

Surgery for acquired cholesteatoma in children: long-term results and recurrence of cholesteatoma

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Abstract

The aim of the study was to evaluate the long-term results after surgery for acquired cholesteatoma in children and to contribute to the search for predictors of recurrence. During a 15-year period, 114 children underwent surgery. The patients were re-evaluated with a median observation time of 5.8 years. At the last re-evaluation 85 per cent of the ears were dry with an intact drum. Recurrence of cholesteatoma developed in 27 ears. The cumulated total recurrence rate was 24 per cent using the incidence rate calculation, applying Kaplan-Meier survival analysis the corresponding recurrence was 33 per cent. Recurrent disease occurred significantly more frequently in children younger than eight years, with a negative pre-operative Valsalva, with ossicular resorption and with large cholesteatomas. In conclusion, young children with poor Eustachian tube function and a large cholesteatoma with erosion of the ossicular chain, are at special risk of recurrence and should be observed for several years after surgery.

Key words: Otitis media, suppurative; Cholesteatoma; Child; Surgery, operative; Recurrence

Introduction

One of the main problems in cholesteatoma surgery is residual and recurrent cholesteatoma. Residual cholesteatoma develops from a remnant of keratinized epithelium left behind at initial surgery (Gyo *et al.*, 1996), and is believed to be closely related to the type, location and extension of the original cholesteatoma, the operative technique as well as the skill of the surgeon (Smyth and Fisch, 1985). Recurrent cholesteatoma develops from a retraction of the membrane, and is believed by some to be closely related to impaired middle ear ventilation (Sade, 1993).

The rate of total recurrency of cholesteatoma (residual and recurrent cholesteatomas) is reported to vary from seven to 70 per cent (Jansen, 1978; Shuring *et al.*, 1990). One reason for the great variability in the reported recurrence rates may be due to the statistical method applied. In this study the analysis of recurrence has been accomplished by applying both the standard incidence rate method as well as the Kaplan-Meier survival analysis which has been used in some recent publications (Rosenfeld *et al.*, 1992; Parisier *et al.*, 1996).

Most authors agree that the recurrence rate is higher in children than adults (Smyth, 1976; Brown, 1982; Vartiainen, 1995), but some authors found no correlation between recidivism and age (Edelstein and Parisier, 1989; Rosenfeld *et al.*, 1992). Vartiainen (1995) found a higher recurrence rate in ears

discharging at the time of operation than in dry ear. Rosenfeld *et al.* (1992) found that ossicular erosion was the only significant predictor of cholesteatoma recurrence. Tos and Lau (1989) found different recurrence rates in different types of cholesteatoma, they were lowest in attic cholesteatoma and highest in tensa retraction cholesteatoma.

The aim of the present study was to evaluate the long-term results after one-stage cholesteatoma surgery in children, and to contribute to the search of predictors which may predispose to recurrence of cholesteatoma, in order to find the children who are predisposed to developing recurrence. Finally we focused on the impact of the applied statistical method for the outcome of the results.

Subjects and method

During a 15-year period, January 1980 to December 1994, 114 children, younger than 15 years of age, consecutively underwent one-stage surgery for acquired cholesteatoma. In January 1996, all the patients were called in for a re-evaluation. Ninety-nine patients (87 per cent) attended the re-evaluation. The median observation time was 7.3 years, (range 1.0 to 16.1 years). The 15 patients who did not attend the re-evaluation were included with the actual follow-up period, because the median observation time for the 15 patients was 1.7 years (range 1.0 to 5.7 years). There was no significant difference in age distribution or total recurrence rates between

