ORIGINAL ARTICLE

Management of Labyrinthine Fistula and Accompanying Findings: The Coexistence of Labyrinthine Fistula and the Facial Canal Dehiscence

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Submitted: 14 April 2008 Revised: 10 July 2008 Accepted: 17 July 2008

Mediterr J Otol 2008; 4: 132-137

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OBJECTIVE: To describe the audio-vestibular results of labyrinthine fistula surgery in patients with cholesteatoma.

PATIENTS AND METHODS: Data of 185 patients who had undergone surgery for cholesteatoma between 2001 and 2007 were reviewed. Three-layer sealing was used for the management of fistula.

RESULTS: Twenty patients were found to have labyrinthine fistula, of which 11 (55%) were male and 9(45%) female. Fistula wase located in lateral semicircular canal in all cases. Correlation of labyrinthine fistula and facial nerve dehiscence was statistically significant. Follow up was done for 1-6 year. Postoperatively, vertigo disappeared in 19 (95 %) patients. Hearing remained unchanged in 18 (90 %) patients. Worsening in bone conduction thresholds was observed in 2 (10 %) patients. Postoperative deafness did not occur.

CONCLUSION: Possibility of facial nerve dehiscence and tegmen defect should be considered in patients with labyrinthine fistula. Three-layer sealing may be a valuable technique in surgical treatment of labyrinthine fistula, lowering the risk of cochleovestibular functions.

Cholesteatoma is a pocket or cystic lesion consisting of stratified squamous epithelium and proliferative keratin within the temporal bone. The invasive expansion and keratin accumulation may cause bony destruction, hearing impairment, facial nerve paralysis, and labyrinthine fistula, as well as intracranial complications such as brain abscess and meningitis [1]. The expression of various cytokines, prostaglandins, nitric oxide, neurotransmitters, and growth factors is known to be associated with chronic inflammation in cholesteatoma - induced bone resorption [2,3].

At present, there is substantial morphological evidence that osteoclastic resorption is the major mechanism of bone destruction in cholesteatoma [1, 3, 4].

Labyrinthine fistula represents an erosive loss of the endochondral bone overlying the semicircular canals without loss of perilymph. This loss of the overlying protective bone allows pressure or mass- induced motion of the underlying endosteum, perilymph, and by contiguity, the endolymphatic compartment, evoking vestibular and sometimes auditory symptoms.

The overwhelming majority of labyrinthine fistulas involve only the lateral semicircular canal (LSCC). In few instances only do these erosions expose the superior or posterior semicircular canal, the vestibule, or the cochlea. This bone resorption is almost exclusively secondary to cholesteatoma [5].

Labyrinthine fistula (LF) is encountered during surgery for cholesteatoma with an average frequency of 2.9% to 21% $^{[6.7,8.9,10,11,12,13,14,15]}$ (Table 1).

Main treatment of labyrinthine fistula is surgical. Removal of the fistula generally improves the vestibular symptoms. Loss of hearing in the involved ear or worsening of existing hearing are the risks that always accompany the procedure, but this has been reported to occur in less than 20% of carefully managed fistula cases^[5].

The aim of our study was to analyze accompanying mastoid findings at surgery of LF and to describe our surgical technique without risk for the cochleovestibular functions

MATERIALS AND METHODS

We reviewed records of 185 patients who had undergone cholesteatoma surgery between 2001 and 2007. All surgeries were performed by one of us (M or N). Tympanomastoidectomy canal wall up (CWU) or canal wall down (CWD) was performed. Primary surgery was performed in 170 ears (89 male and 81 female patients) and revision surgery in 15 ears (6 male and 9 female patients). LF was routinely searched for during microscopic surgery, and its size and location were identified and recorded.

Table-1: Studies with reported incidence of labyrinthine fistula, location of fistula

| Author/year | N | Fistulas | Fistulas (%) | scc | Cochlea |
|------------------|------|----------|--------------|-----------|---------|
| Sheehy 1978 | 1024 | 97 | 9.5 | 83 | 14 |
| Sanna 1984 | 701 | 88 | 12.5 | 78 | 10 |
| Parisier 1991 | 426 | 41 | 10 | 34 | 7 |
| Dornhoffer 1995 | 1265 | 37 | 2.9 | 35 | 2 |
| Gersdorff 2000 | 769 | 54 | 7 | 48 | 6 |
| Lin 2004 | 117 | 7 | 6 | 7 | _ |
| Manolidis 2000 | 111 | 23 | 21 | 21 | 2 |
| Ahmad 2000 | 382 | 25 | 6.45 | 24 | 1 |
| Portier 2005 | 382 | 22 | 5.8 | 22 | _ |
| Soda- Merhy 2000 | 360 | 27 | 7.5 | 26 | 1 |
| Overall | 4437 | 421 | 378(89.8%) | 43(10.2%) | |

N, Number of patients in study examined; SCC, semicircular canal.

