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Tympano-ossicular allografts for cholesteatoma in children[1]

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Abstract

At the Sint Augustinus Hospital, Antwerp, Belgium, all children with cholesteatoma are operated by a canal wall up approach and immediate reconstruction with a tympano-ossicular allograft. In the majority of the cases, a second stage is performed after 1 year. This paper presents the results of a retrospective review of the charts of 103 consecutive children treated between 1979 and 1995. The mean patient age was 10 years and the mean postoperative follow-up was 4.5 years. In 28 children residual cholesteatoma was found at the second stage operation and 20 developed recurrent cholesteatoma in the course of time. So far no residual cholesteatoma surfaced after the staged procedure, but 11 children needed more than two operations to control recurrent disease. An intact, trouble-free graft was present in 79 children at the latest follow-up. The median postoperative bone-conduction thresholds were equal to the preoperative thresholds. The median postoperative air-conduction thresholds improved in 50% of the cases, remained unchanged in 25%, and deteriorated in 25% of the cases. It is concluded that the tympano-ossicular allograft technique is effective and safe and offers good anatomical and acceptable functional results. © 1997 Elsevier Science Ireland Ltd.

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1. Introduction

Management of cholesteatoma, in children as well as in adults, requires surgical intervention. The four goals of cholesteatoma surgery are: (1) to eradicate the disease; (2) to create a dry, trouble-free and 'water-proof' ear; (3) to maintain or improve hearing; and (4) to prevent recurrent disease. The most certain way to achieve the first goal is to remove not only the cholesteatoma sac, but also the affected tympanic membrane and ossicles. A meticulous anatomical reconstruction offers the best chances to achieve the second and the third goal. The last and most challenging aim is to prevent recurrent pathology. Given the still insufficient understanding of the pathogenesis of cholesteatoma, our success is inevitably incomplete. However, the authors believe that anatomical reconstruction of the middle ear allows its return to a physiological homeostasis, and that this is the best defense against recurrent pathology.

Basically, two surgical approaches are available to deal with cholesteatoma [1,17]. First, the canal wall down approach or open tympanoplasty, and second, the canal wall up approach or closed tympanoplasty. Many consider the first approach to be more effective in eliminating the disease, as this technique offers better visualization during surgery and recurrences are less likely to develop in an open mastoid cavity. The functional results, however, are considered to be better by the second approach. The closed technique offers better opportunities for ossicular chain reconstruction. Moreover, an anatomically normal postoperative ear does not require regular suction cleaning, children are allowed to swim and if necessary, a hearing aid can be worn.

The technique introduced by Jean Marquet, 1966 [10], consists of the meticulous dissection of the pathology by a canal wall up approach and immediate reconstruction of the middle ear with tympano-ossicular allografts. It allows both maximum visualization during the operation, as well as optimal possibilities for reconstruction such that the surgeon can be radical in the removal of any diseased structure. In recent years few results of this technique have been reported [4,8,14].

At the Sint Augustinus Hospital in Antwerp, Belgium, all children with cholesteatoma are treated by a closed tympanoplasty and allograft reconstruction. This study presents the results of this approach in 103 consecutive children treated for cholesteatoma between 1979 and 1995. Details on residual and recurrent disease, graft status and hearing levels will be reported.

2. Materials and methods

The charts of all consecutive children undergoing surgery for cholesteatoma at the Sint Augustinus Hospital Antwerp between 1979 and 1995 were reviewed retrospectively. Patients over 16 years of age at the time of surgery, and those followed up for less than 1 year postoperatively were excluded. In children with bilateral cholesteatoma ($n = 15$; 15%), only one ear was included randomly, in order to meet the statistical criterion of independency of data.

All children were operated according to Marquet's technique [10]. Ninety-six percent of the operations were carried out by three senior surgeons (J.M., F.E.O., T.S.). The surgical procedure started with a post-auricular skin incision and dissection of the skin of the bony ear canal. This was followed by excision of the tympanic membrane with its fibrous annulus and the neck of the cholesteatoma sac. A posterior tympanotomy was performed and 'following the pathology', the cholesteatoma was dissected from the mastoid and middle ear. Affected ossicles were removed. A tympano-ossicular allograft was placed, and the skin of the external ear canal was repositioned over the cuff of the allograft. Defects of the attic or posterior meatal wall were restored with remodeled autologous cortical bone grafts. The ear canal was packed for 1 week with synthetic sponges soaked in an antibiotic/steroid ointment for 1 week. A second look operation was usually planned after 12 months. In children with limited disease no second stage was planned, but, like all patients, they were regularly followed up by otomicroscopy.

