

OTOLOGY

Treatment of cholesteatoma with intact ossicular chain: anatomic and functional results

Il trattamento chirurgico del colesteatoma a catena ossicolare integra: risultati anatomici e funzionali

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SUMMARY

In case of cholesteatoma with intact ossicular chain, the primary aims of surgery are complete removal of the cholesteatoma matrix and reconstruction of a dry and safe middle ear; if possible, ossicular chain continuity and therefore the preoperative hearing must be preserved. The aim of this retrospective study is to present the experience of the U.O.C. Otorinolaringoiatria Universitaria of University of Bari "Aldo Moro" in treatment of intact ossicular chain cholesteatoma with Bondy modified radical mastoidectomy (BMRM) and canal wall up tympanoplasty (CWUT). The study group was composed of 65 subjects affected by cholesteatoma with intact ossicular chain. Mean age was 40.7 years (range 6-79), with 42 males and 23 females. 30 patients were treated by a BMRM and 35 by CWUT, in 22 cases without mastoidectomy and in 13 cases with mastoidectomy. Mean follow-up was 24.25 months. In the BMRM group, no cases of residual cholesteatoma located in the middle ear space were detected; at follow-up, 1 patient developed a retraction pocket (3.33%), 1 patient showed a small epidermal cysts of the tympanic membrane (3.33%) and 3 patients (10%) experienced otorrhea. In CWUT, residual cholesteatoma was detected in 2 cases (5.7%); at follow-up, 3 patients presented recurrent cholesteatoma (8.57%; 2, 6 and 8 years after surgery), 3 cases a retraction pocket (8.57%) and one case otorrhea (2.86%). Statistical analysis showed a significant higher number of residual cholesteatoma in CWUT ($p < 0.005$) and differences in terms of long-term complications. No significant changes in hearing occurred post-operatively or at 1 year follow-up in either group. The current trend in our centre is to perform BMRM when indicated and CWUT preferably without mastoidectomy in case of mesotympanic cholesteatoma with normal OC.

KEY WORDS: Cholesteatoma • Intact chain • Modified bondy mastoidectomy • Canal wall up tympanoplasty

RIASSUNTO

L'obiettivo primario del trattamento chirurgico del colesteatoma a catena integra è la completa eradicazione della patologia, la creazione di un orecchio asciutto e se possibile il mantenimento della funzione uditiva pre-operatoria. L'obiettivo di questo studio retrospettivo è di presentare i risultati ottenuti nel trattamento del colesteatoma a catena integra trattati mediante Radicale Modificata secondo Bondy e timpanoplastica chiusa presso U.O.C. Otorinolaringoiatria Universitaria Dell'Università di Bari "Aldo Moro". Il gruppo di studio è composto da 65 pazienti affetti da colesteatoma a catena integra. L'età media era di 40 anni (range 6-79 anni), 42 maschi e 23 femmine. 30 pazienti sono stati sottoposti a Radicale Modificata secondo Bondy e 35 a timpanoplastica chiusa (22 senza e 13 con mastoidectomia). Il follow-up medio è stato di 24,25 mesi. Nei pazienti trattati con radicale modificata secondo Bondy in nessun caso si è verificato un colesteatoma residuo, due pazienti (6,66%) hanno presentato complicanze precoci (3,33% vertigine, 3,33% acufeni) e 5 pazienti (16,67%) hanno manifestato complicanze tardive (3,33% tasca di retrazione, 3,33% cisti epidermica della MT, 10% otorrea). Nel gruppo trattato con timpanoplastica chiusa, 2 pazienti hanno presentato colesteatoma residuo al secondo tempo (5,7%) e 7 pazienti (20%) hanno presentato complicanze tardive (8,75% colesteatoma ricorrente, 8,75% tasca di retrazione, 2,86% otorrea). L'analisi statistica ha dimostrato un numero significativamente più alto di colesteatoma residuo in pazienti sottoposti a timpanoplastica chiusa ($p < 0,005$) e nessuna differenza nelle complicanze a lungo termine. La capacità uditiva in entrambi i gruppi non si è modificata nel post-operatorio e ad un anno di follow-up. Entrambe le tecniche si sono dimostrate efficaci da un punto di vista anatomico e funzionale nel trattamento del colesteatoma a catena integra. La tecnica di Bondy trova indicazione in caso di colesteatoma epi-timpanico laterale alla catena ossicolare, mentre la timpanoplastica chiusa è la nostra tecnica di scelta in caso di colesteatoma mesotimpanico.