We attempted to preserve the cholesteatoma matrix over the fistula during bone drilling with copious irrigation. At the end of operation the matrix overlying the fistula was gently removed, and the area was sealed with three layers: temporalis fascia, bone dust, temporalis fascia (sandwich technique). The bone dust was obtained from non-inflamed bone cortex prior to removal of the matrix. In patients with only hearing ear, we left the matrix overlying the fistula. Canal wall down procedure was performed in all patients with LF.

Symptoms and audiological/vestibular findings were collected pre and postoperatively. Follow- up ranged from 1- 6 years. Data were analyzed using Chisquare and Fisher's exact tests.

Table-2: Correlation between incidence of labyrinthine fistula and facial nerve dehiscence in 185 ears

| Labyrinthine fistula | | | |
|----------------------|---------|--------|-------|
| Facial nerve | | | Total |
| dehiscence | | | |
| | Present | Absent | |
| Present | 11 | 44 | 55 |
| Absent | 9 | 121 | 130 |
| Total | 20 | 165 | 185 |

P= 0.009 (Chi-square test)

Table-3: Correlation between incidence of labyrinthine fistula and dural exposure of mastoid and tympanic tegmen in 185 ears.

| Lal | Labyrinthine fistula | | | |
|--------------|----------------------|--------|-------|--|
| Facial nerve | | | Total | |
| dehiscence | | | | |
| | Present | Absent | | |
| Present | 5 | 17 | 22 | |
| Absent | 15 | 148 | 163 | |
| Total | 20 | 165 | 185 | |

P= 0.009 (Chi-square test)

RESULTS

In 185 patients with cholesteatoma, 20 patients (10.8 %) had LF:, 19 with a single fistula, and one with 2 fistulas, all in LSCC.

LF was identified in 18 (10.6 %) of 170 ears during primary procedure and 2 (13.3%) of 15 ears during revision procedure. The difference in incidence between primary and revision surgery groups was not significant statistically using the Fisher's exact test (p=0.501). Facial nerve dehiscence (FND) was found in 55(29.73%) patients (47 tympanic segments, 4 mastoid segments, 4 tympanic and mastoid segment together). Of these, 11 patients also had LF. Association of LF and FND proved to be statistically significant (p=0.009) using Fischer exact test. (Table2).

Dural exposure of tympanic and mastoid tegmen was found intraoperatively in 22 (14 mastoid tegmen and 8 tympanic tegmen) patients, of whom, 5 having also LF. No significant difference was found between LF incidence and presence of dural exposure of tegmen in Fisher's exact test (Table3).

Of the 20 patients with LF, 11 (55%) were male and 9 (45%) female. Age of patients ranged from 11 to 69 years, with an average of 34.1 years. As for preoperative symptoms and audiological findings, hearing loss was present in 20 (100%), otorrhea in 19 (95%), vertigo in 12 (60%) and tinnitus in 8 (40%) patients. Audiogram showed pure conductive hearing loss in 3 (15%) patients, and mixed hearing losses in 16 (80%) patients. Only one (5%) patient had deafness. The fistula test was performed in all patients. The test was positive in 9 (44.5%) patients with LF.

In patients with LF, we used the CWD tympanomastoidectomy in 17 (85%) patients and radical mastoidectomy in 3 (15%) patients. None of the patients had only hearing ear.

During postoperative follow up vertigo disappeared in 95% of the patients within two months following the surgery. Vertigo did not occur postoperatively.

Hearing remained unchanged in 18 (90%) patients; however, in 2 (10%) patients worsening in bone conduction thresholds was observed varying from 5 to 20 dB. Deafness did not occur post operatively.

DISCUSSION

LF is a common problem in surgery for chronic otitis media (COM). In 10 well-documented series where fistulas were reported, the range in incidence was from 2.9 % to 21% (6-15) (Table 1).

Incidence of LF in our study was 10.8 %. In Manolidis study a higher incidence of 21% was found mainly because the majority of cases were revision surgeries [8]. In our study incidence of coexisting FND and dural exposure was higher, and there was a positive statistical association between FND and LF. One should always suspect that fallopian canal may be eroded and the facial nerve in direct contact with cholesteatoma whenever a LSCC fistula is observed or vice versa. One should also consider the possibility of defects in the tegmen.

In Manolidis study two associations with LF were prominent. The facial nerve was involved with cholesteatoma in 60% of the patients, and dehiscence of the tegmen occurred in 39%. These figures in our study were 55% and 25%, respectively.

