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## ORIGINAL ARTICLE

# Acquired cholesteatoma in children: Strategies and medium-term results

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### KEYWORDS

Cholesteatoma;  
Child;  
Audiometry;  
Deafness;  
Middle ear surgery

### Summary

**Objectives:** To assess paediatric cholesteatoma surgical management strategies, residual disease and recurrence rates and especially the medium-term auditory impact.

**Material and methods:** Retrospective study of 22 cases of acquired middle ear cholesteatoma selected from a series of 77 children under the age of 16 operated for cholesteatoma between 1st January 2000 and 31st December 2003 on the basis of the following criteria: first-line surgical management with postoperative follow-up greater than four years. Surgical strategies, preoperative and postoperative (at 1 year and at the final visit) audiograms and residual disease and recurrence rates were analysed.

**Results:** A canal wall up tympanoplasty was performed in 82% of cases as first-line procedure and a canal wall down tympanoplasty was performed in 32% of cases. Residual cholesteatoma was observed in 9% of cases and recurrent disease was observed in 18% of cases. The mean preoperative hearing loss was 26 dB with an air-bone gap of 23 dB with values of 26 dB and 20 dB respectively at the end of follow-up.

**Conclusion:** The majority of children were operated by two-stage canal wall up tympanoplasty. Long-term hearing results remained stable and close to preoperative values. The recurrence rate (residual disease and relapse) was low (27%), as reported in the literature.

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## Introduction

Acquired cholesteatoma in children is an aggressive disease due to its rapid growth and high recurrence rate. This disease raises two main problems for the surgeon. The first problem obviously concerns local control, which requires eradication of the disease process, while preventing recurrence. The second problem concerns the

hearing rehabilitation strategy, which is essential to prevent learning difficulties. The choice of resection technique, either canal wall down tympanoplasty (CWDT) or canal wall up tympanoplasty (CWUT), is still a subject of debate and must be determined for each individual case, especially according to the extent of the cholesteatoma and the surgeon's experience. Although the chances of eradication and prevention of recurrences are considered to be better with CWDT, the hearing prognosis is considered to be better with CWUT. In reality, the hearing prognosis appears to depend on the status of the stapes and especially preservation of the stapes superstructure [1–3]. In order to evaluate our

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clinical practice, we decided to study all cases of acquired cholesteatoma operated over a four-year period with sufficient follow-up to allow evaluation of the medium-term hearing results and the final disease status. The various surgical strategies used were also reviewed.

## Material and methods

Seventy-seven children under the age of 16 were operated for middle ear cholesteatoma in our paediatric otorhinolaryngology department over a 4-year period between 1st January 2000 and 31st December 2003. Twenty-two cases of acquired middle ear cholesteatoma affecting 21 of these 77 children were included in this study on the basis of the following criteria: first-line surgical management, appropriate postoperative follow-up for more than four years. To ensure a study sample representative of our department's activity, all cases were included regardless of the operator, although a majority (54%) were operated by the senior author (JPM). Cases of congenital cholesteatoma were not included in this study.

The sex distribution, mean age at the time of the first operation and mode of discovery of cholesteatoma were reported.

Surgical strategies, especially the resection and reconstruction techniques and the number of operations were described together with the main operative findings. Canal wall down tympanoplasty is indicated as first-line procedure in our centre in children with a small mastoid with limited air cells or meningeal prolapse and as second-line procedure for recurrence. It is often combined with reconstruction by a musculoperiosteal Palva flap.

The residual and recurrent cholesteatoma rate was calculated.

Preoperative audiometry, and audiometry before the second operation or at one year in the case of a one-stage procedure and the last available audiometry were analysed with calculation of air conduction (AC) and bone conduction (BC) thresholds and air-bone gap by averaging thresholds at 500 Hz, 1 kHz, 2 kHz and 4 kHz, according to the recommendations of the International Bureau for Audiophonology.

## Results

The mean age at the first operation was 9.7 years (range: 5–14 years). This series comprised 14 boys (67%) and seven girls (33%). Cholesteatoma was usually discovered in the context of follow-up of chronic otitis media (serous otitis media or retraction pocket) (54%), otorrhoea (18%) and hypoacusis (18%). One case was discovered incidentally and another case was discovered in the context of acute mastoiditis, the only case presenting at the stage of complications in this series. No intracranial complications were observed.

The mean postoperative follow-up was 74 months (standard deviation 15.1 months) after the first operation and 47 months (standard deviation 29.9 months) after the last operation.