PAROLE CHIAVE: Colesteatoma • Catena integra • Bondy • Timpanoplastica

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Introduction

The erosion of the ossicular chain (OC) in the course of cholesteatoma is a common condition found in over 85% of cases ¹. In recent decades, cases of advanced cholesteatoma have been significantly reduced because of the use of antibiotics and improved diagnostics; this has also led to an increased number of patients with intact OC and good hearing at presentation ².

In case of cholesteatoma with intact OC, the primary aims of surgery are complete removal of the cholesteatoma matrix and reconstruction of a dry and safe middle ear; if possible, OC continuity and therefore the preoperative hearing must be preserved.

All these aims can be fulfilled using two surgical techniques, namely Bondy modified radical mastoidectomy (BMRM) ³ and canal wall up tympanoplasty (CWUT) with or without mastoidectomy ⁴. BMRM is an extremely effective operation when practiced on carefully selected patients ⁵. This technique is indicated in case of patients affected by epitympanic cholesteatoma spreading laterally to an intact OC, normal pars tensa and good preoperative hearing (air bone gap, ABG, < 25 dB) ^{3,6}. CWUT with or without mastoidectomy is a conservative technique that can be performed in all cases of cholesteatoma with intact OC. In case of cholesteatomas involving only the middle ear, CWUT can be performed through a postauricular microscopic approach ⁷ or an exclusive endoscopic transcanal approach ⁸.

The aim of this retrospective study is to present the experience of the U.O.C. Otorinolaringoiatria Universitaria of University of Bari "Aldo Moro" in the treatment of intact OC cholesteatoma with BMRM and CWUT.

Materials and methods

Between November 2000 and December 2014, 673 subjects affected by a previously untreated middle ear and mastoid cholesteatoma were operated on and among these subjects all patients in whom the OC was kept intact during surgery were selected. The study group was therefore composed of 65 consecutive subjects. Mean age was 40.7 years (range 6-79), with 42 were males and 23 females. 30 patients were treated by a BMRM and 35 by CWUT, in 22 cases without mastoidectomy and in 13 cases with mastoidectomy. CWUT was staged in 8 cases (23%). The location and the extension of the cholesteatoma were evaluated as well as the anatomical and functional postoperative results and complications. In particular, the location of the cholesteatoma was recorded in the following anatomical sites: epitympanum, antrum, mastoid, mesotympanum and protympanum.

The guidelines of the Committee on Hearing and Equilib-

rium of the American Academy of Otolaryngology Head and Neck Surgery ⁹ were followed and the pure-tone average (PTA) was calculated as the mean of 0.5, 1, 2 and 4 kHz thresholds. Air-Bone Gaps (ABG) were calculated from air conduction (AC) and bone conduction (BC) thresholds determined in each study. Mean follow-up was 24.25 months (range 6-210); in BMRM, average follow-up was 25.83 months and in CWUT it was 22.8 months. All patients signed an informed consent and the work was performed in accordance with the principles of the 1983 Declaration of Helsinki.

Surgical technique

BMRM was performed as described by Sanna et al. ¹⁰ with some technical differences. Conchal cartilage was not removed and the meatoplasty was obtained through a full thickness incision along the superior wall of the external auditory canal with posterior-inferior displacement of the flap obtained. The sinodural angle was obliterated with bone paté and covered with connective tissue, as previously described ¹¹. Particular care was taken to avoid contact between bone paté and the short process of the incus in order to prevent fixation.

CWUT was performed through a retroauricular incision in all cases under microscopic magnification. In case of cholesteatoma located only in the middle ear spaces, the surgery was performed through a postauricular transcanal approach. When needed, atticotomy was performed and the scutum reconstructed with tragal cartilage. In case of cholesteatoma involving the posterior epitympanum and spreading towards the antrum, a CWUT with posterior tympanotomy was performed. Scutum defect was reconstructed with bone paté. The tympanic membrane was always reconstructed with temporalis fascia.