The majority of Manolidis' cases were revision cases, and this increases incidence of dehiscence of tegmen and FND $^{[8]}$.

Two major surgical techniques are used for treatment of LF. Some surgeons adopt conservative methods, leaving the matrix of the cholesteatoma overlying the fistula, whereas others remove it and seal the fistula. Both techniques are acceptable, however, in the first technique it is considered that exposing the labyrinth can damage the cochlea and cause postoperative sensorineural hearing loss, whereas, in the latter technique the matrix is removed to prevent residual cholesteatoma. When matrix is left, bone erosion will continue because of matrix collagenase responsible for

bone resorption,. Another reason to do so is to eradicate the disease completely to obtain a dry ear.

Our policy was to remove the matrix overlying the fistula with any size and then seal the fistula with three layers. We practiced the open technique because majority of the patients had advanced cholesteatoma exhibiting bone destruction. In patients with only one hearing ear, we may consider leaving the matrix of the cholesteatoma overlying the fistula.

Use of the open technique as opposed to the closed technique is a controversial issue. Sanna et al (1984) and Sheehy and Brackmann (1979) recommend the closed technique in two stages, removing the matrix in the second stage [14, 16].

The closed technique has been recommended in studies by Palva and Ramsay^[17] and Ostri and Pederson ^[18]. The matrix was always removed during the first operation.

Based on 8 studies available, majority of patients (49.6%) with LF underwent a canal wall down procedure. The next commonest procedure was canal wall up (42.2%). (Table 4).

The incidence of pre- and postoperative deafness was reported in 7 studies (Table 5). The average preoperative deafness was found in 11.65%, whereas the average surgical deafness was observed in 9.04%.

In our study, none of the patients ended up with postoperative deafness. We prefer removing the matrix overlying the fistula at the end of the surgery since earlier uncovering, irrigation, or accidental aspiration of the labyrinth may increase the possibility of hearing damage.

The other advantage of this technique (sealing the fistula with three layers), is that it decreases the possibility of postoperative vertigo during cavity cleaning.

A detailed clinical examination is essential for the preoperative diagnosis of LF. In our study, 95% of patients had otorrhea, and 60% had vertigo, which supports the idea that presence of LF should be strongly suspected in patients with otorrhea and vertigo. The fistula test was positive in only 44.5% of

Table-4: Studies with reported incidence of surgical procedures for labyrinthine fistula.

| Author/ year | Fistulas | CWD | CWU | Radical | Subtotal petrozectomy |
|------------------|----------|------------|------------|----------|-----------------------|
| Sheehy 1978 | 97 | 13 | 72 | 12 | - |
| Parisier 1991 | 41 | 39 | 1 | 1 | _ |
| Sanna 1985 | 88 | 31 | 43 | 14 | _ |
| Portier 2005 | 22 | 17 | 5 | _ | _ |
| Ahmad 2002 | 25 | 25 | _ | _ | _ |
| Gersdorff 2000 | 54 | 20 | 34 | _ | _ |
| Manolidis 2000 | 23 | 17 | 2 | 2 | 2 |
| Soda- Merhy 2000 | 27 | 25 | 2 | _ | _ |
| Overall | 377 | 187(49.6%) | 159(42.2%) | 29(7.7%) | 2(0.5%) |

CWD: canal wall down; CWU: Canal wall up

Table-5: Preoperative and postoperative deafness as a result of fistula.

| Author/year | Fistulas | Surgical deafness (%) | Preoperative deafness (%) |
|------------------|----------|-----------------------|---------------------------|
| Parisier 1991 | 40 | 18(7) | 20 (8) |
| Sheehy 1978 | 97 | 13(13) | 12(12) |
| Sanna 1984 | 88 | 5.7(5) | 4.5(5) |
| Dornhoffer 1995 | 37 | 8.1(4) | 11(3) |
| Gersdorff 2000 | 54 | 18. 5(10) | 6.3 (5) |
| Manolidis 2000 | 23 | 0(0) | 13(3) |
| Soda- Merhy 2000 | 27 | 0 (0) | 14.8(4) |
| Overall | 366 | 9.04 (39) | 11.65 (45) |

our patients. In fact, fistula test may have false-negative or false-positive results varying from 33% to 50% [19]. Therefore, we do not consider this test to be predictive of labyrinthine damage [15, 18].

In this study, there was a positive association between FND and LF; furthermore, surgeons should consider the possibility of FND and defects in the tegmen in patients with LF.

We demonstrated that open technique with removal of the matrix and sealing with three layers may be a valuable choice for the surgical treatment of LF with little risk for cochleovestibular functions.

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