

Cholesteatoma in children: results in open *versus* closed techniques

J. MARCO-ALGARRA, F. GIMENEZ, I. MALLEA, M. ARMENGOT, L. DE LA FUENTE (Valencia, Spain)

Abstract

Cholesteatoma in children presents characteristics that differentiate it from the adult condition. Surgery has been performed in 55 ears. The main reason for referral was otorrhoea (83 per cent), either alone (29 per cent) or with hearing loss (54 per cent). Apart from three cases with an intact eardrum, a perforation was always present mostly posterior-superior (50 per cent). Open techniques were performed in 27.3 per cent of the ears and the closed technique in 72.7 per cent. Only in 18 per cent of cases was the ossicular chain normal and mobile. The attic was occupied by cholesteatoma in 79 per cent. Recurrence took place in 37 per cent and 13 per cent of the closed and open techniques respectively. Open techniques tend to be employed in the presence of extensive disease, whereas the closed technique is reserved for those with a more localized problem.

Introduction

The concept of cholesteatoma in children (CC) as a pathological entity in itself, with features that differentiate it from adult cholesteatoma is open to controversy. Although the incidence among children is admittedly lower than in adults, agreement is lacking on practically all other characteristics of the disease. A number of authors claim that CC exhibits greater aggression and incidence of complications and recurrences. Charachon (1985) attributes this to the greater growth potential of tissues in childhood, along with common secondary infection and progression of the normal evolution of mastoid pneumatization that offers less resistance to the cholesteatoma. In this sense, Ruedi (1958) also refers to the presence of fibrous, low-density mastoid tissue that is easily destroyed, thus facilitating expansion. Conversely, other authors come to opposite conclusions (Sheehy, 1985).

Diagnosis is often difficult and laborious. The condition tends to present as small perforations that contrast with the large size of the CC situated behind them. The growth is easily masked by wax scabs, slight oedema of the surrounding tissue, and external desquamative otitis, (Jahnke, 1982), and only a thorough microscopic examination allows detection. Much has been said of its relation to the Eustachian tube; indeed CC may be masked by middle ear suppurative otitis (MSO); the latter may in turn be maintained by frequent superinfection of the CC, although it cannot be established whether these infections trigger CC following retraction of the eardrum, or whether the CC maintains a state of irritation of the middle ear mucosa; perhaps both processes coexist with no correlation whatsoever (Bluestone *et al.*, 1978; Andrieu-Guitrancourt *et al.*, 1980).

Likewise no agreement exists on the surgical procedure of choice in CC. While authors such as Palva *et al.*

(1977), Andrieu-Guitrancourt *et al.* (1980) or Jahnke (1982) prefer open techniques, Glasscock *et al.* (1981), Charachon *et al.* (1984) or Sheehy (1985) among others prefer closed approaches. A third opinion is based on the use of both open and closed techniques, associated with the so-called 'on demand' techniques (Olaizola, 1985); in each case the percentage of residual cholesteatoma varies markedly, between the 50 per cent reported by Charachon *et al.*, (1984) and seven per cent according to Jansen (1978).

Intact ear drum (or congenital) cholesteatoma is of maximum interest in childhood; diagnosis is becoming more common as a result of improved knowledge of the existence of the condition and the development of more sophisticated exploration techniques that have made detection possible before reaching the suppurative stage. Cholesteatoma may in time perforate the membrane (Olaizola, 1985) and exteriorize; the condition may then be diagnosed as either primary or secondary cholesteatoma. This explains the difficulty encountered in applying Derlacki and Clemis (1965) criteria to secure strict diagnosis, and the need for a broader approach to studying the disease.

In the present study we report our experience in a group of 52 patients (55 ears)—maximum age being 14 years—diagnosed as middle ear cholesteatoma and operated in the Hospital Clínico Universitario of Valencia (Spain) between 1974 and 1987. These paediatric patients represented 20 per cent of the total number of cholesteatoma cases diagnosed. Mean duration of follow up after surgery was 7.1 years (range 3–16 years).

Materials and method

Clinical data

Twenty-seven right and 28 left ears were operated on.

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TABLE I
SYMPTOMATOLOGY AT THE FIRST CONSULTATION

Main symptoms		Additional symptoms	
Otorrhoea + hearing loss	30 cases (54%)	Pain	7 cases (13%)
Otorrhoea only	16 cases (29%)	Dizziness	2 cases (3.8%)
Hearing loss	4 cases (7.6%)	Tinnitus	2 cases (3.8%)
Mastoiditis	3 cases (5.4%)	Fever	1 case (1.9%)
Pain	1 case (1.9%)		

There were 25 males and 27 females. Mean age at the time of surgery was 9.8 years (range 4–14). Sex was not a significant factor in age distribution.