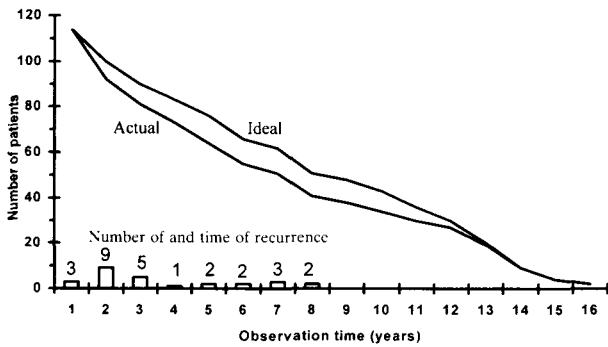


FIG. 1

Ideal and actual follow-up in 114 cholesteatomas, and time of recurrence.

the attendee and non-attendee group. For all 114 patients the median observation time was 5.8 years (range 1.0 to 16.1 years) (Figure 1). The median age at operation was 9.0 years (range 3.5 to 14.9 years).

The material included 33 attic, 45 sinus and 36 tensa retraction cholesteatomas. All patients underwent one-stage individualized surgery including myringoplasty with fascia and ossiculoplasty with autologous ossicles or cortical bone. Type I tympanoplasty with intact chain was conducted in 41 cases, type II with interposition in 49 cases with defective incus, and type III with a columella in 24 cases with resorbed incus and stapedial arch. In 16 ears with attic cholesteatoma, the modified intact wall mastoidectomy (Tos, 1982) was performed, in 14 ears canal wall down and in three ears the cholesteatoma could be removed endaurally by atticoantrotomy only. The sinus cholesteatoma could be removed through the ear canal during tympanoplasty without mastoidectomy or atticotomy in 26 ears. In 19 ears mastoidectomy was performed, of these the canal wall was removed in five cases. In the tensa retraction cholesteatoma, the cholesteatoma could be removed through the ear canal without mastoidectomy in 23 ears (64 per cent). The canal wall down technique was necessary in two ears in order to eradicate the cholesteatoma.

Because the length of follow-up on these ears varied, the standard incidence rate calculation method of recurrence does not reflect the true risk of recurrence, as only those patients observed for that specific period are included in the calculation. In order to overcome this statistical problem we applied

Kaplan-Meier survival analysis which is designed so that all patients, despite the length of follow-up are included in the calculation of the recurrence risk. For comparison reasons, all long-term recurrence rates have been calculated using both two statistical methods (Moeschberger and Klein, 1995). The Cox regression test has been used to analyse the correlation of recurrence between the different predictor groups. The chosen level of significance was $p < 0.05$.

Results

Re-operations

In total, re-operations had been performed in 35 out of 114 ears (31 per cent). In eight ears it was because of residual cholesteatoma, in 17 ears because of recurrent cholesteatoma, in eight because of re-perforation and in two ears because of poor hearing. In seven ears re-operation was performed twice (three cholesteatoma, two perforations and two because of poor hearing). In two ears a recurrent cholesteatoma was detected at the re-evaluation and was scheduled for a re-operation.

Recurrence of cholesteatoma

Residual cholesteatoma occurred most frequently in sinus cholesteatomas (Table I and Figure 4). In six out of eight ears the residual cholesteatoma developed from the tympanic cavity and in two out of eight ears in the attic. The median detection-time for residual cholesteatomas was 1.7 years (range 0.6 to 7.8 years).

Recurrent cholesteatoma was most frequent in attic cholesteatomas (Table I and Figure 4). In most cases the recurrent cholesteatoma developed from a retraction in the attic and in only three out of 19 ears from an extension of the retraction of the posterior part of the tympanic membrane into the tympanic cavity. The median detection-time for recurrent cholesteatomas was 2.8 years (range 1.5 to 7.7 years). The total recurrence rate (residual and recurrent cholesteatoma) was 24 per cent using the standard calculation method(s) and 33 per cent, applying Kaplan-Meier analysis (k-m) (Table I). In both methods of analysis the highest recurrence rate was found in the attic cholesteatoma group, and the lowest in ears with tensa retraction cholesteatomas. In all cases the recurrence was detected within eight years after the initial surgery (Figure 2 and Figure 3).

