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Managing cholesteatoma in labyrinth fistula

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April 05, 2024

Abstract

Objectives: To evaluate the management of cholesteatomas with labyrinthine fistulas (LF), as well as the clinical characteristics and postoperative hearing outcomes in a tertiary general hospital from 2011 to 2018. Design: Retrospective observational cross-sectional study using hospital electronic medical records. Setting: Mastoidectomies done for clinically suspected cholesteatomas. Participants: All patients who underwent primary middle ear mastoidectomy for cholesteatomas. Main outcome measures: Demographic data: age, gender, race, presenting complaints. Preoperative HRCT temporal bone and intraoperative findings. Change in hearing levels preoperatively and postoperatively. Results: 14 of 90 ears (15%) with middle ear cholesteatomas were complicated by LF. HRCT observed 93% sensitivity and 95% specificity in the identification of LFs. Intraoperative findings include stapes erosion in 79% ears ($p<0.001$), malleus erosion in 79% ears, incus erosion in 93% ears, dehiscence of tegmen tympani in 64% of ears ($p=0.016$) and tympanic facial canal dehiscence in 45% of ears. Matrix was removed completely in 85% (n=12) and matrix was left behind in 13% (n=2). 21.5% (n=3) had preoperative dead ears. Postoperative hearing results had a mean follow-up time of 2.1 years (1.5, 0.14-4.84). Matrix removal group (n=9) had hearing levels with 78% no change, 11.1% improvement and 11.1% decrease in hearing levels, while matrix preservation group (n=2) had decreased hearing levels. Conclusions: Preservation of hearing in labyrinthine fistulas is possible with cautious matrix removal and immediate repair of the fistula. Longer follow-up of hearing of LF with matrix preservation may show poorer hearing outcomes.

Key points:

1. We report labyrinthine fistulas in 15% of middle ear cholesteatomas that underwent mastoidectomies.
2. We observe that preoperative HCRT had 93% sensitivity and 95% specificity in the identification of LFs.
3. Intraoperative findings include erosion of 79% stapes, 93% incus, 79% malleus and dehiscence in 29% tegmen tympani and 64% tympanic facial canal.
4. Significantly higher incidence of stapes erosion and tegmen dehiscence observed in cholesteatomas with labyrinthine fistulas compared to without.
5. 6 out of 8 patients with complete matrix removal and fistula repair did not have a change in hearing. Hence it is possible to preserve hearing with complete matrix removal.

INTRODUCTION

Labyrinthine fistulas (LFs) are formed from the erosion of the bony labyrinth and occur as a complication in 2.7-21% (1, 2) of cholesteatomas.

Early identification of LFs allows better surgical planning to reduce potential vestibulocochlear sequelae. While intraoperative diagnosis of LFs occurs with clear visualisation of semicircular canal (SCC) fistula, high-resolution CT (HRCT) temporal bone scans are currently mainstay investigation preoperatively, with sensitivity of 53.8-100% (3, 4) and specificity of 90-100% (3, 5).

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Surgical management of LFs varies. Matrix may be left over the fistula and the mastoid cavity exteriorised to prevent complications such as sensorineural hearing loss and vertigo. Alternatively, matrix may be completely removed and the fistula repaired.

Objectives

We report our local experience in the management of LFs in middle ear cholesteatomas, over an 8-year period, at a major tertiary hospital. We aim to discuss the clinical characteristics, management and hearing outcomes.

Ethical considerations

Ethics approval was obtained from the Singhealth Centralized Institutional Review Board before commencement of the study (2019/2791).

METHODS

Reporting guideline: STROBE

A retrospective review was done for mastoidectomies performed between July 2011 and July 2018 by 2 senior surgeons were identified via the electronic surgical logbook. Patients with primary cholesteatomas were included, excluding congenital cholesteatomas and incomplete data. Data extracted comprised preoperative demographics, high-resolution CT (HRCT) 0.5mm thickness temporal bone scan results, intraoperative findings and hearing results.

