**Objective:** To analyze clinical presentation, preoperative diagnostic methods, surgical treatment and postoperative outcomes of labyrinthine fistula (LF) secondary to cholesteatomatous chronic otitis media.

**Materials and Methods:** Of 582 mastoid surgeries performed, a retrospective review of 33 patients with LF secondary to cholesteatomatous chronic otitis media was performed.

**Setting:** Tertiary referral center.

**Interventions:** Audiologic evaluation with pure tone bone conduction average (PTA-bone) and radiologic imaging studies with temporal bone computed tomography (CT) scanning were evaluated. Open technique with a removal of the cholesteatoma matrix and sealing the fistula site with connective tissue was preferred.

**Main outcome measures:** Auditory results were evaluated as a function of LF size and site.

**Results:** Incidence of LF was 5.7% (n:33). Isolated fistula of the lateral semicircular canal was found in 25 patients (75.7%), and isolated involvement of the oval window was found in one patient (3%). Multiple fistulas were detected in 7 patients (21.2%). In 31 patients (88.2%), the cholesteatoma was completely removed, and the fistula was sealed; and in 2 patients (5.8%) the matrix was left intact after exteriorization. One patient (3%) presented with anacusis. Postoperatively, hearing remained in the same range in 24 patients (72.7%). In 8 patients (24.2%) a reduction of PTA-bone was observed, and 2 patients (6%) presented with postoperative anacusis. Vertigo improved or disappeared in 14 (42.4%) cases, and remained unchanged in 4 (12.1%) cases.

**Conclusion:** LF is an aggressive complication of cholesteatomous chronic otitis media. The postoperative prognosis is correlated to the size and location of the LF.

**Introduction**

Labyrinthine fistula (LF), one of the major complication of cholesteatomatous chronic otitis media, can result in permanent loss of labyrinthine function. The incidence of LF secondary to chronic otitis media with cholesteatoma has been reported to range approximately between 3% and 12%.[1-4] The lateral semicircular canal is the most commonly affected site in nearly 90% of the patients, although other inner ear structures, either alone or in combination, may be involved.[1-4] Preoperative diagnosis and criteria for the surgical management of LF, with or without a perilymph leak, are complex and controversial.

Goal of the current study was to discuss clinical presentation, preoperative diagnostic methods our choice of surgical technique, intraoperative findings and postoperative outcomes in 33 patients who developed LF secondary to cholesteatomous chronic otitis media.

**Patients and Methods**

A retrospective review of the clinical records of 582 patients operated on for cholesteatomatous chronic otitis media between 1988 and 2007, identified 33 patients (5.7 %) with surgically confirmed LF. There were 16 women (48.4%) and 17 men (51.6%). Age range was 14 to 73 years with an average of 37.2 years.
A labyrinthine fistula was defined as a cholesteatomatous destruction of the bony otic capsule, exposing membranous labyrinth, with or without a perilymph leak. Cases with thinning of the bony labyrinth were not included. In the current study, LFs were classified in 3 groups, according to size: small (<1mm), medium (between 1 and 2 mm), and large (>2mm). In patients with more than one fistula site, the fistula with the greater size was taken into consideration.

Clinical records of 33 patients with LF were reviewed to identify: clinical symptoms, audiologic evaluations, radiologic imaging studies, technique of surgery adopted, location and size of LF and postoperative outcomes.

Preoperative examinations included bone conduction audiometry with pure tone averages (PTA-bone) at the frequencies of 0.5, 1, 2, and 4 kHz in decibels hearing level, categorized in 3 hearing ranges (<30 dB, between 31 and 50 dB, and >50 dB) and computed tomography (CT) scanning of the temporal bone. Pneumatic otoscopy was performed in all patients. The fistula test was considered positive if conjugate deviation of the eyes and subjective dizziness were induced by alternating pressure applied to the external auditory canal.