Tympano-ossicular allografts were harvested and prepared at the tissue bank of the Sint Augustinus Hospital [10], according to the standards of the Belgian law (Belgisch Staatsblad 13.6.86). Immediately after removal from the cadaver, grafts were fixed for at least 2 weeks in a solution of 4% buffered formaldehyde. After dissection, tissues were preserved in Cialit (1:5000 aqueous solution of a sodium salt of an organomercuric compound, Hoechst Pharmaceuticals) for a period of 3 weeks to 2 months. Fig. 1 shows an example of an en-bloc tympano-ossicular allograft.

This study does not address the alleged risk of transfer of infectious diseases, such as Creutzfeldt-Jacob disease (CJD) and human immunodeficiency virus (HIV) infections. Transmission of CJD has been reported in 10 cases after implantation of dura mater grafts [11]. It has, however, never been reported after transplantation of tissues other than brain or cadaveric dura mater. In addition, the incidence of CJD is extremely low (1:1 000 000) and stringent criteria for donor selection should exclude donors at risk for CJD. No reports of transmission of HIV by non-vital allograft material have appeared in the literature. Formaldehyde, used in the preservation of allografts, is known to inactivate HIV readily [6].

Outpatient follow-up continued until adulthood, and always included an otomicroscopic examination. Pure-tone audiometry was planned at regular intervals. Audiometry was conducted in a sound-treated room using a Madsen Electronics OB 822 Clinical Audiometer calibrated according to ISO-standards (1987). Air- and bone-conduction thresholds were determined at 5 dB steps. Unless otherwise specified, hearing levels are reported as the pure-tone average (PTA, i.e. the average of the thresholds at 500, 1000 and 2000 Hz).

For the present analysis a standardized form was used to record the following variables: sex; age; surgeon; history of ear surgery; side and site of cholesteatoma [12]; preoperative hearing levels; type of allograft reconstruction; follow-up time; findings at follow-up in terms of state of graft; residual or recurrent cholesteatoma and hearing levels. No response to air-conducted sound was coded as 120 dB, no response to bone-conducted sound as 80 dB. Missing values were coded as such. Descriptive statistical analyses were performed on all variables. Counts, percentages, histograms, and box plots were used to describe nominal data. Hence, audiometric results (often expressed PTA, i.e. the mean of hearing levels at 500, 1000 and 2000 Hz) were described by means of non-parametrical statistics. Fisher exact tests with Yates correction were used to study the dependence of recurrent or residual cholesteatoma on all the other variables. A Kruskal–Wallis analysis of variance was performed to study the dependence of the audiometric results on the different variables. All tests were carried out at a 5% significance level.

3. Results

One hundred and three children were included in this study. In Tables 1 and 2 some of their characteristics are summarized.

Figs. 2 and 3 show the occurrence of residual and recurrent cholesteatoma in our population following initial reconstructive surgery. According to Brackman [2] residual cholesteatoma is defined as epithelium left behind at the primary surgery that has regrown into a cholesteatoma in contrast with recurrent cholesteatoma which is defined as a newly developed cholesteatoma arising from an attic or posterior superior retraction pocket.

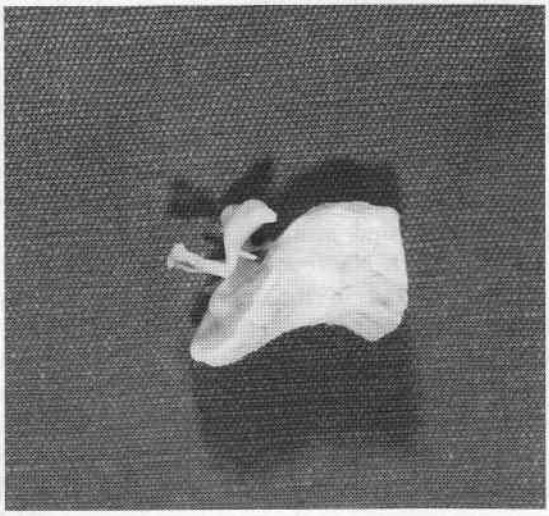

Fig. 1. Tympano-ossicular allograft

Table 1
General patient characteristics $n = 103$

Mean age	10 years (range 4–16 years)
Sex	65 male; 38 female
Previous surgery at another centre[a]	17 children
Mean follow-up	4.5 years (range 12–191 months)

[a]Surgery varied from a myringoplasty to extensive surgery for cholesteatoma.