TABLE I

LOCATION OF RESIDUAL AND RECURRENT CHOLESTEATOMA CALCULATED BY STANDARD INCIDENCE RATE (S) AND BY KAPLAN-MEIER (KM)

Cholesteatoma type	Residual				Recurrent				Total recurrence	
	Tympanic cavity		Attic		Tympanic cavity		Attic		n	s/km %
	n	s/km %	n	s/km %	n	s/km %	n	s/km %		
Attic (33)	–	–/–	1	3/3	–	–	9	27/41	10	30/44
Sinus (45)	6	13/16	–	–/–	2	4/6	4	9/14	12	27/36
Tensa (36)	–	–/–	1	3/3	1	3/4	3	8/14	5	14/21
All (114)	6	5/7	2	2/2	3	3/3	16	14/21	27	24/33

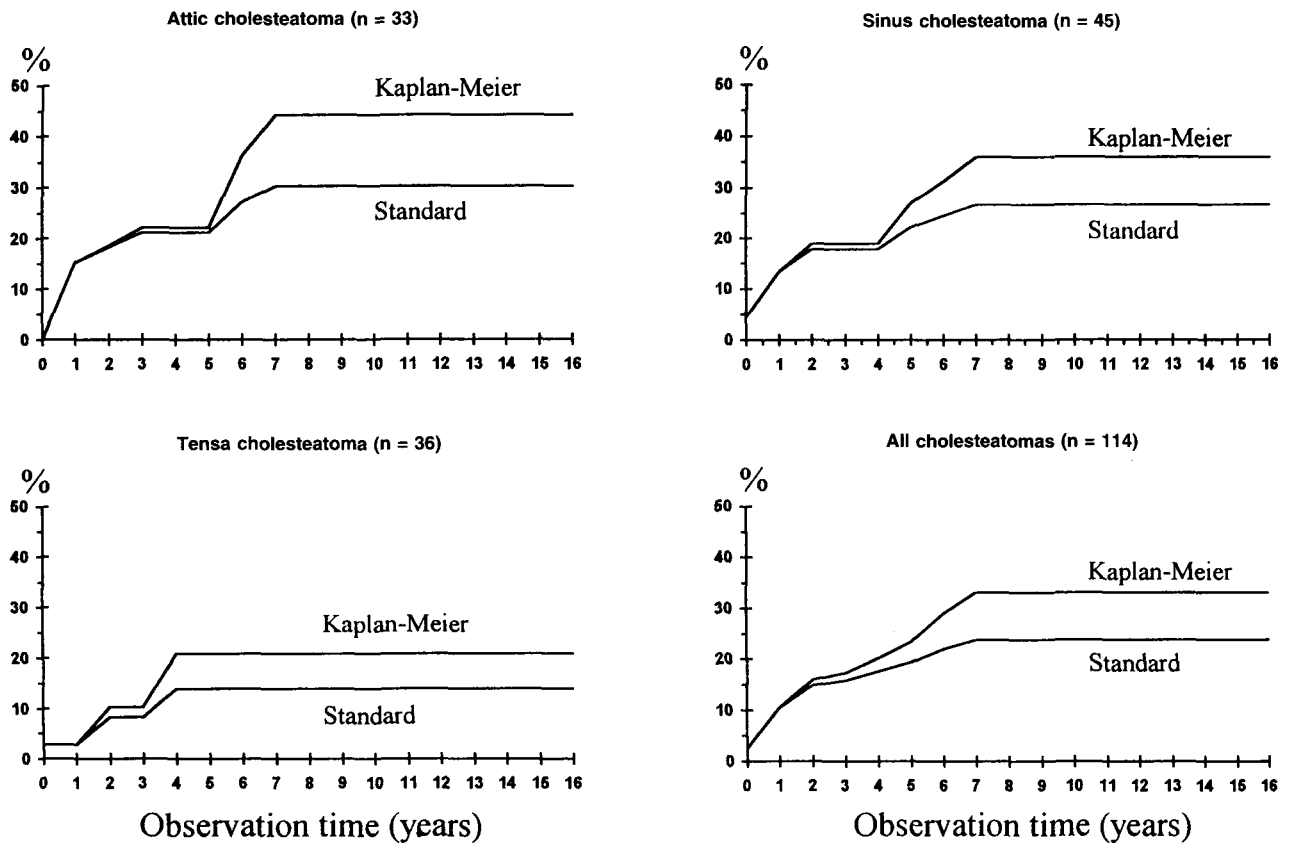


FIG. 2

Cumulated total recurrence in different types of cholesteatoma, calculated by the standard method and by the Kaplan-Meier survival method.