Statistical analysis

Anatomical results are presented as percentiles and comparison between the two groups was done by Chi-square test. Statistical analysis of functional results was performed by ANOVA test for comparison between the two groups and by paired samples t-test for comparison between preoperative and postoperative values for each group of patients. The statistical software STATA_MP11 was used. Significance was set for $p < 0.05$.

Results

Anatomical results

In Table I the anatomical subsites involved by cholesteatoma are reported. Epitympanum (100% vs. 42.86% of the CWUT group; $X^2 = 24.76$; p -value = 0.000001) and antrum

Table I. Anatomical sites involved by cholesteatoma are reported. BMRM; Bondy modified radical mastoidectomy. CWUT; canal wall up tympanoplasty.

| Sites | BMRM | CWUT | p-value |
|--------------|--------|--------|-----------|
| Antrum | 66.67% | 17.14% | p < 0.001 |
| Epitympanum | 100% | 42.86% | p < 0.001 |
| Mesotympanum | 0 | 74.28% | p < 0.001 |
| Mastoid | 13.33% | 5.71% | p > 0.01 |
| Protympanum | 0 | 0 | 1 |

(66.67% vs. 17.14% of the CWUT group; $X^2 = 16.51$; p-value = 0.00048) were most commonly involved in case of BMRM compared to CWUT (p < 0.001). Otherwise, the involvement of the posterior mesotympanum was significantly higher in case of CWUT (p < 0.001).

In Table II the number of anatomical sites involved are reported. In both groups, the cholesteatoma involved one or two sites in a large proportion of cases. In case of CWUT, cholesteatoma involved only one site in significantly more cases (p-value = 0.002).

Dehiscence of the tympanic portion of the facial nerve was observed in 2 patients treated with BMRM (6.67%). A lateral semicircular canal fistula was detected in 2 cases (6.67%) of the BMRM group, while in the CWUT group a fistula of the stapes footplate was found in 1 case. Partial erosion of the long process of the incus was observed in 8 patients submitted to BMRM (26.67%) and 3 patients to CWUT (8.57%). No significant differences were found in terms of intra-operative findings.

Anatomical complications were grouped into three categories: early post-operative, at second-look and at follow-up. In the early post-operative period, 1 patient treated with BMRM experienced dizziness and one reported post-operative tinnitus; analogous symptoms were reported by 2 patients treated with CWUT (5.7%). Second-look procedures were performed in 8 cases operated by CWUT where complete removal of the disease was doubtful. In two cases (5.7% of total and 25% of second look procedures) a residual cholesteatoma was found, in one case adherent to the stapes superstructure and in one case to the long process of the incus. In BMRM, no cases of residual cholesteatoma located in the middle ear space were detected at follow-up. Statistical analysis showed a significant difference (p 0.005) between

Table II. Number (and %) of anatomical sites involved in the two groups.

| | 1 site | 2 sites | 3 sites | 4 sites |
|-------|-------------|-------------|------------|-----------|
| BMRM | 10 (33.33%) | 16 (53.33%) | 4 (13.33%) | 0 |
| CWUT | 23 (65.71%) | 8 (22.86%) | 0 | 1 (2.86%) |
| TOTAL | 33 (50.77%) | 24 (36.92%) | 4 (6.15%) | 1 (1.54%) |

BMRM, Bondy modified radical mastoidectomy; CWUT, canal wall up tympanoplasty.

the two techniques. At follow-up in the BMRM group, 1 patient developed a mesotympanic retraction pocket (3.33%), 1 patient showed a small epidermal cysts of the tympanic membrane (3.33%) and 3 patients (10%) experienced otorrhoea. In patients treated with CWUT, long-term complications occurred in 20% of cases: 3 patients presented recurrent cholesteatoma (8.57%; 2, 6 and 8 years after surgery), 3 cases a retraction pocket (8.57%) and one case otorrhoea (2.86%). Statistical analysis did not show a significant difference between the two techniques in terms of long-term post-operative complications.

Functional results

In Table III the pre-operative, immediate post-operative and 1 year follow-up average AC, BC and ABG thresholds (\pm standard deviation) of the two groups are reported. Statistical analysis showed that, pre-operatively, patients submitted to CWUT presented significantly worse hearing compared to BMRM group in terms of AC (p = 0.008), BC (p = 0.04) and ABG (p = 0.02). No differences were encountered in the post-operative period. No significant changes in hearing occurred post-operatively and at 1 year follow-up in both groups.