In nine children (9%) the disease was controlled in a single stage. A planned two-stage procedure was performed in 83 cases (81%). Eight children (8%) underwent three operations, two children (2%) had four operations, and one child (1%) had five operations because of recurrent cholesteatoma.

Fig. 4 shows the state of the tympanic membrane graft at the latest follow-up (average 4.5 years). A safe, trouble-free graft was present in 79 children (76%). An unsafe retraction, probably requiring further surgery was found in 12 children (12%), and in seven children (7%) the graft had failed, resulting in a perforation.

The hearing results are presented in Figs. 5 and 6 [7]. The median hearing levels at the latest follow-up were similar to those at 1-year-postoperative follow-up (Fig. 5).

Finally, variables that might predispose to either recurrent or residual disease were studied. The occurrence of residual or recurrent disease was not significantly correlated to age, sex, a history of middle-ear dysfunction, extension of cholesteatoma at initial surgery, quality of the middle ear mucosa, and aeration of the middle ear at initial surgery.

Table 2
Otologic characteristics $n = 103$

Side of cholesteatoma	
	49 left; 54 right
Site of cholesteatoma at preoperative otomicroscopic examination	
	32 Attic
	45 Posterosuperior quadrant
	10 Central part pars tensa
	16 Behind an intact TM
Allograft used for reconstruction in initial operation	
	17 TM only
	29 TM + M + I[a]
	47 TM + M + I + Sp[b]
	8 TM + Ir
	2 TM + other ossicular reconstruction

TM, tympanic membrane; M, malleus; I, incus; Sp, partial stapes (i.e. anterior crus + anterior half of the footplate); Ir, remodelled incus [9].
[a]This type of allograft is placed on an intact stapes.
[b]This type of allograft is placed on the footplate (absent stapes suprastructure).

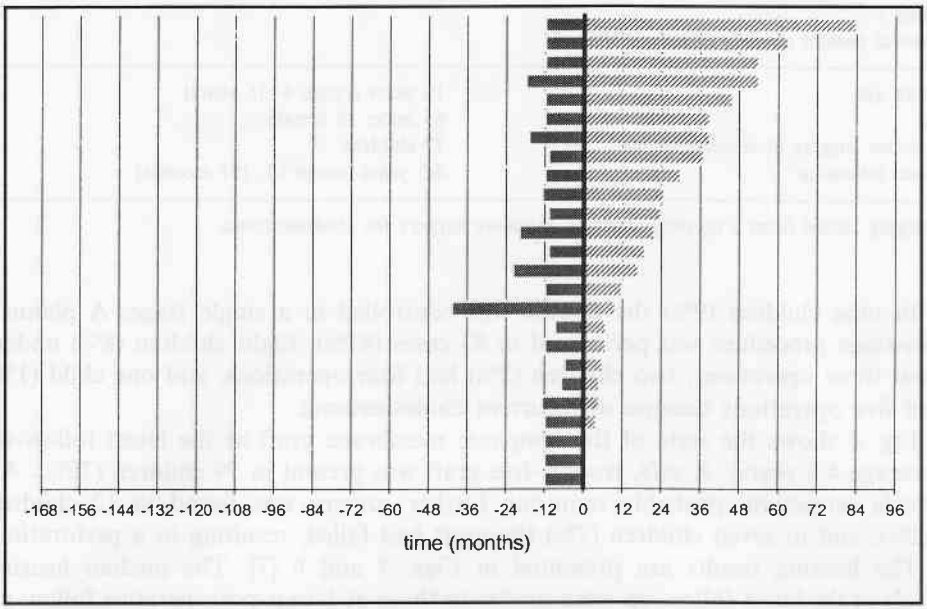

Fig. 2. Residual cholesteatoma ($n = 25/103$). The bold vertical line (time = 0) represents the second stage operation of 25 children at which residual cholesteatoma was found. The dark grey bars at the left hand side represent the time between the first and second stage operation. The light grey bars represent the follow-up time after the second stage operation.

4. Discussion

Reports on cholesteatoma surgery are often difficult to interpret as its results depend on a large number of variables. For a meaningful comparison of the results of one specific center to those of others, it is crucial that a detailed description of the study group and the surgical technique is provided.