In three ears two re-operations were necessary to eradicate the cholesteatoma. In two ears, not previously re-operated, recurrent disease was detected at the re-evaluation three and 4.5 years after the initial operation.

Type of mastoidectomy and recurrency

Of the 114 patients, 44 had canal wall up, 21 canal wall down and the cholesteatoma was removed without mastoidectomy in 49 ears (Table II). In the

canal wall up group, the highest recurrence rate was found in attic cholesteatomas (Table II). Recurrence rates were higher in the canal wall up than the canal wall down mastoidectomy, using the standard calculation method wall up had a 39 per cent recurrence rate, compared to 19 per cent recurrence using the canal wall down method. In contrast when applying Kaplan-Meier analysis, the highest recurrence rate was found in the canal wall down group (64 per cent) compared to 45 per cent in the canal wall up group. In ears with no mastoidectomy, total recurrence developed in 12–14 per cent (s/k-m) of the ears.

Extension of the cholesteatoma

The extension of the cholesteatoma was classified into four locations, attic, antrum, tympanic cavity and mastoid process. The tympanic cavity was involved in all cases of sinus and tensa retraction cholesteatomas since these originate from the tympanic cavity. In 33 out of 45 ears (73 per cent) with sinus and in 32 out of 36 ears (89 per cent) with tensa cholesteatoma, the cholesteatoma was confined to the tympanic cavity only. The sinus cholesteatoma did not extend into the mastoid process in any case and, in only one out of 36 ears (three per cent) with tensa cholesteatoma was the mastoid involved. In contrast, the cholesteatoma extended into the mastoid in 24 per cent of ears with

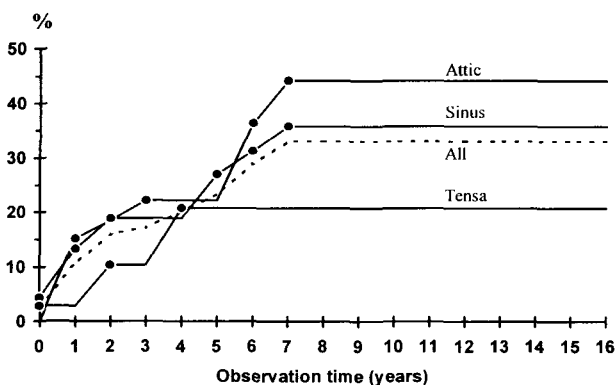


FIG. 3

Cumulated total recurrence in different types of cholesteatomas calculated by the Kaplan-Meier survival method. Dots indicate time of recurrence.

