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LABYRINTHINE FISTULA IN CHOLESTEATOMA AND ITS MANAGEMENT-A RETROSPECTIVE STUDY

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ABSTRACT

Introduction- Labyrinthine fistula is one of the most common intra-temporal complications of cholesteatoma. Objective of study is (1) to evaluate the role of CT Scan in detecting labyrinthine fistulas, (2)to establish fact that fistula grade and location have no correlation with surgical approaches(CWU Vs CWD mastoidectomy approach),(3) to study the impact on hearing after treatment of labyrinthine fistula and(4) establish a management protocol to deal with labyrinthine fistula.

MATERIAL AND METHODS

Retrospective study done for 20 patients of labyrinthine fistula out of 149 cholesteatoma patients who underwent surgery for cholesteatoma in a tertiary care hospital between February 2015 to January 2020. Data collected for preoperative clinical signs and symptoms, HRCT temporal bone 0.5 mm cuts both axial and coronal cuts, Intra-operative fistula grade, site and surgical technique and pre and post operative audiometric outcomes.

RESULTS

Preoperative HRCT temporal predicted fistula in 15(75%) patients. Using the Dornhoffer and Milewski classification, 15(75 %) patients were identified as stage 1, 4(20%) patients as stage II, and 1(5%) patient as stage III. Post operatively 4(20%) patients showed improvement,13(65%) showed no change and 3(15%) showed deterioration in sensorineural hearing. There is statistically significant relationship between grade of labyrinthine fistula and the hearing outcome postoperatively (p value=.036).

Conclusion- Clinical signs and symptoms of labyrinthine fistula are not diagnostic preoperatively. HRCT temporal bone is mandatory in making the diagnosis of labyrinthine fistula preoperatively. Choice of surgery between canal wall up(CWU) or canal wall down(CWD) mastoidectomy is independent of the type of fistula. Although there is an inverse correlation between fistula grade and post operative sensorineural hearing outcome yet overall preservation of hearing is good. Complete and meticulous removal of the cholesteatoma matrix over the fistula in a single staged procedure and its repair using temporalis fascia followed by bone dust is a safe and effective procedure to treat labyrinthine fistula.

KEYWORDS

Labyrinthine fistula , Lateral semicircular canal ,Cholesteatoma,High resolution computed tomography ,Bone conduction threshold

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INTRODUCTION

Labyrinthine fistula is a known complication of cholesteatoma. Erosion of labyrinthine block is caused by multiple factors like: pressure necrosis, ischemia, osteoclasts activation, reduced pH levels, inflammatory mediators related to infection and osteolytic enzymes [1]. About 87 % of labyrinthine fistula are located over the lateral semicircular canal (LSCC) [2]. Preoperative clinical assessment is neither sensitive nor specific for diagnosing labrynthine fistula [2,3]. Prediction of labyrinthine fistula can be done by doing high resolution computed tomography (HRC) temporal bone preoperatively [4]. According to the depth of labyrinthine fistula Palva and Ramsay [5] gave 4 stage classification and Dornhoffer and Milewski [6] gave 3 stage classification. According to size sanna et al [7] divided fistula into 3 stages (small .5-1mm,medium 1-2 mm and large >2mm) and Ikeda et al 8 divided labrynthine fistula into 2 stages - small(< 3mm) and large group(>3mm). Management of labyrinthine fistula is controversial, Broadly two techniques are advocated in literature : first complete removal of cholesteatoma matrix over fistula site with immediate repair, to eradicate potential source of bone resorption [5,8,9] and second advocates leaving the matrix over fistula site undisturbed for maintenance of hearing and vestibular function [10, 11]

Severe sensorineural hearing loss has been reported between 3% to 37% in patients who underwent surgical management of labyrinthine fistula [12-15]

The aim of the our study is to evaluate the role of HRCT temporal bone in detecting labyrinthine fistula preoperatively, to show that fistula grade and location have no correlation with surgical approaches (canal wall down(CWD) versus canal wall up (CWU)approaches), to study the impact on hearing after treatment of labyrinthine fistula and finally to establish a management protocol to deal with labyrinthine fistula.

