**Pediatric Otolaryngology**

Long-term surgical and functional outcomes of the intact canal wall technique for middle ear cholesteatoma in the paediatric population

In this paper, we report the postoperative outcomes in canal wall up procedures with second stage surgery in 40 children undergoing intervention for cholesteatoma of the middle ear. The residuals, recurrences and the hearing results were analysed. All 40 patients had a follow-up of at least five years. Of the 39 patients who underwent two staged surgery, 18 (46.1%) had a residual lesion that was identified and excised during the second surgery. Over a five year follow-up period, there were five (12.5%) patients with recurrences, all belonging to the group in whom a residual cholesteatoma was identified during the second staged surgery. The rate of residual cholesteatoma tends to decrease as age increases. The type of cholesteatoma, acquired or congenital middle ear, were not statistically related to the incidence of residual cholesteatoma. Hearing analysis showed that hearing recovery was excellent with canal wall up procedures and remained stable over five years.

**KEY WORDS:** Cholesteatoma • Paediatric • Canal Wall Up (CWU) • Second stage surgery • Ossiculoplasty • Recurrence • Residual • Hearing results • Follow up

Introduction

The debate between intact canal wall or canal wall up (CWU) and canal wall down (CWD) mastoidectomy both in children and adults is on-going since the 1970s. This issue is pertinent because, in children, the effects of deafness can have social, educational and behavioural consequences, thereby necessitating the need for hearing preservation. Though CWU mastoidectomy achieves the important objective of hearing preservation, two of its main drawbacks are recurrence of cholesteatoma due to limited intraoperative exposure and occurrence of postoperative retraction pockets due to a dysfunctional Eustachian tube (ET). This led some surgeons to focus on disease elimination by CWD mastoidectomy at the cost of hearing. However, the drawbacks of CWU, namely recurrence and retraction, were addressed respectively by the addition of a second look surgery and positioning of cartilage in the attic region to prevent retraction. This technique has gained favour among most surgeons for cholesteatoma in the paediatric population. Despite this, the CWU technique is not always a suitable option. It is not advisable to perform CWU in certain situations like extensive disease, disease in a well pneumatised mastoid, low lying dura, anterior placed sigmoid or a cleft palate, where CWD surgery is the surgery of choice.
It has been shown in various studies that recurrences of cholesteatoma in CWU surgery normally take several years to manifest. Sivola et al., reported that 92% of all their recurrences occurred within 5 years. Similar observations have been made by other authors who advocate a minimum of a 5-year follow-up. However, there have been very few studies in the past with a long-term follow-up. It is in this context that our series assumes importance as we present the results of 5-year follow-up in homogeneous study conditions.

Materials and methods
A retrospective chart review of patients operated for cholesteatoma at the ENT Department of the University of Parma, Italy from 2001 to 2005 was carried out. Of a total of 273 patients treated for COM with cholesteatomas, 54 patients in the age group of two years to 16 years who underwent surgery for acquired or congenital middle ear cholesteatomas were identified. 14 patients with simple retraction pockets and patients with revision surgeries resulting from operations performed elsewhere were excluded from the study. Two additional patients who underwent CWD procedures were excluded, resulting in a total of 40 patients operated by the CWU technique in the study population. Otomicroscopy findings, preoperative pure tone air conduction (AC), bone conduction (BC), speech reception audiometry and tympanometry were noted. HRCT was done in all cases. Otomicroscopy and pure tone audiometry findings in the 1st, 3rd, 6th, 12th, 24th months and at the 5th year were recorded. Pre- and postoperative pure tone averages (PTA) were calculated using thresholds at 500, 1000, 2000 and 3000 Hz. Residual cholesteatoma at second stage surgery and recurrences during the follow-up period were recorded. Residual disease was defined as persistence of disease due to incomplete removal revealed during second stage surgery. Recurrent cholesteatoma was defined as a newly formed disease process secondary to a retraction pocket after second stage surgery.