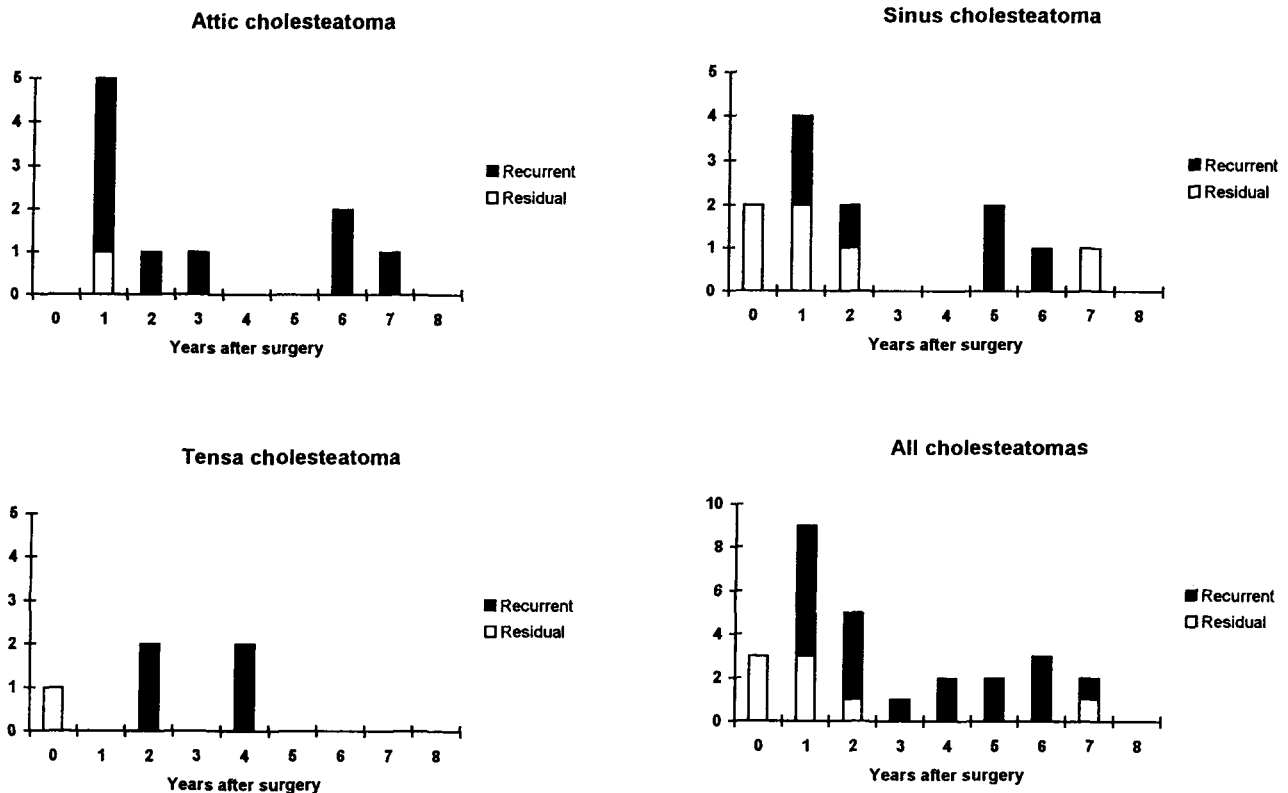


FIG. 4

Time of occurrence of residual and recurrent cholesteatoma in different types of cholesteatomas

attic cholesteatoma (Table III). For statistical calculations all cholesteatomas were classified into cholesteatomas limited to one location only, and cholesteatomas extending into two or more locations. In the attic cholesteatoma group only two were confined to one location only. In tensa cholesteatoma the distribution was the opposite where 32 out of 36 cholesteatomas extended into one location only (Table III).

Extension of the cholesteatoma and recurrence

In attic cholesteatomas there was no significant difference in the recurrence rate between small and large cholesteatomas. In sinus and tensa cholesteatomas there were a highly significant increase in recurrence in ears with cholesteatoma extending into two or more locations compared to ears with a small cholesteatoma (Table III). The total recurrence rate

for ears with large cholesteatomas, analysed with method (k-m) was 63 per cent and 38 per cent when using method (s). In ears with small cholesteatomas (one location only) the corresponding figures were 16 per cent and 13 per cent respectively (Table IV).