Diagnostic accuracy and correlations of non-contrast HRCT scans with intraoperative findings were analysed for 1) erosion of structures: ossicular chain, tegmen tympani, tympanic facial canal and semicircular canal, 2) location of squames and 3) ossicular chain encasement. The technique of matrix preservation or removal was dependent on surgeon choice and intraoperative findings, such as presence of intact endosteum. Material to repair LFs was also noted. Average air conduction (AC), bone conduction (BC) and air-bone gap (ABG) pure tone threshold (PTA) were performed at 500Hz, 1kHz and 2kHz for all patients pre- and post-operatively. A change of BC PTA of 10dB or more is considered clinically significant.

Statistical analysis was conducted using chi square test using IBM SPSS statistic package, version 25. Cholesteatomas were categorized into "LF" and "No LF" groups. Continuous variables such as age and hearing levels were reported as mean (standard deviation, range). Comparisons of categorical variables were performed using the Pearson chi-squared test.

RESULTS

Participants

A total of 90 ears underwent mastoidectomies for primary middle ear cholesteatomas during the 8-year period. 5% (5/90) had bilateral cholesteatomas. 15% (14/90) of ears in 13 patients were found to have LFs intraoperatively, with an equal number of left and right sided LFs. 54% (7/13) of patients were male and 69% (9/13) were Chinese. Mean age of diagnosis was 52 years (19.6, 8-6).

Descriptive data

Presenting complaints of ears with LFs were 80% (12/14) hearing loss, 79% (11/14) otorrhea, 29% (4/14) giddiness, 7% (1/14) tinnitus, 7% (1/14) facial palsy and 7% (1/14) otalgia. 65% (9/14) ears had both hearing loss and otalgia.

HRCT accurately identified 13 of the 14 LFs diagnosed intraoperatively, with a false positive of 4 ears and false negative of 1 ear. This reflected a sensitivity of 93% (13/14) and specificity of 95% (72/76) in the identification of LFs through semicircular canal dehiscence. All fistulas were located in lateral SCC and 7% (1/14) in superior SCC.

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In LF group, intraoperative dehiscence was observed – 29% (4/14) in facial canal, 64% (9/14) tegmen tympani. Ossicular erosion occurred in 79% (11/14) stapes, 79% (11/14) incus and 93% (13/14) malleus. Intraoperative squames were found, 93% (13/14) attic, 93% (13/14) middle ear and 71% (10/14) mastoid. Ossicular chain encasement occurred in 71% stapes (10/14), 93% (13/14) incus and 79% malleus (11/14). Compared to non-LF group, LF group noted a significantly higher incidence of stapes erosion ($p<0.001$), dehiscence in tegmen tympani ($p=0.016$) and SCC ($p<0.001$) [Table 1].

Outcome data

All cholesteatomas with LFs underwent canal wall down mastoidectomy and primary mastoid obliteration with local tissue flaps (middle temporal artery or inferior musculoperiosteal flaps). Middle ear reconstruction utilized cartilage placed onto remnant stapes suprastructure if present or stapes footplate if absent stapes. Surgeon 1 employed complete matrix removal in 86% (12/14) of LFs, by leaving the matrix and cholesteatoma over the SCC till the last stage of operation, before delicately removing the matrix. This was followed by immediate bone-wax sealing of fistula in 50% (6/12) and temporalis fascia in 8% (1/12). As the endosteum was intact in the remaining 5 cases, these cases were not sealed. Surgeon 2 performed matrix preservation in the remaining 14% (2/14) SCC fistulas. Revision mastoidectomies for recurrent cholesteatoma were required in 14% (2/14). These 2 cases had complete matrix removal, whereby time to recurrence was 5 years and 6 years respectively.

Postoperative hearing PTAs were taken at a mean of 2.1 years (1.5, 0.14-4.84). Mean preoperative and postoperative BC thresholds were 38.4dB and 45.3dB while AC thresholds were 73.6dB and 79.4dB respectively. 21.5% (3/14) ears had preoperative severe to profound sensorineural hearing loss. The remaining 11 ears were evaluated on their pre and postoperative hearing [Table 2]. Of the 11 ears, 9 ears had their matrix completely removed and LF repaired, and 2 ears had matrix preservation with a modified radical mastoidectomy performed as the fistula was large.