Intraoperative prophylactic antibiotics (amoxicillin-clavulanic acid) were systematically used for cholesteatoma

surgery at the time of this series, but have no longer been used since 2005 [4].

CWUT was performed as the first-line procedure in 82% of cases, combined with a mastoid procedure in 95% of cases and posterior tympanotomy in 50% of cases. CWDT was performed in 32% of cases at various stages: 18% as the first-line procedure and 14% as third-line procedure. All CWDTs were completed by reconstruction of the cavity by a musculoperiosteal Palva flap.

At least two surgical procedures were performed for 82% of patients, while the other patients were submitted to clinical and CT follow-up. When a second surgical procedure was initially planned, follow-up CT was not systematically performed prior to this second procedure. Eighteen percent of children underwent a single operation, 50% were operated twice, 23% were operated three times and 9% were operated four times. The mean interval between the first two procedures was 19.2 months (range: 9–56 months).

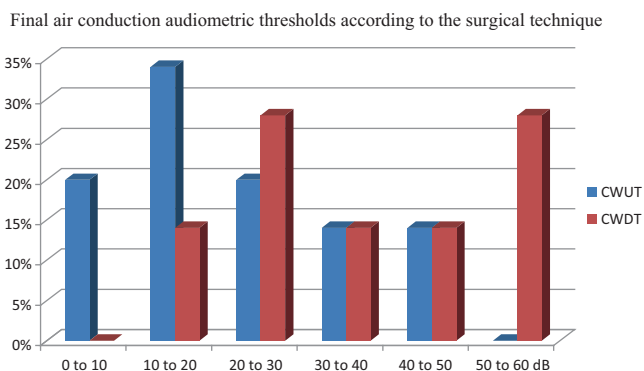
Ossicular reconstruction was possible in 18% of cases during the first-line procedure and was deferred until the second procedure in 68% of cases. Ossiculoplasty was not required in 14% of cases. Ossicles were removed during the first procedure in 64% of cases (incus in 64% and head of malleus in 18%). When ossicular reconstruction could not be performed during a one-stage procedure, fine Silastic sheeting was placed at the back of the tympanic cavity over the stapes. Reconstruction consisted of incus transposition in 33% of cases, cartilage graft onto stapes in 27% of cases, titanium total ossicular replacement prosthesis in 33% of cases and titanium partial ossicular replacement prosthesis in 6% of cases.

Cartilage augmentation of the tympanic membrane was systematically performed during the first or second stage of CWUT.

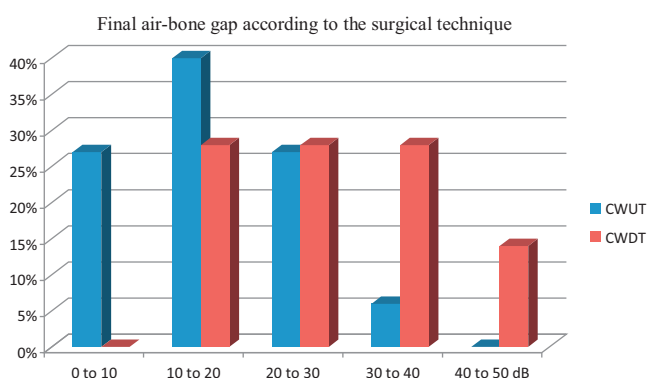
Residual or recurrent cholesteatoma was detected in 18% of cases during the second operation (residual disease in 9% of cases and recurrence in 9% of cases). Two cases of late recurrence (9% of cases) were treated by a third procedure. All recurrences occurred after CWUT, requiring subsequent CWDT in one half of cases. No subsequent recurrences were observed.

The region most commonly affected in this series was the atrium (77% of cases), followed by the attic (41%). All middle ear cavities (atrium, attic and mastoid) were invaded in 14% of cases. The ossicular chain was disrupted in 41% of cases. The ossicle most commonly destroyed was the incus (36%), followed by the stapes (18%). No cases of malleus destruction were observed in this series.