Condition of the ossicular chain

Resorption of the ossicular chain was most frequently found in attic cholesteatoma where all types of resorption were seen (Table V). Involvement of the malleus was found only in attic cholesteatoma. The incus body was partly resorbed in 15 out of 33 attic cholesteatomas and in only two out of 45 ears with sinus cholesteatoma, in tensa retraction cholesteatoma, resorption of the incus body was not seen. Resorption of the long process of the incus was seen in all types of cholesteatoma (55–69 per cent), most frequently in sinus cholestea-

TABLE II
TYPE OF MASTOIDECTOMY RELATED TO TOTAL RECURRENCE (R) USING STANDARD INCIDENCE RATE (S) AND BY KAPLAN-MEIER (KM)

Cholesteatoma type	Canal wall up		Canal wall down		No mastoidectomy	
	n/r	s/km %	n/r	s/km %	n/r	s/km %
Attic (33)	19/8*	42/49	14/2	14/14	-/-	-/-
Sinus (45)	14/5	36/42	5/2	40/100	26/5	19/20
Tensa (36)	11/4	36/44	2/-	-/-	23/1	4/8
All (114)	44/17	39/45	21/4	19/64	49/6	12/14

*In three ears atticoantrotomy only was performed, one had recurrence.

TABLE III
EXTENSION OF CHOLESTEATOMA AND RECURRENCE

Extension of cholesteatoma	Type of cholesteatoma			All (114) n/r
	Attic (33) n/r	Sinus (45) n/r	Tensa (36) n/r	
At	2/1			2/1
At + An	3/-			3/-
At + An + Tc	18/6	3/-	1/-	22/6
At + An + Tc + Ma	6/2		1/1	7/3
At + Tc	2/-	3/2	2/1	7/3
At + An + Ma	2/1			2/1
An + Tc		6/5		6/5
Tc		33/5	32/3	65/8
Only one location	2/1	33/5	32/3	67/9
Two or more locations	31/9	12/7	4/2	47/18

Attic (At), Antrum (An), Tympanic cavity (Tc), Mastoid process (Ma), Recurrence (r).

toma. Total resorption of the stapedial arch was found in nine per cent, 20 per cent and 14 per cent in the attic, sinus and tensa cholesteatomas respectively.

Condition of the ossicular chain and recurrency

There was a significantly higher ($p < 0.05$) recurrence rate in ears with some degree of resorption of the ossicular chain, compared to ears with an intact chain (Table IV).

Age at the operation and recurrence

At the time of the operation 42 children were younger than eight years and 72 children were eight years or older. Recurrence occurred significantly more frequently in the younger age group, where 36 per cent had recurrence, compared to 17 per cent in the older children. Applying method (k-m) the recurrence rate for the two age groups was 47 per cent and 24 per cent respectively (Table IV).

Pre-operative middle ear ventilation and recurrence

From the patient's journal the result of the Valsalva test was noted in about half of the cases. In those cases where the Valsalva test was not mentioned, the middle-ear ventilation was consid-

ered positive in case of normal middle-ear pressure or airfilled middle ear at one of the pre-operative otomicroscopy. If not recorded the middle-ear ventilation was considered negative in case of retracted tympanic membrane and effusion at surgery.

In each of the cholesteatoma groups, there was no significant difference in the recurrence rate of cholesteatoma between ears with negative and positive middle ear ventilation at the time of operation. In the entire cholesteatoma group however, significantly more children with negative Valsalva developed recurrence, compared to children with a positive pre-operative Valsalva (Table IV).

Ear discharge at operation and recurrence

At the time of operation 62 ears were dry and 52 ears discharging. Although recurrence occurred in 27 per cent of the dry ears and in 43 per cent of discharging ears (k-m), we could not demonstrate any significant difference between the two groups (Table IV).

TABLE IV
PREDICTORS OF RECURRENCE OF CHOLESTEATOMA CALCULATED BY STANDARD INCIDENCE RATE (S) AND BY KAPLAN-MEIER (KM)

Predictive factors	Number/ recurrence n/r	Total recurrency		p value	
		s %	km %		
Extension	Only one location	67/9	13	16	$p < 0.01$
	Two or more locations	47/18	38	63	
Chain	Intact	38/5	13	16	$p < 0.05$
	Resorbed	76/22	29	42	
Age	Younger than 8	42/15	36	47	$p < 0.05$
	8 years or older	72/12	17	24	
Valsalva	Positive	47/6	13	15	$p < 0.05$
	Negative	67/21	31	42	
Condition	Dry	62/13	21	27	n.s.
	Discharging	52/14	27	43	

TABLE V
CONDITION OF THE OSSICULAR CHAIN IN DIFFERENT TYPES OF CHOLESTEATOMA RELATED TO RECURRENCE (R)

Ossicular pathology	Type of cholesteatoma			
	Attic (33) n/r	Sinus (45) n/r	Tensa (36) n/r	All (114) n/r
Mh + Ib	2/-			2/-
Mh + Ib + Lp	5/2			5/2
Mh + Ib + Lp + Ts	1/-			1/-
Ib	5/4			5/4
Ib + Lp	1/1			1/1
Ib + Lp + Ps	1/-			1/-
Ib + Lp + Ts		2/2		2/2
Lp	4/1	21/6	13/-	38/7
Lp + Ps	4/1	1/1	2/1	7/3
Lp + Ts	2/-	7/1	5/2	14/3
Intact chain	8/1	14/2	16/2	38/5
Resorbed chain	25/9	31/10	20/3	76/22

Malleus head (Mh), Incus body (Ib), Long process (Lp), Partial (Ps) and Total stapes crura (Ts).

Otomicroscopic findings at the re-evaluation

At the re-evaluation 85 ears (86 per cent) were dry, with an intact drum and with no sign of recurrence. Three ears had discharging cavities, nine ears had a perforation of the drum and in two ears a recurrent cholesteatoma was detected (Table VI). In the group with canal wall up mastoidectomy one ear had a perforation and in one ear a recurrent cholesteatoma was found, this ear had 4.5 years previously been operated for tensa retraction cholesteatoma. All ears with canal wall down mastoidectomy were dry and perforation or recurrence of cholesteatoma was not seen in any ear.

Hearing results, pre-operatively, post-operatively early and at re-evaluation

Related to the type of cholesteatoma, the poorest pre-operative hearing, as evaluated by absolute hearing, was found in ears with tensa retraction cholesteatomas (Table VII). In ears with an intact chain the median absolute hearing was 25 dB compared to 38 dB in ears with total resorption of the stapes. The difference in median absolute hearing between ears with a positive and negative Valsalva was small, 28 dB and 32 dB respectively.

The hearing results obtained post-operatively (one to six months after surgery) are shown in Table VII. The best hearing was obtained in ears with an intact chain (15 dB) whereas the greatest hearing gain

obtained was seen in ears with tensa cholesteatoma (16 dB).

There was no difference in the pre- and early post-operative median absolute hearing in ears with total resorption of the stapes.

At the re-evaluation the median absolute hearing was better than 20 dB in all types of cholesteatomas (Table VII). Compared with the primary results, the hearing was stable and even with a small improvement up to the last follow-up in all three types of cholesteatomas. The stability of the hearing results up to the last evaluation was significantly ($p < 0.05$) higher in ears with a positive pre-operative Valsalva, where 77 per cent had absolute hearing better than 20 dB, compared to ears with a negative Valsalva where only 49 per cent had a hearing better than 20 dB.

Discussion

In the present study the priority of the surgery has been a complete eradication of the cholesteatoma during the initial surgery. None of the procedures were staged, but the children were followed up over time and underwent revision surgery only when recurrence was suspected. Recurrence of cholesteatoma developed up to eight years after the surgery. We found the overall recurrence rate, depending on the statistical method, to be between 24 and 33 per cent others have reported the rate of cholesteatoma recidivism to vary from seven to 70 per cent (Jansen,

TABLE VI
OTOMICROSCOPIC FINDINGS IN 99 PATIENTS AT THE RE-EVALUATION

Cholesteatoma	No major pathology		Discharging cavity		Perforation of the drum		Recurrence of cholesteatoma		
	Type	n	n	%	n	%	n	%	
Attic	31	29	94	1	3	1	3	-	-
Sinus	37	31	84	1	3	4	11	1	3
Tensa	31	25	81	1	3	4	13	1	3
All types	99	85	86	3	3	9	9	2	2