In the patient's histories there was a high proportion of tonsillectomies and, in particular, adenoidectomies—a point of no significance in view of its frequency among children. In only one patient were bilateral ventilation tubes fitted for MSO.

The main reason for referral was otorrhoea (83 per cent), either alone (29 per cent) or accompanied by hearing loss (54 per cent). Other symptoms are presented in Table I.

Otorrhoea was foetid in 83 per cent of cases, and in 87 per cent was closely related to 'colds'. The condition of the eardrum is shown in Table II. The discharge was cultured and strains of *Proteus*, *Staphylococcus aureus* and *Pseudomonas aeruginosa* were isolated in equal portions.

Audiometry showed hearing to be normal or near normal (threshold under 20 dB) in four cases (seven per cent). In another 37 patients (69 per cent), the threshold appeared between 20 and 40 dB, whereas in the 12 remaining cases (21 per cent) it was over 40 dB. One case presented with a mixed hearing loss with the bone conduction threshold at 40 dB and air conduction at 90 dB. The final case presented a dead ear with a history of cranioccephalic trauma and loss of consciousness one year before.

As to complications, we observed only three cases of mastoiditis already mentioned, with no involvement of the facial nerve or intracranial complications. The posterior labyrinth was not affected in any case.

Radiography revealed the mastoid bone to be sclerotic in 25 cases (45 per cent), diploetic in ten (18 per cent) and well pneumatized in 13 (23 per cent).

The contralateral ear (the second being diagnosed in the case of bilateral disease) was otoscopically normal in 38 cases; it was retracted and dull in six, with an attic retraction pocket in three (two with otorrhoea), an attic perforation in two (both with otorrhoea), central perforation in one (also with otorrhoea), spontaneous type III eardrum in one case, and an intact eardrum with disease in the middle ear in one patient. The two attic perforations were operated on, together with the last case described above. Cholesteatomas were encountered in all three. Forty of these contralateral ears presented

normal hearing, whereas 12 presented conductive hearing loss secondary to the above mentioned processes.

Surgical data

Surgery was performed in all cases as follows:

—Open techniques: 15 ears (27.3 per cent)—five with Palva-type musculoplasty. Tympanoplasty was associated in two cases.

—Closed techniques: 40 ears (72.7 per cent) 19 with posterior tympanotomy and in four cases with simultaneous functional reconstruction.

During surgery, the middle ear structures showed the following conditions:

—Mucosa; hyperplastic and/or polypoid in 92 per cent of cases and normal in only 8 per cent.

—Ossicular chain; intact and mobile in nine cases (18 per cent) and damaged in 43 (82 per cent). The most commonly affected ossicle was the incus (70 per cent)—particularly its long process, as is common in all the studies reported, due to its precarious vascular supply. The next most commonly ossicle to be damaged was the malleus (43 per cent) and then the stapes (38 per cent), the footplate being intact in all cases.

—Cholesteatoma extension: the cholesteatoma occupied the attic in 39 cases (79 per cent), antrum in 28 (57 per cent), the tympanic cavity in 18 (36 per cent), the posterior recesses in 15 (31 per cent), the mastoid in ten (21 per cent), the peritubal zone in five (10 per cent), and the posterior portion of the external auditory canal was destroyed in one case (two per cent).

—Involvement of the horizontal semicircular canal: two cases. In one patient a true fistula of the canal was found, whereas in the other the involvement was a simple dehiscence of the bony cortex. Both cases corresponded to marginal posterosuperior perforations, coexisting with important lesions of the three ossicles but without clinical vestibular manifestations.

Results

The results obtained after the above mentioned follow-up period are evaluated in terms of three categories (Table III). We consider that the results were highly sat-

TABLE III
RESULTS: OPEN VERSUS CLOSED TECHNIQUES

	Technique employed		
	Closed	Open	Total
Success	21 (52%)	6 (40%)	27 (49%)
Otorrhoea	4 (10%)	7 (46%)	11 (20%)
Residual Ch.	15 (37%)	2 (13%)	17 (30%)
Total	40 (100%)	15 (100%)	55 (100%)

TABLE II
SITE OF EARDRUM PERFORATIONS

Normal eardrum	3 cases (5.4%)
Attic perforation	28 cases (50%)
Marginal post-perforation	16 cases (29%)
Central perforation	5 cases (8%)
Marginal anterior perforation	2 cases (3.6%)
Double attic and central perforation	1 case (1.8%)

TABLE IV
RECURRENCE AFTER SURGERY

	Technique employed		
	Closed	Open	Total
Before 1 year	1 (6%)	0	1 (6%)
Between 1–2 years	9 (60%)	0	9 (52%)
Between 2–5 years	5 (33%)	2 (100%)	7 (41%)
Total	15 (100%)	2 (100%)	17 (100%)

isfactory or a success when surgery eliminated the cholesteatoma and otorrhoea disappeared (49 per cent of the cases)—the figures being better with closed techniques. Partial success, with elimination of the cholesteatoma but otorrhoea persisted in 20 per cent of the cases most commonly in those undergoing the open technique (49 per cent). Residual cholesteatoma reappeared in 30 per cent of the patients, the results being worse with closed techniques (37 per cent) than with the open procedures (13 per cent).