To ensure the eradication of the cholesteatoma, surgery with open techniques (classical or modified radical mastoidectomy) was performed. None of the patients had undergone prior tympano-mastoidectomy. Choice of the surgical technique was determined by the ipsilateral and contralateral hearing thresholds, location and size of the fistula and extent of cholesteatomatous destruction. The cholesteatoma matrix was preserved over the fistula site until the whole surgery was completed. Before the exploration of the fistula site, a gelatin sponge impregnated in ampicillin solution was applied to the fistula site for several minutes to reduce the potential infection. The matrix overlying the fistula was gently removed under constant irrigation with physiological saline and the fistula was immediately covered with temporalis fascia or tragal perichondrium.

Intraoperatively, intravenous corticosteroid (prednisone, 1mg/kg, to a maximum dose of 60mg) was administered; and during the postoperative period, an appropriate systemic antibiotic therapy was prescribed. The pure tone audiometry evaluation obtained within 3 months postoperatively was accepted for comparison.

“House and Brackmann facial nerve grading scale” (HBS) was used to evaluate preoperative and postoperative facial nerve function. Facial nerve decompression was tailored according to the facial nerve problem.

Chi-square testing was performed using the Statistical Package for the Social Sciences 9.0 for Windows (SPSS, Inc., USA) in order to calculate significance of factors analyzed. $p<0.05$ was considered significant.

**Results**

Of 582 surgeries performed for chronic otitis media with cholesteatoma, 33 patients with LF have been encountered. This represents 5.7% of the total number of cholesteatoma cases during this period. Twenty-one patients (63.6%) had unilateral chronic otitis media, and 12 patients (36.4%) had bilateral chronic otitis media.

The isolated fistula of the lateral semicircular canal was found in 25 patients (75.5%), and isolated involvement of the oval window (footplate) was found in one patient (3%). Multiple fistulas were detected in 7 patients (21.2%): simultaneous involvement of lateral semicircular canal and superior semicircular canal was found in 5 patients (15.1%), simultaneous involvement of lateral semicircular canal and posterior semicircular canal was found in 1 patient (3%), and simultaneous massive involvement of the three semicircular canals was found in 1 patient (3%).

The LFs were classified according to the size. Eighteen patients (51.5%) had small sized (<1mm), 10 patients (30.3%) had medium sized (between 1 and 2 mm), and 5 patients (15.1%) had large sized (>2mm) fistulas. Multiple fistulas (n=7, 21.2%) were medium and large sized (>1mm).
Clinical presentation
All patients presented with hearing loss. Fourteen patients (42.4%) had a history of tinnitus. Conductive, sensorineural and mixed type hearing loss were in 14, 11 and 8 patients respectively. Eighteen patients (54.5%) presented with vertigo, which was intermittent in 13 (39.3%), and persistent in 5 patients (15.1%). Duration of vertigo varied from 2 months to 3.5 years. Twenty patients (60.6%) had otorrhea in the involved ear, and 7 patients (21.2%) presented with otalgia. Six patients (18.1%) presented with various degrees of facial nerve dysfunction: HBS 2-3/6 (n=3), HBS 4/6 (n=2), HBS 5/6 (n=1).

Duration of otologic symptoms attributable to chronic otitis media varied from 2 weeks to 57 years. One patient with an attic cholesteatoma presented with facial nerve paralysis 2 weeks prior to surgery. Sixteen patients (48.5%) had otologic symptoms for more than 10 years.

Preoperative diagnostic methods
All patients were subjected to preoperative audiometric evaluation: PTA-bone was less than 30 dB in 12 patients (36.4%), between 31 and 50 dB in 14 patients (42.4%), and more than 50 dB in 7 patients (21.2%). One patient (3%) with a simultaneous LF of 3 semicircular canals had anacusis in the affected ear. The fistula test was positive in 14 patients (42.4%), all of whom complained of vertigo preoperatively. Five patients (15.1%) with persistent vertigo had positive fistula test.