This study focuses on the results of surgery in children. Consistent with Sheehy [17] and Brackmann [2] a cut-off age of 16 years was defined. Many have questioned whether cholesteatoma in children differs from the condition in adults, and if so, whether children require a different surgical technique than adults [1,15–17].

Even more than adults, children may benefit from the advantages of a closed technique. This study reports on the tympano-ossicular allograft technique for cholesteatoma in children and at present it is the standard technique of the Antwerp School of Otology. Since it is applied as the technique of choice in every case, this study is not biased by selection of the 'better' cases for a closed technique and the 'poorer' cases for an open technique.

The canal wall up technique is essentially a two-staged technique, so whether or not complete eradication of disease is achieved, may only be evaluated at the second stage. In 27% of children residual cholesteatoma was found at the second

stage operation. This corresponds to the results of other centers where a staged closed tympanoplasty technique is routinely performed in children and residual disease rates of 23–44% were found [2,5,15,17]. There is no point in comparing these figures with those obtained by an open technique [13]. It merely proves that a second look is essential in a closed technique. Since the two stages of the operation are considered to be part of one single treatment and no second residuals were observed in the present population one could say that in our series complete eradication of disease is achieved in 100% of cases. Therefore, the authors claim their closed tympanoplasty technique to be safe and effective.

The take rate of allografts at an average follow-up of 4.5 years was 93%. This is as good as the take rate of fascia grafts obtained by Sheehy [17] and Brackmann [2]

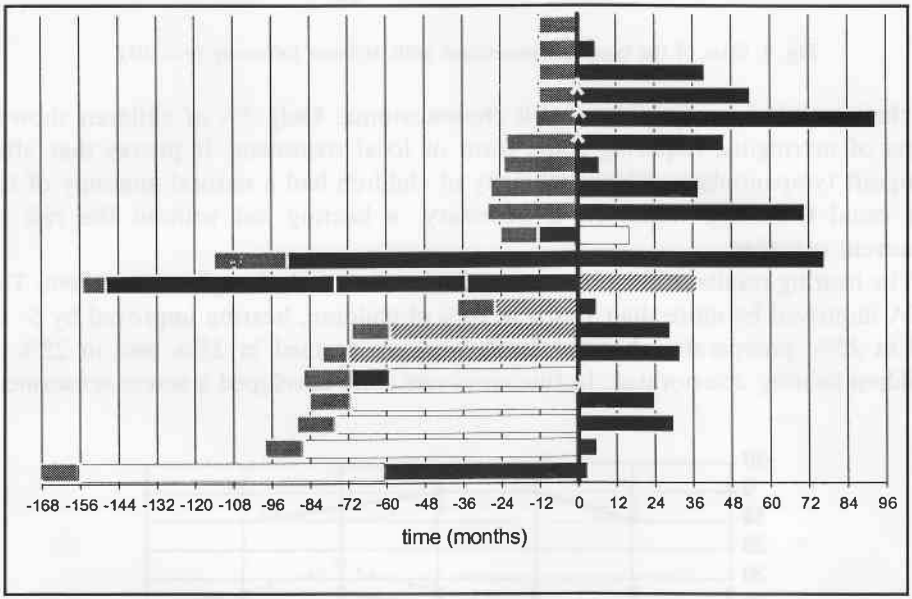

Fig. 3. Recurrent cholesteatoma ($n = 20/103$). The bold vertical line (time = 0) represents the latest surgical intervention of 20 children who developed recurrent cholesteatoma. The bars on the left hand side of this line refer to the operations and course of time up to this final operation. The bars on the right hand side refer to the follow-up time after the latest operation. The colour of the bars refers to the findings at the operation at the beginning of the bar: dark gray, first stage operation; black, recurrent cholesteatoma; light gray, residual cholesteatoma; white, no cholesteatoma found. For example bar number 11 from above represents a child who had recurrent cholesteatoma at the second stage operation 22 months after the first stage. At the third stage, 92 months after the second stage, again recurrent cholesteatoma was found. During the follow-up there after (77 months) there were no signs of recurrent disease. Bar number 17 from above represents a child who showed no residual or recurrent cholesteatoma at the second stage operation, 11 months after the first stage. After a follow up of 72 months she developed a recurrent cholesteatoma for which a canal down procedure was performed. She had a further follow-up of 24 months. * Canal wall down procedure. