of SL generally are a sensorineural hearing loss, tinnitus, and vertigo.[1] Interestingly, there is scarce data in the literature documenting isolated SL as the only cause of meningitis[2,3]. There are no data describing which treatment is more effective, how often this complication occurs, or when to expect this fatal outcome. In addition, the occurrence of SL without otorrhea or cholesteatoma is extremely rare. We report a case of an immunosuppressed adult patient, evolving from an otitis media and SL to late-onset meningitis. This case highlights the importance of correct and timely treatment of SL. The indications of mastoidectomy in these cases are discussed.

## Case Report

A 58-year-old female, diabetic and hypertensive with poor control presented to the ENT outpatient department of a secondary care hospital. Informed consent was obtained. She had a previous history of hysterectomy and appendectomy due to a tubo-ovarian abscess and left submandibular abscess drainage. She begun with suppurative otitis media and was treated with systemic levofloxacin and topical ciprofloxacin. She attended the clinic 2 weeks later with no symptoms and no perforation. Two weeks after, she complained of vertigo and dizziness, sudden unilateral right hearing loss and headache, otoscopy showed an opaque right tympanic membrane. The sensorineural right hearing loss was found [Figure 1], and tympanogram revealed curves A. There was no evidence of fever, neurologic focalization, or cerebellar disease. Computed tomography (CT) showed occupation of mastoid cells [Figure 2]. We suspected signs of meningism. Neurology assessment was requested, no evidence on CT [Figure 3] and clinical findings of central nervous system disease was found.

She was diagnosed by a certified otologist with SL, and tympanostomy tube was placed on the right ear, and no suppurative fluid was seen after myringotomy. After 4 days, she was discharged from the hospital with antibiotic treatment (ceftriaxone 10 days and clindamycin) and closed clinical surveillance. One month after, she presented to the emergency room with severe vomiting,

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somnolence, fever, headache, and intense vertigo. On physical examination, she presented with neck stiffness and fever. A lumbar puncture confirmed meningitis [Table 1]. Empirical treatment with antibiotics was prescribed (Vancomycin and Meropenem) insulin, and enalapril. Subsequently, she underwent mastoidectomy. Inflammatory mucosa in mastoid cells was found [Figure 4], amber secretion obtained from the mastoid was sent for culture. Cultures and biofilms were negative. Four months after, the patient is free of disease, tympanic membrane is normal and with no evidence of neurologic disease. The sensorineural hearing loss was unchanged.

**Table 1. Cerebrospinal fluid cytological and cytochemical analysis.**

| Parameters | Result |
|---|---|
| PH | 8.5 |
| Density | 1.01 |
| Color | Turbid |
| Glucose | 100 mg/dl |
| Total proteins | 82 mg/dl |
| LDH | 587 U/L |
| Leucocytes | 1440/mm$^3$ |
| Polymorphonuclear Neutrophil | 65% |
| Mononuclear | 35% |

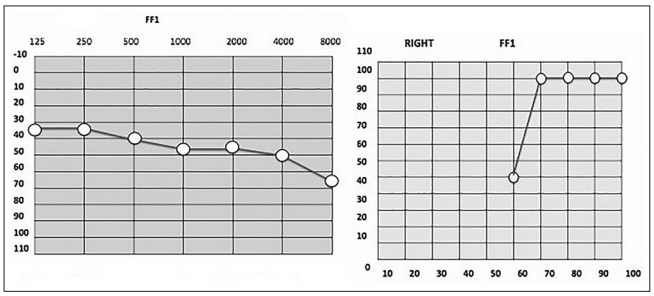


**Figure 1:** Audiometry. Right moderate sensorineural hearing loss. Speech reception threshold 100% at 70 db

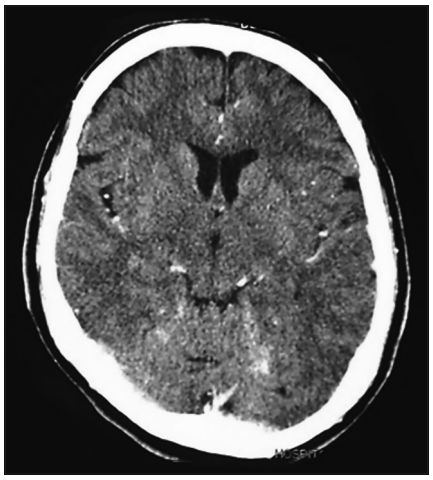

**Figure 3:** Computed tomography scan. Soft tissue window. Axial View. No pathological data was found associated with meningitis

## Discussion

SL is a diffuse bacterial infection of the labyrinth, which mainly occurs as a complication of otitis media. The incidence of labyrinthitis has been reduced considerably due partly to the development of new antibiotics and greater awareness of the complications of otitis media.[2-4] Labyrinthitis may develop from 4 to 24 months after a suppurative otitis media, and 12–38 months after meningitis.[2]

Complications of otitis media can be extracranial and intracranial, 47.1% and 30.6%, respectively; however, 22.4% of these combinations are observed.[3] The main intracranial complication is meningitis that occurs mostly when the involvement of the middle ear is chronic.[4] In previous reports, meningitis represents 37% of intracranial complications of otitis media.[1]

Although meningitis caused by labyrinthitis has been described, there are treatment controversies in the management