MATERIALS AND METHODS

Retrospective study done in a tertiary care hospital between February 2015 to January 2020. We included 20 patients having single labrynthine fistula confirmed intra operatively out of 149 patients who underwent mastoidectomy (CWU or CWD) for cholesteatoma, irrespective of age and sex. Cholesteatoma with cochlear promontory fistula with profound hearing loss, multiple fistula , cholesteatoma with intracranial complication was excluded from study.

Preoperative settings:

Records of age, gender, diseased ear side, patient symptoms, audiometric analysis by PTA (pure tone audiometry) to evaluate hearing loss , endoscopic ear evaluation by 4mm 0 degree telescope, HRCT temporal bone axial and coronal 0.5 mm cuts to look for labyrinthine fistula were collected.

Audiometric data were based on the American Academy of Otolaryngology Head and Neck surgery guidelines at 0.5, 1, 2, and 4 kHz. Pure tone averages were calculated on these 4 frequencies [16].

Intraoperative settings:

CWU or CWD mastoidectomy was done in all cases through post aural route. Labrynthine fistula identified and catergorized as per Dornhoffer and Milewski classification . Dornhoffer et al divided labrynthine fistula into 3 stages : stage I represents erosion of the bony labyrinth with intact endosteum; stage II is a true fistula with an opened perilymphatic space; and stage III is an opened perilymphatic space with concomitant involvement or destruction of the underlying membranous labyrinth. Stage II fistula is categorized further into stages IIa and IIb, but we did not subclassify stage II in this study.

Complete excision of cholesteatoma was done in all the cases along with repair of labrynthine fistula. Temporalis fascia followed by bone dust was used to repair labyrinthine fistula in all cases. All patients underwent excision of cholesteatoma and repair of labrynthine fistula in single stage.

Post operative settings:
Post operative audiometric evaluation was done after 4 to 6 weeks of surgery. Averaged air and bone conduction (BC) thresholds were compared before surgery and after surgery. Postoperative averaged BC thresholds were classified in three groups: unchanged, improved hearing, decreased hearing .

We analyzed association between patients age, sex, location of labyrinthine fistula, disease duration, surgical approaches, bone conduction threshold change and the fistula stage using chi-square ,Fisher's exact test.(p value of <0.05 was considered significant)

RESULTS
20 (13.42%)(11 males, 9 females) out of 149 patients of cholesteatoma presented with labyrinthine fistula. Age distribution ranged between 13–61 years (mean 31.65). As per Dornhoffer and Milewski classification, 15(75%) patients were identified as stage 1, 4(20%) patients as stage II, and 1(5 %) patient as stage III. Primary surgery was performed in 17 cases out of 20. Three patients underwent revision surgery. (Table no 1)

Table No. 1: Summary of clinical and radiological features, operative findings and hearing results.

Case no.	Sex	Age	Semi circular canal	Stage	Pre op BC(dB) threshold	Post op BC(dB) threshold	FN dehiscence intraopeartively	Pre op HRCT	Surgery
1	F	36	LSCC	1	10	10	–	–	CWDM*
2	F	13	LSCC	1	10	8.75	+	+	CWDM
3	M	19	LSCC	1	58.75	58.75	–	–	CWDM*
4	F	18	LSCC	2	55	90	+	+	CWDM
5	F	49	LSCC	1	25	25	+	+	CWUM
6	M	59	LSCC	1	25	25	+	+	CWDM
7	F	22	LSCC	3	0	25	+	+	CWUM
8	M	21	LSCC	1	10	10	+	+	CWUM
9	M	17	LSCC	1	60	60	+	+	CWDM*
10	M	14	LSCC	1	0	0	+	+	CWUM
11	F	46	LSCC	1	15	15	+	+	CWDM
12	M	58	LSCC	2	25	15	+	–	CWDM
13	M	37	LSCC	1	40	40	+	+	CWUM
14	F	50	LSCC	2	10	10	+	+	CWUM
15	F	19	LSCC	1	40	40	+	+	CWUM
16	M	61	LSCC	1	60	60	+	+	CWDM
17	M	32	LSCC	1	10	55	+	+	CWDM
18	M	18	LSCC	2	20	15	+	–	CWDM
19	F	21	LSCC	1	27.5	2.5	+	+	CWUM
20	M	23	PSCC	1	15	15	–	–	CWDM

LSCC-lateral semicircular canal, PSCC-posterior semicircular canal ,BC-bone conduction
CWUM-canal wall up mastoidectomy, CWDM – canal wall down mastoidectomy
*-revision surgery

1. Clinical assessment-
Preopeartively
Otorrhoea and hearing loss was seen in all (100 %). Dizziness was seen in 13 patients (65%), tinnitus in 3 patients (15%), headache and vomiting in 3 patients (15%).Duration of symptoms was between 6 to 40 yrs (mean 18.18 yrs). A positive fistula sign was present in only 2 patients (10%). (Table no 1)

Postoperatively:
Follow up done for 6 months to 5 years(mean 2.1 years). 40% patients complaints of dizziness in immediate post operative period, which was resolved within 2 weeks by labrynthine sedatives. No postoperative facial palsy or intracranial complications occurred.

2. Audiological assessment-Pure tone audiometry(PTA)
Preoperative:
Preoperative PTA showed conductive hearing loss in 10 patients (50 %) and mixed hearing loss in 10 patients (50%).Pre operative averaged AC threshold was 64.625 dB and BC threshold was 25.81 dB .

Postoperative:
Postoperative averaged AC threshold was 53.5 dB and BC threshold was 29 dB .

Preoperative and postoperative BC thresholds were compared. 13(65%) patients had stable hearing, 4(20%) patients showed improvement and 3(15%) patients cases showed worsening. There was statistically significant relationship between labyrinthine fistula and hearing outcome (P value = 0.036).
In stage I labyrinthine fistula (n-15)- 14 (93.33%) patients showed no change or improvement and 1(6.67%) showed worsening.
In stage II labyrinthine fistula (n-4) - 3(75%) %)

patients showed no change or improvement and 1(25%) showed worsening.

In stage III labyrinthine fistula(n-1) - 1(100%) %)patients showed worsening of hearing.(Table no 2) so we observed that as the stage of fistula increased ,there are more chances of worsening of post operative cochlear functions.

Table No. 2 : Correlation of labyrinthine fistula grade with fistula sign, post operative BC change, pre-operative HRCT prediction and surgical technique.

FISTULA GARDE	Type of surgery		HRCT fistula Detection		Post operative BC change			Fistula sign	
	CWUM	CWDM	Yes	No	No change	better	worst	Yes	No
1st	6	9	12	3	12	2	1	1	14
2nd	1	3	2	2	1	2	1	1	3
3rd	1	0	1	0	0	0	1	0	1
Total	8(40%)	12(60%)	15(75%)	5(25%)	13(65%)	4(20%)	3(15%)	2(10%)	18(90%)

Hearing outcomes are not affected by surgical technique.

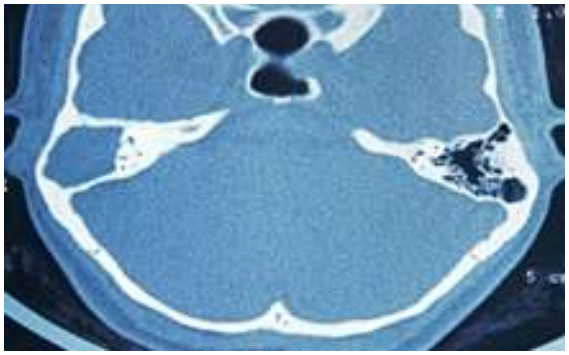
Table No 3 : Surgical technique versus post operative sensorineural hearing change.

Surgery type	Post operative Bone conduction hearing			
	worse	better	No change	Total
CWUM	2	3	7	12
CWDM	1	1	6	8
Total	3(15%)	4(20%)	13(65%)	20

3. Preoperative HRCT temporal bone prediction :

Labyrinthine fistula was suspected in 15 cases(75%).

(a)



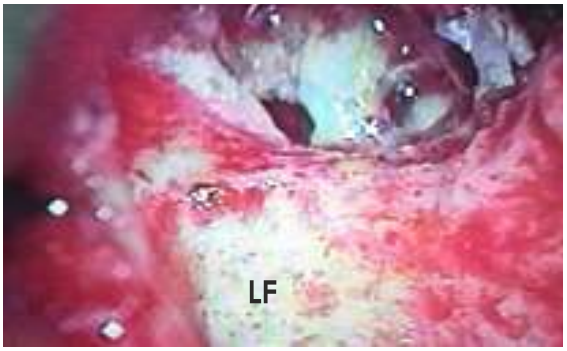
(b)

Fig 1- High resolution computed tomography scan temporal bone coronal (a) and axial (b) view showing right lateral semicircular canal fistula(white arrow).

There is no correlation between preoperative CT prediction and fistula grade .(Table no 2)

4. Intraoperative findings:

Using the Dornhofferand Milewski classification, 75% were classified as stage 1, 20 % as stage II,



and 5% as stage III .facial nerve was dehiscent in 17 cases(85%). 19 out of 20 involving LSCC (95%)and 1 involving PSCC(5%).(Table no 1)

Fig 2- Intra operative picture right canal wall down mastoidectomy showing grade I labyrinthine fistula over lateral semicircular canal.
LF-lateral semicircular canal fistula.

5. Surgical procedure (canal wall up or canal wall down)
CWU mastoidectomy was performed in 8 (40 %) patients whereas CWD mastoidectomy was done in 12(60%) patients, 3 of them were revision surgery (Table no 1). The choice of surgical technique was determined by status of posterior canal wall (eroded or intact), extension of disease, and size of mastoid and prior ear surgery. The choice of procedure was independent of the presence and grade of fistula. (Table no 2)

DISCUSSION
Labyrinthine fistula is one of the common complications of cholesteatoma. Incidence of labyrinthine fistula in literature varies between 3 to 16% with a average of 8% [17,18] . In our study, the incidence of labrynthine fistula is 13.42%. In 5 years, 20 out of 149 patients who were operated for cholesteatoma presented with labrynthine fistula. LSCC fistula was seen in 19 patients (95%) and PSCC in 1 (5%).LSCC is the most common location for labyrinthine fistula [2,6,17,19] due to preferred anatomical location of cholesteatoma in concordance with our study.

The average age in our study was 33.65 years, ranging between 13 to 61 years, which is similar to that reported in the literature [18-20] .Preoperative symptoms like otorrhoea and hearing loss which occurred in 100 % of the cases are non specific for labrynthine fistula and could be seen in chronic otitis media without labrynthine fistula.

Clinical signs and symptoms for labyrinthine fistula in cholesteatoma are positive fistula test and vertigo. Only 10 % cases in our study found positive for fistula test, which was low as compared to a systemic review study by Copeland and Buchman et al [2] where they found positive fistula sign in 50% patients . Discrepancy in results may be due to presence of cholesteatoma causing interruption of transmission of pressure from the external ear canal to the site of fistula. vertigo was present in 65% patients in our study compared to 45 % as seen in study by Meyer et al 18 and 100% for Hakuba et al [21] . we observed that positivity of the fistula test was not in correlation with the fistula grade (Table no 2). Vertigo and positive fistula test are poor indicators of labyrinthine fistula, as seen in existing literature [2,8,18,20] . Tinnitus was present in 15% patients in our study similar to seen in study by Meyer et al [8] (16%) . In our study there was no case with pre operative facial palsy with labrynthine fistula but some studies reported upto 10% of labrynthine fistula associated with pre operative facial nerve weakness [1,8,22].

HRCT temporal bone was done in all case of cholesteatoma to see anatomy, extent of disease , rule out complication and to tailor our surgical approach. The sensitivity of the HRCT temporal to detect labyrinthine fistula is 75 %(Table no 1) in our study which is slightly lowered compared to current literature showing 85 to 100% sensitivity [8,18,19]. This could be due to poor quality of HRCT temporal bone film cuts done at peripheral diagnostic center and long duration between imaging and date of surgery. For better planning and results, HRCT temporal bone 0.5 mm axial and coronal cuts should be done within a period of one month from date of surgery.