Audiometric assessment showed that the affected ear presented a mean preoperative AC hearing loss of 26 dB with a mean air-bone gap of 23 dB, associated with a mean AC hearing loss of 15 dB with a mean air-bone gap of 13 dB in the contralateral ear. One year after the first-stage procedure, before performing the second stage, the mean AC hearing loss was 46 dB with a mean air-bone gap of 37 dB and a mean AC hearing loss of 7 dB in the contralateral ear. The mean final AC hearing loss was 26 dB with a mean air-bone gap of 20 dB. In the CWUT group, the mean final AC hearing loss was 22 dB with a mean air-bone gap of 16 dB while in the CWDT group; the mean final AC hearing loss was 36 dB with a mean air-bone gap of 30 dB (Figs. 1 and 2). In patients with initial ossicular destruction, the mean final hearing loss was 34 dB



**Figure 1** Distribution of final air conduction audiometric thresholds (four or more years after the diagnosis) with canal wall up tympanoplasty (CWUT) and canal wall down tympanoplasty (CWDT).



**Figure 2** Distribution of final air-bone gap (4 or more years after the diagnosis) with canal wall up tympanoplasty (CWUT) and canal wall down tympanoplasty (CWDT).

with a mean air-bone gap of 25 dB. In the case of destruction of the stapes superstructure, the mean final hearing loss was 30 dB with a mean air-bone gap of 23 dB. Hearing results as a function of the type of ossiculoplasty are represented in Table 1.

Five of the 21 (24%) children use a hearing aid: an air conduction hearing aid in four cases (three children operated by CWUT and one operated by CWDT) and a bone conduction hearing aid in one case (operated by CWDT).

**Table 1** Mean hearing loss according to the type of ossicular reconstruction.

	CA (dB)	Air-bone gap (dB)
Incus transposition <i>n</i> = 6	23	16
Cartilage graft onto stapes <i>n</i> = 5	30	23
Titanium TORP <i>n</i> = 6	38	31
Titanium PORP <i>n</i> = 1	35	33

TORP: titanium total ossicular replacement prostheses; PORP: partial ossicular replacement prosthesis.

All cavities resected by CWDT are currently stable. No cases of facial palsy, labyrinthization or cophosis were observed.

### Discussion

The main objectives of management of cholesteatoma are eradication of the disease and hearing rehabilitation. Cholesteatoma in children is reputed to be more aggressive than in adults. For example, De Corso et al. [5] reported a recurrence rate (defined as the residual disease plus relapse rate) of 26.6% in children versus 6.8% in adults and more frequent ossicular lesions in children. Charachon et al. [3] reported residual disease in 31% of children after CWUT versus 16% in adults. In the present series, the recurrence rate was 27% with 9% of residual cholesteatoma and 18% of relapses, in line with the results published in the literature (Table 2).

Eradication is obviously more complex in the presence of advanced disease, hence the importance of early diagnosis. The majority of cholesteatomas of the present series were diagnosed in the context of follow-up of chronic otitis media (54%), indicating the need for long-term follow-up of children with a history of chronic otitis media.

CWUT is the preferred operative technique in our centre, performed as first-line procedure in 82% of cases. Some authors consider that this technique allows more effective hearing rehabilitation [1,6] with, in our series, a mean final AC hearing loss of 22 dB with an air-bone gap of 16 dB for CWUT versus a mean final AC hearing loss of 36 dB with an air-bone gap of 30 dB for CWDT. CWDT also requires the creation of a large cavity, sometimes associated with difficult healing and frequent episodes of otorrhoea, and for which fitting of a hearing aid may be more complex. However, we perform first-line CWDT in patients with a small mastoid, limited mastoid air cells or meningeal prolapse. Reconstruction by a musculoperiosteal flap decreases problems related to the healing of the cavity by reducing its volume. None of the children of the present series presented an unstable cavity.

In our centre, ossicular reconstruction is primarily performed using autologous material: incus transposition (33% of reconstructions), cartilage graft onto stapes (28%), which avoids the problems of long-term tolerability of prostheses in this paediatric population, as well as the risks of extrusion, as observed in one case of this series, requiring redo surgery.

Up until recently, the treatment strategy adopted by our team systematically involved a two-stage surgical procedure. High-resolution CT scanners and MRI, especially using diffusion and late sequences, now allow good quality radiological follow-up, eliminating the need for the second stage of the procedure, provided several conditions are met during the first stage: experienced operator, resection considered to be complete, healthy mucosa and satisfactory aeration of the middle ear. When these conditions are met, ossiculoplasty can be performed during the first operation. This change of strategy occurred during the inclusion period of the present study, which explains why this series comprised children treated by a one-stage procedure and by a systematic two-stage procedure. Postoperative follow-up after