One patient in the BMRM group presented a decline of bone conduction > 20 dB and one patient submitted to BMRM underwent a revision surgery because of a post-operative increase in the ABG at 6 months after surgery. During revision, bone dust was found at the level of the incudo-malleolar joint that impeded the normal vibration of the OC; after removal of the bone dust the ABG returned to 8 dB HL.

Discussion

The aim of this study was to analyse the results of our experience in the surgery of cholesteatoma with intact OC by BMRM or CWUT.

Table III. Postoperative hearing results at follow-up. PTA-AC; pure tone average air conduction at 0.5, 1, 2 and 4 kHz. PTA-BC; pure tone average bone conduction at 0.5, 1, 2 and 4 kHz.

| | | Preoperative | Postoperative | 1 year follow-up |
|-------|------|-------------------|-------------------|-------------------|
| PTAAC | BMRM | 27.72 \pm 11.46 | 32.66 \pm 13.77 | 23.18 \pm 12.41 |
| | CWUT | 41.2 \pm 20.58 | 42.55 \pm 22.19 | 35.19 \pm 22.33 |
| PTABC | BMRM | 15.92 \pm 9.14 | 18.8 \pm 10.29 | 15.91 \pm 9.92 |
| | CWUT | 23.75 \pm 16.07 | 23.02 \pm 13.13 | 22.21 \pm 16.83 |
| ABG | BMRM | 11.79 \pm 6.48 | 13.86 \pm 9.03 | 7.27 \pm 4.87 |
| | CWUT | 17.45 \pm 9.18 | 19.53 \pm 13.62 | 12.98 \pm 10.40 |

ABG, air bone gap; BMRM, Bondy modified radical mastoidectomy; CWUT, canal wall up tympanoplasty.

It is clear from this series that the two surgical techniques have different indications. BMRM has very strict indications, namely epitympanic cholesteatoma lateral to an intact OC. The involvement of the posterior mesotympanum was, in fact, never encountered in the present series, while involvement of the antrum and mastoid was not a contraindication of the technique as well as the partial erosion of the OC. CWUT with or without mastoidectomy was instead used in cases of limited cholesteatomas involving only one or two sites in 97.14% of cases, with the posterior mesotympanum involved in 74.28% and the epitympanic spaces in 42.86% of cases. Surgery was performed exclusively through a postauricular transcanal approach in 2/3 of cases and with mastoidectomy and posterior tympanotomy in the remaining.

It should be noted that the present series includes only patients where the OC was maintained intact during surgery; in fact, all subjects in whom the intact OC was not maintained during surgery were excluded. We usually try to preserve an intact OC only when the cholesteatoma sac does not involve the medial surface of the body of the incus and/or of the incudo-malleolar joint. In these cases, even with the use of angled endoscopes and angled instruments the likelihood to radically remove the cholesteatoma and preserve the OC has been reported to be around 15%⁹.

In case of patients with intact OC, the indications of the two techniques overlap only in case of limited cholesteatoma involving the epitympanum without medial extension, while the involvement of the posterior mesotympanum represents a contraindication to the use of BMRM. In case of a pure epitympanic cholesteatoma, both an atticotomy and a BMRM can be performed, although a higher risk of recurrence is possible with the former technique.

In the present series, patients treated with CWUT presented residual cholesteatoma in 5.37% of cases (25% of patients undergoing second look), tympanic membrane retraction pocket in 8.57% of cases and recurrent cholesteatoma in 8.57% of cases, while patients treated with BMRM presented no cases of residual disease medial to the tympanic membrane, one case (3.3%) of tympanic membrane retraction pocket and no cases of recurrent cholesteatoma.