In our study there is no correlation between fistula stage introperatively and pre operative HRCT scan detection sensitivity (Table no 2) .ikeda et al [8] found paradox results which showed preoperative HRCT detection sensitivity 70% in stage I fistula,94% in stage II fistula and 100% in stage III fistula. Stephenson et al[19]showed 100% sensitivity and specificity in fistula detection irrespective of fistula stage.

Stephenson et al [19] also demostrated identification of membranous fistula versus bony fistula with 66% sensitivity and 71 % specificity respectively on preoperative HRCT temporal bone.

The management of labyrinthine fistula is controversial. Whether to do complete or partial removal of cholesteatoma matrix over fistula site/ do it in single stage or two stage/ approach of surgery (CWU Mastoidectomy vs CWD Mastoidectomy)?

Ritter and Freeman left matrix over fistula to avoid opening of labyrinth and preserve hearing [10,11] . Plava et al[23] reported a case of dead ear after 3 years of matrix preserving surgery due to suppurative labyrinthitis . Some authors left cholesteatoma matrix over fistula site during primary surgery and removed it during the second stage after 6 months [7,24,25] to obtain better functional results with reduced risks of hearing loss . Sanna et al [7] presented concept of “reverse metaplasia” i.e mucosalization of cholesteatoma matrix in 69% of post operative cases during reexploration. This could be due to restoration of normal ventilation of middle ear following disease removal in primary surgery. In a systemic review analysis Lim et al [26] found that rate of hearing preservation were similar in both staged and unstaged procedure.

Recent literature favor total eradication of cholesteatoma in a one-stage surgery [8,18,19] because complete cholesteatoma matrix removal results in reduction of bone resorption and reduced risk of suppurative labyrinthitis, [2,21] but it has potential to cause postoperative sensorineural hearing loss, but according to Copeland and Buchman et al [2] and Lim et al [26] both techniques (total removal or preservation of matrix over fistula) are alike in preserving hearing.

In our study, we performed single stage surgery in all cases with complete removal of cholesteatoma matrix and immediate repair using temporalis fascia followed by bone dust at the end of procedure irrespective of fistula grading.

RLF

Fig 3- Right ear canal wall down mastoidectomy with titanium TORP ossiculoplasty and lateral semicircular canal fistula repaired with bone dust and temporalis fascia. RLF-repaired lateral semicircular canal fistula, TORP-total ossicular replacement prosthesis

Freeman, Hakuba and Abramson et al [11,21,27] strongly believed that if labyrinthine fistula is present, radical or modified radical mastoidectomy is the preferred option. Sheey et al [17] advocated classical modified radical mastoidectomy if labyrinthine fistula was suspected in only hearing ear. Sanna et al [12] advocated canal wall down surgery in case of only hearing ear with multiple fistula or revision surgery . Sanna et al [12] showed that hearing outcome are not affected by surgical technique which is concomitant to our study. (Table no 3).

Most studies shows that choice of the technique(CWUM vs CWDM) adopted was dependent on the extent of disease, mastoid size, the history of prior ear surgery but not on the presence of labyrinthine fistula and its grade [12,18,19] which is again in concomitant to our study.