In cases of residual cholesteatoma, the time interval after primary surgery is presented in Table IV. We found that with the open techniques, residual cholesteatoma always appeared late (after more than two years), whereas with the closed procedure they were infrequent (six per cent) during the first year—maximum incidence occurring in the second year (60 per cent), followed by a decrease thereafter (33 per cent).

Treatment of recurrence (17 cases): in the two ears subjected to radical mastoidectomy, a surgical cleaning of the granulation tissue was carried out, with no subsequent recurrence or suppuration.

In all 15 cases of recurrence following closed techniques, an open procedure was carried out, in one case with simultaneous tympanoplasty. Results were: elimination of the cholesteatoma and suppuration in ten cases; elimination of the cholesteatoma but persistence of suppuration in three ears, reappearance of the cholesteatoma in two cases.

Functional reconstruction: the middle ear ossicular system was intact in nine ears; in a further 39 ears with tympano-ossicular damage no functional reconstruction was attempted. Of the seven remaining cases, reconstruction was carried out in six cases simultaneously with cholesteatoma removal. In the seventh ear, reconstruction was carried out five years after the initial surgery.

Of the seven cases of tympano-ossicular reconstruction, two were open cavities (type III tympanoplasties) and five closed cavities. In one of these, a stapes was interposed between the footplate and malleus, as the latter was normal while the incus was destroyed. In two of the five ears, a type III reconstruction was carried out, with columelization from the eardrum to the footplate in further two cases. In the patients in whom functional

TABLE V
SITUATION OF THE AUDIOMETRIC GAP AFTER SURGERY

	No reconstructed ossicular chain	Reconstructed
Bigger gap	6 (12%)	0
Same gap	31 (65%)	3 (42%)
Smaller gap	9 (19%)	4 (57%)
Cochlear hearing loss	2 (4%)	0

reconstruction was carried out, pre-existing air-bone gaps of 20 and 30 dB were maintained in the two cases subjected to open techniques (Table V), whereas in the closed techniques four ears improved 15–30 dB and the fifth stayed the same as before surgery—with frequent otorrhoea secondary to an allergic state.

Discussion

Our experience agrees with that of other authors in that cholesteatomas do not appear during the first three to four years of life (Ruedi, 1958). Our earliest cases were diagnosed at the age of four. This suggests that the process requires time to develop and lead to clinical symptoms, although it is not possible to establish at what point in time, prior to diagnosis, development of the disease begins, or whether it is even present at birth. On the other hand, we found the incidence to increase markedly after nine years of age, with a maximum incidence at age 11.

Tubal dysfunction is known to be common among children and this probably plays a role in the appearance of CC. In our series, 50 per cent of cases exhibited soft tissue damage—of which 29 per cent were posterosuperior in the middle ear—suggesting a relationship between negative pressure secondary to tubal dysfunction and retraction pockets that finally generate cholesteatoma (Olaizola, 1985).

Mention has also been made of the iatrogenic role played by the use of transtympanic ventilation tubes (Charachon *et al.*, 1984; Edelstein *et al.*, 1988). These had only been fitted previously to one of the patients in our series, in the anteroinferior quadrant. At the time of the diagnosis a marginal posterosuperior perforation was noted—an interconnection between the two being improbable in view of the topographic discrepancy involved.

Much as been said of the close relationship in adults between the degree of mastoid pneumatization and incidence of cholesteatoma. However, this relationship is

TABLE VI
PERCENTAGE OF RESIDUAL CHOLESTEATOMA AND THE TECHNIQUE USED

Author	Year	Open	Closed
Abramson	1977	35	9
Austin	1977	29	15
Bellucci	1977	23	7
Brown	1982	34	12
Charachon <i>et al.</i>	1980	49	27
Chila and Schroeder	1980	30	15
Cody and Taylor	1977	35	22
Deguine	1978	50	—
Fombeur <i>et al.</i>	1980	25	8
Glasscock <i>et al.</i>	1981	36	—
Lacher	1978	31	12
Mallea <i>et al.</i>	1983	29	17
Marquet	1982	11	—
Palmgren	1979	15	6
Portman <i>et al.</i>	1984	11	8
Sade <i>et al.</i>	1981	27	13
Sanna <i>et al.</i>	1984	21	—
Schwartz	1984	23	—
Sterkers and Sterkers	1980	13	—
Vase	1981	18	—
Wayoff <i>et al.</i>	1982	44	20
Wright	1977	15	—
Our series	1991	37	13

not so apparent in childhood (Palva *et al.*, 1977; Jahnke, 1982). In fact in our series a sclerotic mastoid was observed in only 45 per cent, while 18 per cent were diploic and 23 per cent well pneumatized—two cases corresponding to intact eardrums. The contralateral ear (free of disease) presented pneumatization as good, or better than the operated ear in all.