TABLE VII

EARLY AND LATE HEARING RESULTS RELATED TO TYPE OF CHOLESTEATOMA, CONDITION OF OSSICULAR CHAIN AND PRE-OPERATIVE VALSALVA

	n	Absolute hearing level (mean of 500–2000 Hz)		
		Pre-operatively	Post-operatively	Last evaluation
		median	median	median
Attic cholesteatoma	33	30	18	17
Sinus cholesteatoma	45	29	22	18
Tensa cholesteatoma	36	34	18	18
Intact chain	37	25	15	15
Stapes present	51	30	18	18
Stapes absent	24	38	38	28
Pre-operative Valsalva	47	28	17	15
Positive				
Negative	65	32	18	22

1978; Schuring *et al.*, 1990). In this series we found the extension of cholesteatoma to be the most important predictor of recurrence and in decreasing order; negative pre-operative middle ear ventilation and ossicular chain resorption. In agreement with others we found a significantly higher recurrence rate in young children where 47 per cent had recurrence, compared with children eight years old or older (24 per cent), this finding is consistent with the aggressive nature of childhood cholesteatoma, additionally the poorest middle ear ventilation and the highest rate of ossicular resorption was seen in this group. The reason for the high recurrence rate in the mastoidectomy group, may be that ears which required mastoidectomy were those with the most extensive cholesteatomas. In ears with no mastoidectomy only two out of 52 ears had large cholesteatomas, compared to 25 out of 41 ears in the canal wall up group and 20 out of 21 in the canal wall down group. Looking at the extreme groups, the total recurrence rate in a child younger than eight years, with a negative pre-operative Valsalva, resorption of the ossicular chain and a large cholesteatoma was 100 per cent. However, in children older than eight years, with a positive pre-operative Valsalva, intact ossicular chain and a small cholesteatoma there was no recurrence.

In this study where the length of follow-up of the patients varied from one to 16 years, half of the subjects have been observed for a shorter period than it may take to develop recurrence, especially recurrent cholesteatoma, therefore the standard incidence rate calculation method does not reflect the true risk of recurrence, since only those patients observed for that specific period are included in the calculation. The Kaplan-Meier survival analysis is designed so that all patients are included in the calculation of the recurrence risk. In other studies where Kaplan-Meier survival analysis on the cholesteatoma recurrence have been applied, the reported recurrence is the high rate of 40 to 70 per cent (Rosenfeld *et al.*, 1992; Parisier *et al.*, 1996). In the present study where we have applied the Kaplan-Meier survival analysis parallel to the standard method, it is seen that the difference between the

calculated rates in all cases is higher, especially when estimating the cumulative rate of recurrent cholesteatomas. When applying Kaplan-Meier to the cumulative rate of residual cholesteatomas the results accomplished by the two methods are almost identical. The reason for this is that most of the residual disease occurs few years after surgery, when there is still a high number of patients being observed, in contrast to most cases of recurrent cholesteatomas which typically occur after a longer observation time, where the number of patients is reduced (Figure 4). In our next study we will discuss the problem and correlation of applied statistical methods to length of post-operative observation, number of patients, detection-time of recurrence and follow-up rates.

Conclusion

Classification of cholesteatoma according to pathogenesis seems essential in order to discover the predisposing factors for recurrence of cholesteatoma. A young child, with poor middle ear ventilation, a large cholesteatoma and resorption of the ossicular chain is at special risk of developing recurrent disease and should be followed by the otologist for several years post-operatively. In contrast, a child older than eight years, with a positive Valsalva, intact ossicular chain and only a small cholesteatoma, is unlikely to develop recurrent disease.

The choice of statistical method for calculation of recurrence rates is sensitive to the length of post-operative observation period and time of the recurrence. In cases where the observation period for the patients followed is shorter than the maximal detection time for recurrency, survival analysis should be the method of choice for calculation of recurrence rates.

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