CT scanning of the temporal bone was done preoperatively in all patients (n=33) and radiographically suspected LFs were elicited intraoperatively in 11 patients (33.3%) with moderate (n=6, 18.2%), and large (n=5, 15.1%) sized fistulas (Figure 1). The remaining CT studies revealed findings related to cholesteatoma with an intact otic capsule.

Surgical management
The choice of the surgical technique was determined by the criteria previously stated. Twenty-one patients (63.6%) had classical radical mastoidectomy, and 12 patients (36.4%) had modified radical mastoidectomy performed. In 31 patients (93.9%), the cholesteatoma was completely removed, and the fistula was sealed. The cholesteatoma was exteriorized, and the matrix was left in situ in one patient (3%) with a LF of the oval window, and in one patient (3%) with LFs of the lateral and posterior semicircular canals. A significant adherence between the cholesteatoma and the underlying membranous labyrinth was not detected.

The materials inserted to seal the fistula was temporalis fascia graft (n=23, 69.7%) and tragal perichondrium (n=10, 30.3%). Tympanomastoid obliteration was performed with an abdominal fat graft and fibrin glue in 2 cases (6%).

Facial decompression was performed in 7 patients (21.2%) with facial nerve dysfunction. Three patients (9.1%) had facial canal destruction isolated to the tympanic segment, whereas 3 patients (9.1%) had tympanic and mastoid involvement. One patient (3%) underwent petrous apicectomy with facial decompression of the vertical and horizontal segments and the first genu.

Fourteen patients (42.4%) had cholesteatoma extending to supratubal recess. Six patients (18.1%) had dehiscence of tegmen with open dura, with one patient (3%) having a sagging of the temporal lobe. In 2 patients (6%), conchal cartilage was used with fibrin glue at the tegmen tympani to support the dura. We did not put anything to the tegmen in four patients with dehiscence of tegmen because of minimal dehiscence. Three patients (9.1%) had external auditory canal
destruction. One of these patients (3%) had polyp protruding and occluding the external auditory canal, and 2 patients (6%) had postauricular subperiosteal abscess.

**Postoperative outcome**

The average period of follow-up was 4.2 years (8 months-8.5 years). One patient (3%) presented with anacusis. Hearing remained in the same hearing range in 24 patients (72.7%). In 8 patients (24.2%) a reduction of PTA-bone was observed, going from 5 to 34 dB hearing level, and 2 patients (6%), in the groups with medium and large sized LF, presented with postoperative anacusis (Tables 1 and 2). Three patients (9.1%) with a LF of the lateral semicircular canal in the small and medium sized LF groups, had an improvement in PTA-bone of 13 dB on average, but remained within the boundaries of the same hearing range.

Of the 12 ears (36.4%) with a preoperative PTA-bone of less than 30 dB hearing range, the postoperative PTA was in the same range in 9 patients (27.2%) (Table 1). In the remaining 3 ears (9.1%), the PTA fell to 31 to 50 dB hearing range postoperatively.

Of the 14 ears (42.4%) with a preoperative PTA-bone between 31 and 50 dB hearing range, the postoperative PTA was in the same range in 10 patients (30.3%), and more than 50 dB in 3 patients (9.1%) (Table 1). One patient (3.1%) had anacusis postoperatively. The patient (3.1%) with a LF of the oval window presented with PTA-bone of 43 dB, the postoperative PTA-bone fell to 77 dB.

Of the 6 patients (18.2%) with a PTA-bone more than 50 dB hearing range, the postoperative PTA-bone was in the same range in 5 patients (15.2%), but one patient (3%) had postoperative anacusis (Table 1).