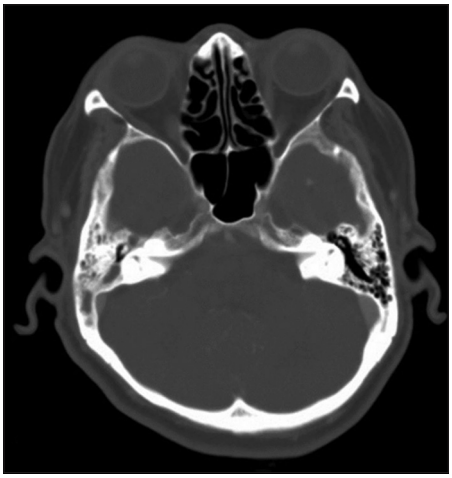

**Figure 2:** Computed tomography scan. Axial view shows right mastoid occupation with coalescence of mastoid cells

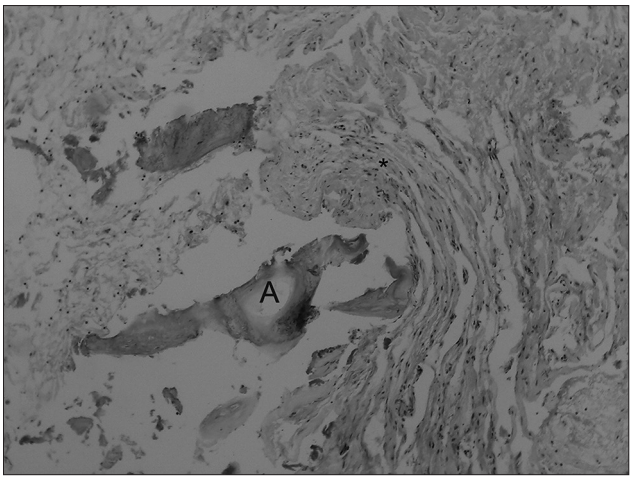


**Figure 4:** Histopathology image of mastoid sample. Hematoxylin and eosin stain shows inflammatory lymphocytic infiltrate (*) and fibro osseous material (a). No biofilms were found

of these patients. In the pathophysiology of labyrinthitis, it is known that suppurative middle ear secretions can access the central nervous system by direct or indirect means. Direct diffusion occurs through normal anatomical pathways (round and oval window) or through bone erosion areas (caused by cholesteatoma or granulations). After a labyrinth compromised, it may be possible an intracranial infection, being the cochlear aqueduct the most likely diffusion path.[5] Hematogenous dissemination occurs due to bacteremia or thrombosis of the emissary veins of the dura.[1,6] Although studies of temporal bones have not found any vascular damage, the presence of modiolar inflammation may be indicative of this form of dissemination.[5] Furthermore, the presence of biofilms are relevant to persistent infection. Nonetheless, samples of mastoid tissue were negative for biofilms in our patient.

The association of meningitis and middle ear disease in patients with diabetes or other immune suppression is described and contributes to the pathology.[7,8]

The clinical presentation of labyrinthitis is due to bacterial invasion of the labyrinth and is followed by the total loss of hearing and vestibular function. The patient suffers from sensorineural hearing loss and vertigo.[1] Treatment of SL is both surgical and medical, and timing is critical to minimize inner ear trauma or further complications. The main surgical treatment described previously includes pressure equalization myringotomy and tube placement, both are essential for the evacuation of middle ear disease as well as to obtain culture and sensitivities to tailor antibiotic therapy. Steroids should be considered if the sensorial neural hearing loss is persistent despite adequate antibiotic treatment.[1]

On the other hand, treatment of otogenic meningitis may include performing a radical mastoidectomy within the first 24 h; however, there is a high rate of recovery with only antibiotics.[9] Barry *et al*. recommended to start treatment with antibiotics and myringotomy alone; according to them, mastoidectomy should be reserved for cases of neurological deterioration or lack of improvement after 48 h of drainage and antimicrobial therapy.[6] Furthermore, Felisati *et al*. described three patients with otogenic meningitis, and normal tympanic membrane, whom underwent mastoidectomy. Interestingly, all purulent material was found in the mastoid, the middle ear was unaffected, which was attributed to aditus ad antrum blockage due to hypertrophy of the lining of the mastoid antrum. In these cases, myringotomy may have a limited effect, and antibiotic treatment with mastoidectomy may be essential. Isolated myringotomy in otogenic meningitis patients is controversial.[7] In addition, Singh and Maharaj performed a radical mastoidectomy for cases with otogenic meningitis and cholesteatoma, and a cortical mastoidectomy for patients with otitis media without cholesteatoma. Although due to the similar mortality rates for both groups, they concluded that mastoidectomy with canal wall up was sufficient to control meningitis.[9]

Our patient developed meningitis after otitis media and SL. Previously treated with antibiotic therapy, at the second visit probably presented with meningism, although no clinical evidence of neurological disease was found at that time.

In addition, in cases with SL and meningitis, Maranhao *et al*.[10] have described that labyrinthitis frequently occurs with other intracranial or extracranial complications. Meningitis and labyrinthitis were present in 21% of their patients. Nevertheless, we found no literature describing treatment for these patients, partly due to meningitis secondary to isolated labyrinthitis (noncholesteatomatous) is rare. However, it is clear that aditus ad antrum blockage due to hypertrophy of the lining mastoid cavity could be the source of invading organisms, and could explain the absence of suppurative fluid after myringotomy. Therefore, mastoidectomy could be indicated as a primary treatment in isolated SL cases. We describe the progression from suppurative otitis media to labyrinthitis, causing meningitis in the lapse of 3 months, when SL was diagnosed adequate antibiotic treatment and a prompt ventilation tube were indicated, and nonetheless, the patient develop 1 month after meningeal disease. The treatment of isolated SL is controversial but based on our experience we encourage early mastoidectomy in patients with isolated SL and immunosuppression. This can represent only a minimal additional risk and could prevent intracranial complications that can threaten a patient's life.

## Conclusion

Isolated SL should be managed with early mastoidectomy, particularly in immunocompromised patients.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

There are no conflicts of interest.

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