Surgical technique
A postauricular approach was used in all patients. Mastoidectomy was performed preserving the posterior canal wall. A posterior tympanotomy was done and the facial recess was opened. The cholesteatoma sac was identified and excised using a combined approach. A silastic sheet was placed in the middle ear and antrum followed by reconstruction of the tympanic membrane by temporalis fascia. Bone paté in case of small defects or homologous rib cartilage was used. For large defects, reconstruction was done using autologous incus. When the incus was absent, a homologous rib cartilage was used.

Reconstruction
When the stapes was present and mobile, a partial ossicular cartilage prosthesis (PORP) was sculpted. At the end of the shaft, an indentation (1 mm wide and 0.5-1 mm in depth) was created with a diamond burr in order to accommodate the capitulum of the stapes. The head of the prosthesis was in contact with the tympanic membrane or the graft used for its repair. When the malleus was present, the prosthesis was placed parallel to it and a groove was created to accommodate the handle. When the stapes superstructure was absent, a total ossicular cartilage prosthesis (TORP) was used. In this case, the shaft was longer than in a PORP but the head of the T-shaped prosthesis was identical. The end of the shaft was placed in contact with the footplate and the head was placed in contact with the tympanic membrane or the graft used for its repair.

Statistical analysis
Data was analysed with a statistical software programme (SPSS Statistics for Windows version 20, Chicago, IL). Continuous data was summarised as mean ± interval of confidence at 95% (CI). Categorical data was presented as frequencies and percentages. Factors related to the rate of recidivism were estimated with univariate analysis. Preoperative and postoperative hearing results were evaluated for the entire sample and the acquired and congenital middle ear cholesteatoma groups. P values below 0.05 were considered statistically significant.

Results
40 patients fitted into this study criteria of CWU procedures for paediatric cholesteatomas. 31 (77.5%) patients had acquired cholesteatoma and 9 (22.5%) had congenital middle ear cholesteatoma. 24 (60%) were males and 16 (40%) were females, with an average age of 10.10 years (95% CI 8.83-11.37; range 2-16 years) at the time of surgery. The age distribution is shown in Figure 1.

Clinical findings
The most common symptoms were otorrhoea seen in 34 (85%) of patients, followed by hearing loss in 19 (47.5%) patients. Otorrhoea and hearing loss coexisted in 15 (37.5%) patients. According to Zini & Sanna’s classification, the cholesteatoma was localised in the mesotympanum (type A) in 20 (50%) patients, in the epitympanum (type B) in 10 (25%) patients, in the mesoepitympanum (type AB) in one (2.5%) patient and retro tympanum (congenital middle ear) in nine (22.5%) patients (Fig. 2). In two (5%) patients a polyp was found in the external auditory canal. Otomicroscopy in the contralateral ear showed...
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a retraction pocket in the tympanic membrane in 13 (32.5%) patients, middle ear effusion in 2 (5%), atelectasis of the middle ear in 3 (7.5%), a large tympanosclerosis in one (2.5%) and a tympanic membrane perforation in one (2.5%) patient.

Surgical procedure
All 40 patients underwent CWU surgery. No labyrinthine fistulae were detected intraoperatively in any patient. The cholesteatoma was found to be diffuse in 30 (75%) and encapsulated in 10 (25%) cases. In all but one patient, CWU mastoidectomies were performed in two stages. In the remaining case, CWU was performed in a single stage owing to the small size of the cholesteatoma and preservation of the ossicular chain. In 22 (55%) patients, the scutum was found to be eroded and was reconstructed with bone patè or homologous rib cartilage. Ossicles were found to be eroded in 39 (97.5%) patients and were reconstructed in all. Homologous rib cartilage was used in 21 (53.8%) patients, autologous malleus in 4 (10.3%) and incus in 14 (35.9%).