**Table 2** Recurrence rate (residual + relapse) reported in various paediatric series.

	Residual disease rate	Relapse rate	Recurrence rate
Darrouzet et al. [1]	21.5%	10%	31.5%
Mutlu et al. [2]	38%	10%	48%
Charachon [3]	31% with CWUT 36% with CWDT	—	—
De Corso et al. [5]	—	—	26.6%
Soldati [8]	—	—	29%
Schraff [11]	—	—	16%
Dodson et al. [12]	—	—	36%
Shirazi et al. [13]	1.2%	3.5%	4.7%
Lerosey et al. [14]	29%	11%	40%
Göçmen et al. [15]	—	—	26% with CWUT 16% with CWDT
El Jerrari et al. [16]	—	—	54% with CWUT 29% with CWDT
Roger et al. [17]	34% at 5 years	—	—
Our series	9%	18%	27%

CWUT: canal wall up tympanoplasty; CWDT: canal wall down tympanoplasty.

a one-stage procedure is ensured by CT, completed by MRI only when the nature of suspicious opacities needs to be more clearly defined.

The final audiometric results observed in this series (AC hearing loss of 26 dB and an air-bone gap of 20 dB) are in line with those reported in the literature. Darrouzet et al. [1] reported a final AC hearing loss of 28.1 dB with an air-bone gap of 22.8 dB, while Iino et al. [7] reported a final AC hearing loss of 27.1 dB with an air-bone gap of 20 dB. Similarly, Soldati et al. [8] reported an air-bone gap less than 20 dB in 51% of cases versus 57% of cases for De Corso et al. [5]. Mutlu et al. [2] reported an air-bone gap of less than 25 dB in 68% of cases and Mishiro et al. [9] reported a final AC hearing loss of less than 20 dB for 12.5% of cases.

Air-bone gaps reported in the various series are shown in Table 3.

Initial ossicular involvement represents a factor of poor hearing prognosis. A final hearing loss of 34 dB with a mean air-bone gap of 25 dB was observed in these cases. One of the essential elements determining the hearing prognosis appears to be preservation of the stapes superstructure [1]. For example, Mutlu et al. [2] reported an air-bone gap of less than 25 dB in 85% of cases when the stapes superstructure was preserved versus 53% of cases when the stapes superstructure was eroded. Similarly, Charachon et al. [10] reported an air-bone gap of less than 20 dB in 63% of patients treated by CWUT and 59% of patients treated by CWDT with an intact stapes versus 45% and 46% in the case of erosion of

**Table 3** Mean postoperative audiometric air-bone gap reported in various paediatric series.

	Mean postoperative air-bone gap		Postoperative air-bone gap $\leq$ 20 dB
	CWUT	CWDT	
Darrouzet et al. [1]	21 dB	31.5 dB	—
De Corso et al. [5]	—	—	57%
Murphy [6]	19	29.6 dB	—
Soldati [8]	—	—	51%
Schraff [11]	20.2 dB	33.1 dB	—
Lerosey et al. [14]	16 dB	27 dB	—
El Jerrari et al. [16]	—	47% with CWUT 20% with CWDT	—
Our series	16 dB	30 dB	59%

CWUT: canal wall up tympanoplasty; CWDT: canal wall down tympanoplasty.

the stapes superstructure, respectively. No significant audiometric difference was demonstrated in the present series, but based on a limited sample size.

The contralateral ear is generally also pathological with a mean air-bone gap of 13 dB preoperatively, corresponding to serous otitis media. The long-term course of the contralateral ear is favourable, with an air-bone gap of 5 dB one year after surgery that subsequently remained stable and AC hearing loss inferior or equal to 10 dB in 64% of cases and inferior or equal to 20 dB in 92% of cases.

## Conclusion

Eradication of cholesteatoma is more difficult to obtain in children than in adults. The various ossiculoplasty techniques provide good audiometric results and constitute a major element of the management, as the contralateral ear also initially presents a hearing loss that may impair the child's learning capacities. The use of autologous material allows recovery of the columellar effect and titanium total ossicular replacement prostheses (TORP) give good results in the case of destruction of the stapes. Early diagnosis, allowing early treatment, is a key element of the prognosis. In this small series of children mostly operated by a two-stage canal wall up tympanoplasty, the medium-term hearing results at 4 years were stable and close to the preoperative results. The recurrence rate (residual and relapse) in this series is in line with those reported in the literature. All these results support our team's management strategy for this disease, which now comprises imaging (CT and MRI). The results of this clinical practice evaluation study can be used as a basis for subsequent evaluations.

## Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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