Matrix removal group had hearing levels with no change in 78% (7/9), improvement in 11.1% (1/12) and decreased in 11.1% (1/12). Mean preoperative and postoperative BC thresholds were 33.1dB (17.8, 13-68) and 38.8dB (19.3, 13-68) respectively. Postoperative hearing thresholds done within 120 days showed worsening hearing in 22% (2/9) ears, that subsequently had hearing restoration achieved at the last follow-up of 3.2 years and 1.6 years respectively.

The 2 LFs with matrix preservation had decreased hearing levels, after a follow-up time of 3.2 years and 2.7 years respectively. Increase in BC thresholds preoperatively to postoperatively were 30dB to 65dB over 2.7 years for patient 1 and 40dB to 72dB over 3.2 years for the other.

DISCUSSION

Key results

Our study reported LFs in 15% (14/90) of middle ear cholesteatomas. The evaluation of cholesteatomas pre-operatively with HRCT yielded a high sensitivity (92.9%) and specificity (94.7%) for LFs. Significantly higher incidence of tegmen tympani dehiscence, stapes erosion were seen in ears with LF. Matrix management method in our centre is based on surgeon preference, considering the approximate fistula size based on preoperative imaging and initial hearing. The two LFs with matrix preservation were done due to Surgeon 2's suspicion of larger fistulas and both patients hearing thresholds. Significantly better postoperative hearing outcomes was observed in complete matrix removal than matrix preservation in our study.

Limitations

Analysis was limited to a small sample size in this case series and variability of timing to postoperative hearing outcomes could have affected the conclusiveness of results. In addition, only 2 patients had matrix preservation and more patients are needed to make a better comparison. Surgeon's preference of matrix management was also subjected to selection bias.

Interpretation

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Labyrinthine fistula (LF) is a known complication of cholesteatomas, most common in the lateral SCC, but also affects superior SCC in 6% and posterior SCC in 2% of LFs (6). Clinical suspicion of LFs is limited by non-specific symptoms – mostly sensorineural or mixed hearing loss, accompanied by vertigo, otorrhea and otalgia also present in chronic otitis media. Classic clinical signs such as fistula sign may only have a sensitivity of 21.7% to 60% (7), false negative rate of up to 54% (8). Also, there is lack of uniformity in performing the test.

While some centres have used the Dornhoffer and Milewski classification to stage fistulas based on bony defects and size of fistulas to determine surgical method, the fistula size, size and grade have been reported to not correlate with surgical outcomes (9). Surgeon 1's technique of matrix removal was applied to all LFs, irrespective of size of fistula. Sealing of fistulas included bone wax and temporalis fascia, consistent with literature also reporting the use of bone wax, temporalis fascia, bone dust, tissucol, fibrin glue, Spongiostan and autogenous tissues (6, 9).

Postoperative hearing deterioration rate has been reported in 2.8-26.9% (9), possibly attributed to the removal of cholesteatomas and granulation tissue previously acting as conduits in conducting sound into ear. Postoperative deterioration in sensorineural hearing loss occurs in 23% to 50% that underwent complete matrix removal and 1 to 44% for matrix left in-situ (9). Unlike the review by Lim et al (9), our patients with matrix preservation had poorer hearing outcome, perhaps due to longer follow-up time. Matrix removal, however, had hearing preservation in 78% although 2 ears had initial worsening within 120 days postoperatively. This is in contrary to greater deterioration of long-term hearing outcomes found by Katsura et al (10), despite initial improvements within the first year, postulated to the involvement of third window during the initial recovery stage. Postoperative persistent hearing loss could be a result of inflammatory effects of the remnant cholesteatoma matrix.

Generalisability

We present our experience in matrix management in LFs, based on surgical preference and labyrinthine manipulation, with findings consistent with literature. Long-term hearing results for fistulas with matrix preservation compared with removal could be explored further in future studies.

(1493 words)

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