Recidivism in the second stage surgery
Of the 39 patients who underwent a second stage surgery, 18 (46.1%) had a residual lesion. In eight (44.4%) patients the cholesteatoma appeared as a single pear shaped lesion, in seven (38.9%) as multiple pear shaped lesions and in three (16.7%) as a diffuse lesion. In three of the 18 (16.7%) patients, ossicular reconstruction was postponed to a third stage after another 6-12 months due to infiltrative disease. Of nine patients with congenital middle ear cholesteatomas, four (44.4%) had a residual cholesteatoma found during second staged surgery. The clinical type of cholesteatoma (acquired vs. congenital middle ear) and histological type (encapsulated vs. diffuse) were not statistically related to the incidence of residual cholesteatoma (p = 0.85 and 0.24 respectively, chi-square test). The average age in the group of patients that had a residual lesion was 8.72 years (95% CI 6.80-10.70), and 11 years (95% CI 9.30-12.79) (p = 0.06, U Mann-Whitney test) in the group that did not have a residual lesion. This difference, although not statistically significant, is worthy of mention as this was the only factor closest to achieving significance among all factors analysed in relation to the incidence of residual lesion. Moreover, when patients were categorized by age into three groups (≤5 years old, 6 and 10 years and ≥11 years), the rate of residual cholesteatoma was 66.7%, 46.7% and 38.9%, respectively (p>0.05 Fisher’s exact test).

Follow-up
All patients were followed-up for at least five years. The mean follow-up was 84 months (range 60-120 months). Eight (20%) patients had a 10 year follow-up, 24 (60%) between six and nine years and eight (20%) patients for five years. In the follow-up period, there were five (12.5%) patients with recurrences, all belonging to the group in whom a residual cholesteatoma was identified and removed during the second staged surgery. Of these, one was converted into a CWD mastoidectomy, while in another case, due to extensive pathology and erratic follow up, a radical mastoidectomy was done. The cavity was obliterated with fat and a cul-de-sac closure of EAC was performed. In the only patient who underwent a single stage CWU mastoidectomy, a suspicious residual cholesteatoma was detected after 2 years and the patient underwent a second stage surgery. The cholesteatoma was confirmed to be near the stapes, which was removed. The average duration of onset of recurrence was 2.5 years after surgery (range 1-4 years). There were no recurrences in the group of patients who were disease-free during the
second staged surgery. Three patients developed a retraction pocket, one developed an atelectasis and three patients developed a glue-ear. There were no cases of perforations of the tympanic membrane.

**Hearing outcomes**

Preoperatively, pure tone audiometry could not be conducted in two patients because one suffered from Down’s syndrome and the other was too young (two years) at the time of surgery. Pre- and postoperatively pure tone audiograms of 38 patients with CWU mastoidectomies with a follow-up of five years were available and analysed. Of the 38 patients, six were excluded from further study because one suffered from Down’s syndrome and five had recurrent cholesteatoma after the second stage surgery. In addition, the audiogram of one patient was incomplete and therefore excluded. Consequently, pre- and postoperative audiograms of 31 patients were compared. No patients developed a dead ear postoperatively.

**Hearing results in the whole sample**

The mean air conduction (AC), bone conduction (BC) and air bone gap (ABG) noted preoperatively, six months and five years postoperatively are shown in Table I and Figure 3. Postoperatively, AC improved significantly from 36.21 dB to 28.06 dB (p = 0.02, Wilcoxon test). This result remained significant at five years of follow-up (p=0.006). There was also a significant improvement between preoperative ABG (23.53 dB) and postoperative ABG at 6 months (17.24 dB) (p = 0.01). This improvement consolidated at five years of follow-up and remained significant (p = 0.001). Mean preoperative BC was 12.47 dB and remained unchanged postoperatively at six months (p = 0.75) and five years (p = 0.43) after surgery. Preoperatively, the average ABG was < 10 dB in four (10.5%) patients, between 11 and 20 dB in nine (26.3%) patients, between 21 and 30 dB in 10 (28.9%) patients and > 30 dB in 11 (34.2%) patients. Postoperatively at six months follow-up, ABG was < 10 dB in 12 (34.3%) patients, between 11 and 20 dB in 10 (28.6%) patients, between 21 and 30 dB in seven (20%) patients and > 30 dB in six (17.1%) patients. At five years of follow-up, the ABG was < 10 dB in 10 (32.3%) patients, between 11 and 20 dB in nine (29%) patients, between 21 and 30 dB in 8 (25.8%) patients and > 30 dB in four (12.9%) patients (Fig. 4).