In large sized LFs, auditory function was negatively influenced (Table 2; p<0.05). Postoperatively, in patients with medium sized LF group (n=11, 33.3%), PTA-bone of 4 patients (12.1%) fell to 31-50 dB hearing range, 5 patients (15.1%) had PTA-bone in more than 50 dB hearing range, and 1 patient (3%) had anacusis. In patients with large sized LF group (n=5, 15.1%), PTA-bone of 3 patients (9.1%) fell to more than 50 dB hearing range, and 2 patients (6%) had anacusis. In small sized LF group (n=18, 54.5%), all patients had postoperative PTA-bone of less than 50 dB.

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**Table 1.** Comparison of preoperative and postoperative pure tone averages for bone conduction audiometry (number of ears / %).

<table>
<thead>
<tr>
<th>Preoperative PTA-bone</th>
<th>Postoperative PTA-bone</th>
</tr>
</thead>
<tbody>
<tr>
<td>&lt; 30 dB (n=12, 36.4%)</td>
<td>31-50 dB</td>
</tr>
<tr>
<td>&lt; 30 dB</td>
<td>9 / 27.2%</td>
</tr>
<tr>
<td>31-50 dB (n=14, 42.4%)</td>
<td>–</td>
</tr>
<tr>
<td>&gt;50 dB (n=6, 18.2%)</td>
<td>–</td>
</tr>
<tr>
<td>Anacusis (n=1, 3%)</td>
<td>–</td>
</tr>
</tbody>
</table>

*PTA-bone, pure tone average for bone conduction.*

**Table 2.** Comparison of preoperative and postoperative pure tone averages for bone conduction audiometry versus size of LF (number of ears / %).

<table>
<thead>
<tr>
<th>Size of LF</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
</tr>
</thead>
<tbody>
<tr>
<td>Small (&lt;1mm) (n=18, 51.5%)</td>
<td>10 / 30.3%</td>
<td>8 / 24.2%</td>
<td>8 / 24.2%</td>
<td>10 / 30.3%</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Medium (1-2mm) (n=10, 30.3%)</td>
<td>2 / 6%</td>
<td>1 / 3%</td>
<td>6 / 18.2%</td>
<td>4 / 12.1%</td>
<td>2 / 6%</td>
<td>4 / 12.1%</td>
<td>–</td>
<td>1 / 3%</td>
</tr>
<tr>
<td>Large (&gt;2mm) (n=5, 15.2%)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>4 / 12.1%</td>
<td>3 / 9.1%</td>
<td>1 / 3%</td>
<td>2 / 6%</td>
</tr>
</tbody>
</table>

*PTA-bone, pure tone average for bone conduction; Preop, preoperative; Postop, postoperative.*
Although, there was no statistically significant difference, auditory function was worse in patients with multiple LFs, (Table 3; p>0.05). All of our patients with multiple LFs, had preoperative PTA-bone of more than 30 dB hearing level (n=7). Postoperatively, of the patients with multiple LFs (n=7, 21.2%), PTA-bone fell to 31-50 dB hearing range in 2 patients (6%), and more than 50 dB hearing range in 2 patients (6%). Three patients (9.1%) had anacusis: one of these patients with LFs of the lateral and posterior semicircular canals had matrix left in situ over the fistula site after exteriorization.

Vertigo improved in 14 (42.4%) cases, and remained unchanged in 4 (12.1%) cases. All patients were able to return to their original activities. Postoperative hearing was worse in ears with a preoperative positive fistula test (n=13, 39.4%) compared to those with negative tests (n=21, 63.6%): the average PTA-bone for the patients with positive fistula test was 38 dB, versus 31.5 dB for the patients with negative fistula test. Tinnitus improved in 5 (15.1%), persisted with minor intensity in 2 (6%), and remained unchanged in 3 patients (9.1%). Facial paralysis improved in 3 (9.1%), and completely recovered in 2 (6%) of the 7 patients (21.2%). Facial nerve function remained unchanged in 2 patients (6%), with HBS scores of 3/6 and 4/6, after an average follow-up period of 13 months. No postoperative facial paralysis occurred. In 2 patients (6%), the mastoid cavity was revisited, after an average period of 13 months. One patient had persistent otorrhea, and the other patient developed a cholesteatoma pearl in the mastoid bowl.