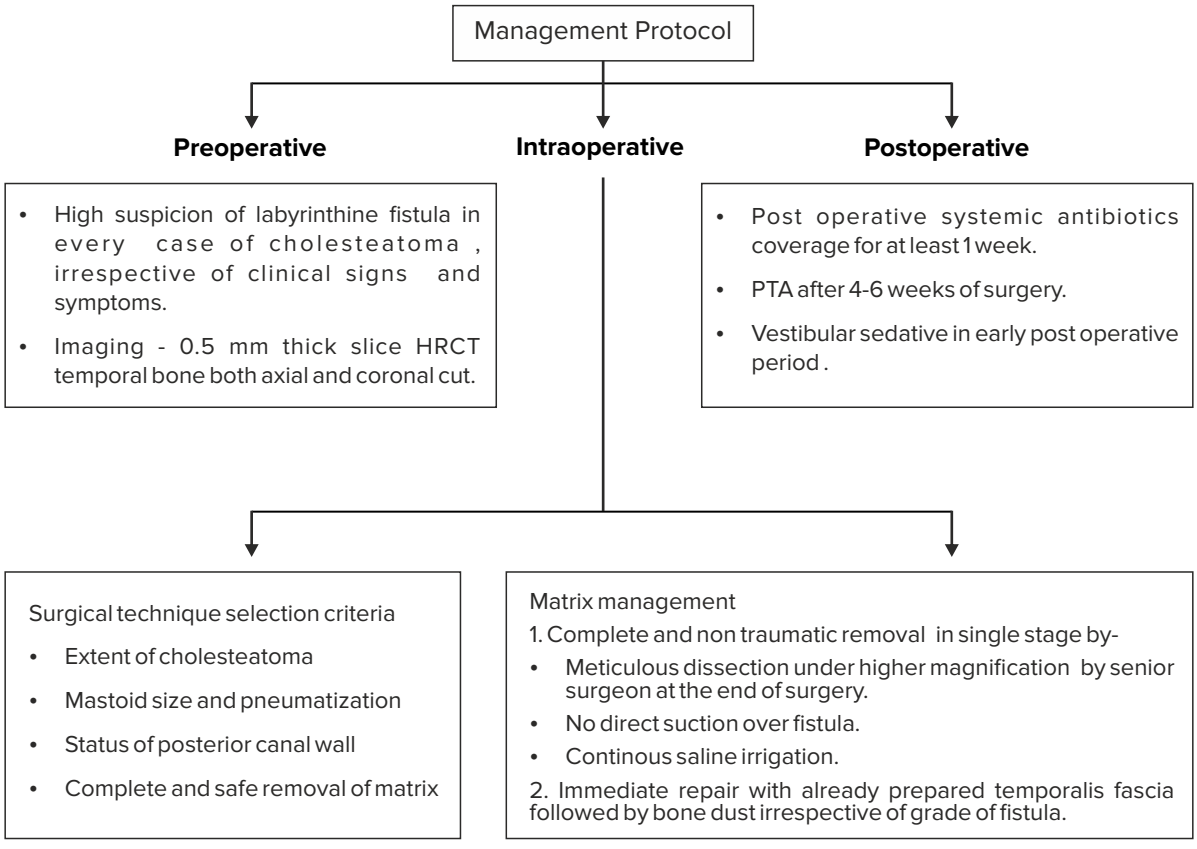
We advocate possible preferred technique to be canal wall up mastoidectomy, to maintain natural anatomy, avoid cavity related problem and minimize the need for long term care.

In our study post operatively hearing improvement was seen in 20% patients, hearing was preserved in 65 % cases and in 15% cases there was deterioration in sensorineural hearing

which was similar to existing literature [8, 18, 20]. Intraoperative labrynthine fistula staging correlated with the outcome of SNHL (sensorineural hearing loss) postoperatively. In stage I labrynthine fistula (n-15) 14(93.33%) patients showed no change or improvement in hearing status postoperatively and 1(6.67%) patient showed detoriation in hearing status postopeartively. In stage II labrynthine fistula (n-4) 3(75%) patients showed no change or improvement in hearing status postoperatively and 1(25%) patient showed detoriation of hearing status postoperatively. In stage III labrynthine fistula (n-1) 100% patients showed detoriation of hearing status postoperatively. So we would like to conclude that as the stage of fistula increases ,there are more chances of detoriation of post operative cochlear functions. By classifying labrynthine fistula stage intraopeartively we can asses post operative hearing status. Some series do not show any correlation between labrynthine fistula grade and post operative sensorineural hearing outcome [8,18,20].

CONCLUSION

Labyrinthine fistula is a common complication of extended cholesteatoma with most common site being LSCC. Since preoperatively sign and symptoms of labyrinthine fistula are not diagnostic , HRCT temporal bone is of utmost importance in making its diagnosis. The choice of canal wall up mastoidectomy or canal wall down mastoidectomy is determined by the characteristics of cholesteatoma, but is independent of the presence of labrynthine fistula and its grade. Complete and meticulous removal of cholesteatoma matrix over the labrynthine fistula, followed by its repair in a single-staged procedure is a safe and effective procedure. Intraopeartive staging of fistula can be used to prognosticate post operative hearing outcome of patient. We have tried to give a management protocol for management of labrynthine fistula with cholesteatoma for good outcomes.



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