**Hearing results in acquired cholesteatoma**

The mean AC, BC and ABG preoperatively, and postoperatively at six months and five years in patients with acquired cholesteatoma are shown in Table II. The mean AC improved from 34 dB before surgery to 28 dB at 6 months and at 5 years (p = 0.07, U Mann-Whitney test). The reduction in ABG was statistically significant from a mean ABG of 22 dB before surgery to 17 dB at 6 months and 5 years (p = 0.007). Mean preoperative BC, which was 12 dB, remained unchanged postoperatively at six months (p = 0.13) and five years (p = 0.22).

**Hearing results in congenital middle ear cholesteatoma**

The mean AC, BC and ABG preoperatively and postoperatively at six months and five years for patients with congenital middle ear cholesteatoma are shown in Table III. Among the nine patients with congenital middle ear cholesteatomas, one patient suffering from Down’s syndrome was non-compliant. In the remaining patients, there was significant improvement between preoperative AC (mean PTA of 42.50) and postoperative AC (mean PTA of 25.63 dB) at six months (p = 0.042, Wilcoxon test). The reduction in ABG from 27.96 dB preoperatively to 17 dB postoperatively at six months was statistically significant (p = 0.006). Both results were statistically significant at

### Table I. Hearing outcomes in middle ear cholesteatomas after CWU surgery preoperatively, 6 months and 5 years of follow-up.

|                       | Preoperative | Postoperative | Follow-up at 6 months | Follow-up at 5 years |
|-----------------------|--------------|---------------|-----------------------|----------------------|
| Mean PTA (95% CI)     |              |               |                       |                      |
| Air conduction        | 36.21 (26.96-42.45) | 28.06 (23.23-32.89) | 26.46 (21.66-31.26)   |                      |
| Bone conduction       | 12.47 (9.41-15.54)   | 10.80 (9.06-12.54)    | 10.43 (8.21-12.64)    |                      |
| Air bone gap          | 23.53 (18.99-28.08) | 17.24 (12.69-21.80)   | 16.12 (11.91-20.32)   |                      |

![Fig. 3. Box-plot showing median AC, BC and ABG according to follow-up. Preoperatively (PRE), 6 months after surgery (6M) and at 5 years follow-up (5Y).](image-url)
five years follow up \((p = 0.028\) and \(p = 0.028\) respectively). Mean preoperative BC, which was 13 dB, remained unchanged postoperatively at six months \((p = 0.86)\) and five years \((p = 0.22)\).

There were no statistically significant differences in the preoperative AC, BC and ABG between acquired and congenital middle ear cholesteatoma groups \((p > 0.05, \ U \ Mann-Whitney\) test). However, in the congenital group, the improvement in AC \((20.20\) dB, 95% CI 28-12) and ABG \((-14.20\) dB 95% CI, -21 -[-7.5]) at five years was better than the AC \((7\) dB, 95% CI 15-[-1]) and ABG \((-5.6\) dB 95% CI, -11 -[-0.33]), in the acquired group \(p=0.017\) and \(p=0.029\), respectively \((U \ Mann-Whitney\) test).