The disease process exhibited marked aggression in our series; the incus was damaged in 70 per cent of cases, the malleus in 43 per cent and the stapes in 38 per cent. Multiple ossicular lesions were therefore very frequent; in only 25 per cent of all ears, was a single ossicle affected, whereas in 30 per cent two ossicles were damaged, and in 28 per cent all three were involved.

Three cases (5.5 per cent) presented with mastoiditis. Two other cases (3.6 per cent) showed lesions of the horizontal semicircular canal, demonstrated intraoperatively. There were no cases of facial nerve paralysis or intracranial complications, this being difficult to compare with the corresponding figures in adults, due to the few cases studied. In other series, the values vary for the lateral semicircular canal between 2.6 per cent (Olai-zola, 1985) to 12.2 per cent (Charachon *et al.*, 1984), whereas facial nerve involvement varies from 1 per cent (Sheehy, 1985) to 1.7 per cent (Jahnke, 1982). As indicated by Olai-zola (1985), this limited incidence of complications may be due to the fact that CC tends to evolve over a short time span, whereas the above complications are more common in prolonged processes such as may recur in adult cholesteatoma.

We found that the process shows a marked tendency towards affecting the attic (79 per cent), antrum (57 per cent) and posterior recesses (67 per cent), with a much more limited involvement of other structures. Even residual cholesteatomas tend to present at the same sites. In this aspect we agree with the reports of others (Charachon *et al.*, 1984; Olai-zola, 1985), with the exception of the tympanic cavity involvement, which tends to be higher (Sanna, 1983).

Three ears presented with a cholesteatoma behind an intact eardrum (5.4 per cent)—two with a normally pneumatized mastoid bone. According to Edelstein *et al.* (1988), CC originates in the anterior region of the tympanic cavity and preferentially extends towards the anterior attic and petrous apex; symptomatology is scarce compared with acquired cholesteatoma, which originates and develops in the posterior middle ear, attic and antrum—with clear clinical manifestations. On the contrary, three of our patients presented cholesteatomas occupying the attic, antrum and posterior portion of the tympanic cavity, with hearing loss as the sole symptom—and no otorrhoea.

Open techniques have the disadvantage of the necessity of periodic cleaning by the specialist, together with occasional suppuration that may go on for years and is due to deficient epithelization of the cavity or to tubal dysfunction. Their main advantage is the lower percentage of residual cholesteatomas observed. Closed techniques present opposite characteristics. As mentioned earlier, authors such as Palva *et al.* (1977), Andrieu-Guitrancourt *et al.* (1980) and Jahnke (1980) prefer open techniques to treat CC. Conversely, Glasscock *et al.* (1981), Charachon *et al.* (1984) and Sheehy (1985) favour closed approaches. We have always performed a

closed technique in those cases in which complete removal of the cholesteatoma could be fully visualized and controlled; otherwise, we have not hesitated in using an open technique—always bearing in mind the need to preserve hearing in these patients while at the same time securing complete removal of the cholesteatoma and reduction of the risk of residual cholesteatoma.

The percentage incidence of residual cholesteatoma varies greatly according to different authors, and depends on the type of technique employed (Table VI). The results reported in the literature fail to clarify whether CC is more aggressive and present with more recurrences than adult cholesteatoma.

Moreover, the comparison between open and closed techniques can never be very rigorous, as the former tend to be employed with more extensive disease, whereas the closed approaches are reserved for more localized presentation. Our figures for CC point to 37 per cent residual cholesteatomas after closed techniques versus 13 per cent after open methods. Globally, these figures are somewhat greater than the mean values in adults, but are still within the normal range of values.

Conclusions

CC requires time to develop, being the earliest cases diagnosed at the age of three to four years. There is a tendency towards affecting the attic, posterior recesses and antrum. The first and main symptom in CC is otorrhoea either alone or associated with hearing loss.

In our hands closed show better results than open techniques, but residual cholesteatoma occurs in a higher rate in closed techniques. On the other hand open techniques have the disadvantage of the necessity of periodic cleaning.

From our point of view the type of technique used depends upon two main points: surgeon's experience and cholesteatoma extension. Our goal is to perform a closed technique only when complete removal of cholesteatoma can be guaranteed by fully controlled visualization.

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Address for correspondence:
 Prof. Jaime Marco,
 Departamento de Cirugía,
 Facultad e Medicina,
 Av. de Blasco Ibañez 17,
 46010 Valencia,
 Spain

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