**Discussion**

In our otologic practice, the incidence of LF complicating cholesteatomatous chronic otitis media was 5.7%, which is similar to those documented in the literature. The lateral semicircular canal was the most common site involved (75.5%) in our study, as in previously reported series. Definitive preoperative diagnosis of LF is not always possible. An extensive clinical examination and modern imaging techniques are essential for the preoperative diagnosis of LF. The presence of decreased bone conduction threshold in association with vertigo, and/or otorrhea should alert the otolaryngologist to the possibility of LF. The long term disease and facial nerve dysfunction also rises the suspicion of LF.

The fistula test does not have a predictive value in preoperative diagnosis of LF, with false-negative rates approximately as high as 50%. The range of true-positive results varies approximately from 20% to 70% 8-10 and in our study, this finding was positive in 38.2% of the cases, which squares with reports by other authors.

Preoperative CT imaging is a more reliable indicator for the assessment of LF. The large sized LFs are easier to be detected on CT scans due to an aggressive

**Table 3.** Comparison of preoperative and postoperative pure tone averages for bone conduction audiometry versus location of LF (number of ears / % ).

<table>
<thead>
<tr>
<th>Size of LF</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
<th>Preop</th>
<th>Postop</th>
</tr>
</thead>
<tbody>
<tr>
<td>LSCC</td>
<td>11/25</td>
<td>9/33.3</td>
<td>27.3%</td>
<td>9/27.3</td>
<td>30.3%</td>
<td>15.1%</td>
<td>18.2%</td>
<td></td>
</tr>
<tr>
<td>LSCS &amp; SSCC</td>
<td>–</td>
<td>–</td>
<td></td>
<td>3/2</td>
<td>2/9.1</td>
<td>6/6</td>
<td>9.1%</td>
<td>6%</td>
</tr>
<tr>
<td>LSCC &amp; PSCC</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>LSSS &amp; SSCC &amp; PSCC</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
<tr>
<td>Oval window</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>1/3</td>
<td>–</td>
<td>–</td>
<td>–</td>
</tr>
</tbody>
</table>

PTA-bone, pure tone average for bone conduction; LSCC, lateral semicircular canal; SSCC, superior semicircular canal; PSCC, posterior semicircular canal.
otic capsule destruction. Small and medium sized LFs (<2mm) may be missed, depending on the positioning of the patient. In our series, the presence of LF has been detected in 11 patients (33.3%) by preoperative CT examination. Preoperative CT imaging was more sensitive for diagnosing large sized LFs (of 5 large sized LFs, 4 were revealed versus 5 of 11 medium sized LFs).

The management of the LF still represents a challenge for the otolaryngologist. The criteria considered in the management of LF before deciding the surgical approach are the surgeon’s ability and experience, extent of cholesteatomatous destruction, location and size of the fistula, hearing thresholds of both the ipsilateral and contralateral ears and revision status of the surgery.

The optimal surgical technique for LF complicating cholesteatomatous chronic otitis media is controversial. Some surgeons prefer open technique, providing a more extensive intraoperative exposure and a safer postoperative observation whereas others recommend closed technique. Removal of the cholesteatoma matrix is another controversial issue in the management of LF. Use of various approaches depend on the matrix removal: matrix left undisturbed at the fistula site with an exteriorization through an open cavity, matrix removed completely and matrix preserved at the first stage, and then removed at a planned second stage.

The surgeon must utilize a delicate technique and attempt to remove the matrix by developing a dissection plane between the membranous labyrinth and matrix under constant irrigation. Matrix preservation is reported to be dangerous for the development of a delayed sensorineural hearing loss due to further progression of disease and supplicative labyrinthitis. Collagenase activity of the cholesteatoma, persistent inflammation and pressure exerted are responsible for continued erosion of the underlying bony labyrinth.