**Discussion**

The incidence of cholesteatoma in childhood is estimated to be 3-6 per 100,000 individuals \(^6\). Various arguments dictate the surgical choice in paediatric cholesteatoma. While some authors \(^7-10\) consider that cholesteatoma is a more aggressive in children with a higher percentage of recidivism than in adults and hence warrants more radical surgery, others \(^11\, 12\) are of the opinion that the behaviour of the cholesteatoma is the same in children as in adults. The second disagreement is that of CWU versus CWD surgery in dealing with paediatric cholesteatomas. While it is important to preserve hearing in the child, which plays a major role in social and educational integration after surgery, it is even more important to eradicate the disease without complications. The introduction of the staged procedure in CWU, use of cartilage for reconstructing the attic to prevent postoperative retractions and advances in ossicular reconstruction have helped surgeons to tackle cholesteatomas in the paediatric population with a CWU technique, thereby preserving hearing and eliminating the risk of recurrence. Furthermore, studies using DW-non-EPI and DWI propeller sequences show promising results in improved diagnostic sensitivity and specificity for even small (< 5 mm) cholesteatomas, thus allowing avoidance of second-look surgery in the future \(^13-16\).

**Principles behind surgery**

We prefer a CWU mastoidectomy with a planned second stage surgery as the primary option. A CWD is used in patients with extensive cholesteatoma, difficult anatomy, cleft palate and unsure follow up. The second stage sur-

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**Table II.** Hearing outcomes in acquired cholesteatoma.

| PTA results | Preoperative | Follow-up 6 months | Postoperative | Follow-up 5 years |
|-------------|--------------|---------------------|---------------|-------------------|
| PTA AC      | 34 (27-41)   | 29 (23-34)          | 28 (22-33)    |
| PTA BC      | 12 (9-16)    | 12 (10-13)          | 11 (8-13)     |
| ABG         | 22 (17-28)   | 17 (12-22)          | 17 (12-22)    |

**Table III.** Hearing outcomes in congenital middle ear cholesteatoma.

| PTA results | Preoperative | Follow-up 6 months | Postoperative | Follow-up 5 years |
|-------------|--------------|---------------------|---------------|-------------------|
| PTA AC      | 42.50 (31.97-53.03) | 25.63 (11.02-40.23) | 22.29 (12.70-31.87) |
| PTA BC      | 13.00 (7.65-18.35)    | 7.42 (1.29-13.54)   | 8.33 (2.91-13.75) |
| ABG         | 27.96 (20.10-35.82)   | 17.01 (11.80-22.22) | 13.75 (4.32-23.17) |
gery is planned 6-12 months after the 1st surgery. This is because the healing process of a middle ear with active infection, granulation tissue, and/or areas of absent mucosa will be variable. Formation of adhesions between the medial wall of the mesotympanum and the tympanic membrane or the ossicular implant can promote tympanic membrane retraction and/or implant displacement. To avoid this, we place a silastic sheet in the middle ear which remains until the time of the second stage surgery, when it is removed and ossicular reconstruction is performed. If residual cholesteatoma is present and is a small pearl, we remove it and continue with ossicular reconstruction in the same session. If it is found to be extensive, or in the presence of infection, the cholesteatoma is removed and the ossiculoplasty is postponed to another session. We do not hesitate to convert the CWU into a CWD as and when necessary.

In assessing outcomes in our series, we agree with previous authors that the total percentage of cholesteatoma recurrence should be the basis for evaluation of the outcome of surgery. We defined residual disease as persistence of disease after incomplete removal as revealed during second stage surgery. Recurrent cholesteatoma was defined as a newly formed disease process secondary to a retraction pocket after second stage surgery. It is unlikely that a good surgeon will leave behind (residual) cholesteatoma after two surgeries. This differentiation is crucial in reporting data. We compared our study to those of other authors, and the results are summarised in Table IV. In our series, as in most others, the residual cholesteatoma was identified during the second stage procedure mostly in the form of a white pearl that could be removed easily. The recurrent cholesteatoma presented to us between a minimum of one year and a maximum of five years, possibly due to poor Eustachian tube function. The age difference between patients who had a residual lesion (average 8.72 years), and those who did not (average 11 years) was close to significance (p = 0.06). It is also important to note that the rate of residual cholesteatomas decreases as the age progresses [(66.7% (≤ 5 years), 46.7% (6-10 years) and 38.9% (≥ 11)], respectively (p > 0.05 Fisher’s exact test). This may point to the fact that younger patients may indeed have a more aggressive disease. The reported rate of residual cholesteatoma in closed procedures is between 19-63.5% and that of recurrent cholesteatoma is between 7.5-22%. While none of our patients who did not have a residual cholesteatoma identified at the second stage went on to develop a recurrence, Schraff et al. reported a 1% recurrence rate in such a category. A period of five years of follow-up is essential for all children undergoing CWU surgery.