Similar results are reported by other authors in patients with an intact OC. In a large series of 258 patients affected by epitympanic cholesteatoma and intact OC treated by BMRM with at least 5 years follow-up, Prasad et al.¹² reported the occurrence of residual cholesteatoma lateral to the tympanic membrane in 8.1% of patients, no cases of residual cholesteatoma medial to the tympanic membrane and 3.1% of retraction pocket and no recurrent cholesteatoma. Berrettini et al.⁵ in a series with at least 5 years of follow-up reported that 84.5% of patients were free of

complications. Marchioni et al.⁸ presented a series of 20 patients affected by epitympanic cholesteatoma with intact OC treated with exclusive endoscopic surgery. They reported a recurrence rate of 8.7%, and, as in the present series, they were able to preserve the OC only in cases of limited cholesteatoma not involving the medial attic⁸. Tarabichi¹³ in a series of 73 cases affected by limited attic cholesteatoma treated with endoscopic surgery reported OC preservation in 69.8% of cases, tympanic membrane retraction pocket in 38% of cases and recurrent disease in 6.8% of cases. Smouha and Javidfar¹⁴ in series of 39 patients affected by mesotympanic and epitympanic cholesteatoma with normal hearing and intact OC treated with microscopic CWUT reported OC preservation in 77% of cases and a recidivism rate of 26%, being 6% residual cholesteatoma and 20% recurrent cholesteatoma.

In terms of post-operative hearing in the present series, maintenance of the OC was associated with a not significant change in the post-operative ABG with either technique. Similar results were reported by Prasad et al.¹² in case of BMRM; no significant change in hearing was reported by Tarabichi¹³, while Smouha and Javidfar¹⁴ were able to preserve hearing in 78% of cases where the OC was maintained intact.

In case of epitympanic cholesteatomas, in our experience MRBM represents the technique of choice, since it gives the best results in terms of recidivisms as well as hearing. One of the problems that have been associated with "open cavities" such as BMRM is the poor quality of life reported by patients because of the cavity itself and a large meatoplasty. Our group has, however, recently demonstrated that with the obliteration of the sinodural angle and a "cosmetic meatoplasty", the reported quality of life in these patients is not worse than that reported by patients submitted to CWUT¹¹.

The real challenge for the surgeon is therefore treatment of patients affected by mesotympanic cholesteatomas with intact OC. In these cases, the creation of a cavity may represent an overtreatment considering the pure involvement of the middle ear spaces. In our opinion, in cholesteatomas with intact OC localised in the posterior mesotympanum with or without involvement of the epitympanum and antrum a CWUT is indicated. The main complications associated with this technique are residual and recurrent cholesteatoma. Residual cholesteatoma originates from epidermal cells that are left behind during surgery, while recurrent cholesteatoma derives from a newly formed retraction pocket or a perforation of the reconstructed tympanic membrane¹⁵. Two recent papers have evaluated disease recurrence after CWUT and canal wall down tympanoplasty (CWDT)^{16 17}; the

authors performed a systematic review in one case¹⁶ and a meta-analysis in the other¹⁷; in both papers, the relative risk of recurrent or residual disease was 2.87 after CWUT compared to CWDT. The higher risk of residual disease has been correlated with poorer visualisation of the hidden recesses of the middle ear in CWUT such as the tympanic sinus and medial wall of the attic²⁰. Use of endoscopes in middle ear surgery increase visualisation of middle ear spaces²¹ and has the potential to reduce the risk of residual disease^{8 13 18 19}. Recurrent disease is caused in most of the cases by retraction of the tympanic membrane and formation of a cholesteatoma sac. An entered mastoid behind an intact canal wall has been proposed as a precondition for recurrence, together with the presence of fibrous tissue and adhesions in mastoid and middle ear that impair normal mucosal function²²⁻²⁶. In the present series, two of the three patients who developed a recurrent cholesteatoma and one of the three patients who developed retraction pockets underwent mastoidectomy. The maintenance of the middle ear mucosa, reconstruction of any scutum defect and ventilation of middle ear spaces are the mainstay in prevention of recurrences¹⁰.

Conclusions

It is clear from the literature that there is no single procedure that can be used in all cases of cholesteatoma, and therefore the surgeon should be able to choose the right technique for the individual patient. The high percentage of complication-free cases, associated with excellent functional results, lead us to consider that both BMRM and CWUT are extremely effective when performed correctly and following the right indications. The advantages of the Bondy technique are good short- and long-term functional results and a low risk of residual and recurrent cholesteatoma. The downside is the risk of cochlear trauma, with a consequent deterioration of bone conduction and the limited indications. The current trend in our centre is to perform BMRM when indicated and CWUT preferably without mastoidectomy in case of mesotympanic cholesteatoma with normal OC. In these cases, the use of endoscopes can help the surgeon in dealing with cholesteatoma in hidden areas.

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