Significant forces in removal of the matrix may result in destruction throughout the membranous labyrinth, and deterioration in bone conduction threshold. If the matrix is adherent to the membranous labyrinth, it is best to leave the matrix in place. Current indications for an exteriorization technique are confined to LFs in an only hearing ear, fistula of cochlea or vestibule, or very large fistulas (>2mm). Our policy is to remove matrix carefully whenever feasible, and then seal the fistula with temporalis fascia or tragal perichondrium. Upon the discovery of a LF, we tried to perform an open and a more radical surgery, because many of our patients presented with an advanced disease, and some had coexisting complications due to cholesteatoma (facial nerve involvement, postauricular subperiosteal abscess, posterior canal wall destruction and tegmen erosion) which makes a more radical approach necessary. Removal of the canal wall enhances the exposure needed in efficiently removing the pathology, and was considered less risky for postoperative follow-up. Our surgical routine of matrix preservation overlying the fistula site until the end of surgery protects the labyrinth against continuous washing or accidental aspiration of the perilymph, and thus lowers the risk of sensorineural hearing loss.

Our hearing results correlated with the literature. Incidence of postoperative deterioration in bone conduction thresholds with the practice of removing the cholesteatoma matrix, has been reported in up to 70% of the patients. In the current study, 6 patients (18.1%) with medium and large sized LFs, had postoperative worsening of the bone conduction threshold range. Surgical removal of the matrix entered the labyrinth, and 2 (6%) patients in the medium and large sized LF groups, with a preoperative PTA-bone of more than 50 dB hearing level, presented with postoperative anacusis. One of these patients had matrix peeled off the fistula site, whereas the other patient with LFs of the lateral and posterior semicircular canals had matrix preserved over the fistula site with exteriorization. Postoperative anacusis after matrix preservation in a patient with simultaneous LFs of the lateral and posterior semicircular canals can be explained by the presence of a labyrinthine invasion and labyrinthitis.

In large sized LFs, auditory function was negatively influenced (p<0.05). Postoperatively, (n=14) of the patients with medium (n=11) and large (n=5) sized fistulas, had PTA-bone of more than 30 dB hearing level.
Although, statistically insignificant, multiple LF's caused increasing impairment of hearing function (p>0.05). All of our patients with multiple LF's, had preoperative PTA-bone of more than 30 dB hearing level (42.8% between 31 and 50dB, and 42.8% in more than 50dB hearing range; and 28.5% had anacusis). Postoperatively, of the patients with multiple LF's (n=7, 100%), PTA-bone fell to 31-50 dB hearing range in 2 patients (28.5%), and more than 50 dB hearing range in 2 patients (28.5%). Three patients (42.8%) had anacusis. Postoperative bone conduction threshold improvement was observed in 3 ears (9%) after surgery. It has been speculated that, this phenomenon related to the resolution of an initial labyrinthitis due to cholesteatoma.\[4, 8, 10]\n
Additional complications of chronic otitis media can exist in the presence of LF.\[4, 9\] The incidence of facial paralysis is higher in patients with LF.\[8\] Facial nerve involvement necessitated facial nerve decompression tailored according to the facial nerve problem in 7 patients (21.2%) in our series. Incidence of tegmen dehiscences in patients with LF's was 2.6 times that which was found in patients without a LF.\[2,4,5,9\] Tegmen dehiscences were detected in 6 patients (18.1%), in our series. Posterior canal wall destruction (n=3) and postauricular superiosteal abscess (n=2) have been encountered.

**Conclusion**

LF is an aggressive complication of cholesteatomous chronic otitis media. Postoperative prognosis is correlated to the size and location of the LF. Objective in treating LF secondary to cholesteatomatous chronic otitis media is to conserve labyrinthine function and prevent complications, but a larger fistula (>2mm) endangers the labyrinth function, with a deterioration in bone conduction threshold. An appropriate surgical technique must be undertaken.

**Acknowledgement**

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