**Hearing outcomes**

Our analysis of the hearing outcomes validates the fact that CWU procedures bring about a significant improvement in AC that is statistically significant. Moreover, this improvement is consolidated at five years and remains statistically significant. Likewise, the improvement in ABG is also maintained at five years. Patients with congenital middle ear cholesteatoma tend to have similar preoperative AC, BC and ABG than with acquired cholesteatoma. However, at 5 years, the postoperative improvement in hearing was superior in congenital middle ear cholesteatoma patients compared to those with acquired cholesteatoma.

Studies indicate that the status of the ossicles, their reconstruction and the type of surgical technique are important factors for postoperative hearing restoration in cholesteatoma surgery. Incus transposition is most frequently used for reconstructing an ossicular discontinuity. However, cholesteatoma usually causes significant destruction of ossicles, especially the incus, so that other materials must be considered. In the absence of the incus, we routinely use rib cartilage for the reconstruction of the attic and the

| Authors | # patients | Residual % | Recurrent % | Mean follow-up (years) |
|---------|------------|------------|-------------|-----------------------|
| Dodson et al. | 66 | 19 | 12 | 22 | 0 | 9.2 |
| Mutlu et al. | 83 | 38 | 11 | 11 | 0 | 4 |
| Sanna et al. | 151 | 40 | 50 | 11 | 25 | 1.5 |
| Schimd et al. | 57 | 24 | 7.5 | 12 | 7.5 | 7 |
| Magnan et al. | 210 | 26 | 19.5 | 0 | NA |
| Triglia et al. | 80 | 41 | 35 | 16 | 8 | 4 |
| Desaulty et al. | 80 | 63.5 | 0 | 7.5 | 0 | 2.5 |
| Roger et al. | 199 | 54 | 72 | 19 | 0 | 2.5 |
| Lerosey et al. | 57 | 26 | 28 | 12 | 12.5 | 7 |
| Charachon et al. | 160 | 31 | 38 | 20 | 0 | NA |
| Our series | 43 | 45 | 0 | 12 | 0 | 8 |

NA=Not available
ossicles. Because of their flexibility and elasticity, cartilage prostheses rarely damage the stapes or the footplate during the surgical maneuver. As stated by Dodson et al., serviceable hearing depends primarily on middle ear parameters such as mucosal status (stenosis and granulation), condition of the TM (thickness, contour), depth of the middle ear cleft and presence or absence of stapes superstructure erosion. Mutlu et al. demonstrated a ≤ 25 dB hearing loss in 85% of those with an intact stapes superstructure. Historically, worse hearing results have often been cited as a criticism of CWD but several studies dispute this notion. Schraff et al. showed that those undergoing CWD have worse hearing pre-operatively and less improvement post-operatively compared with children using the ICW approach. This may be a result of more aggressive disease rather than failure in technique. Nevertheless, CWU surgeries provide a distinct advantage due to the fact that children do not need to worry about mastoid cavities, can swim and may benefit from a good fitting of a hearing aid when necessary.

Conclusions

A two-staged CWU mastoidectomy is the surgery of choice for paediatric cholesteatomas. Placing a silastic sheet to promote regeneration of mucosa with ossiculoplasty in the second stage and reconstruction of the scutum to avoid retraction have made CWU procedures an effective option in dealing with paediatric cholesteatomas. Hearing analysis showed that hearing recovery was excellent with canal wall up procedures and remained stable over five years. There is a higher incidence of recurrence during long-term follow-up in patients in whom residual cholesteatoma was detected during second stage surgery. The rate of residual cholesteatoma tends to decrease as the age increases. Follow-up for a minimum of five years is recommended in all patients with paediatric cholesteatoma.

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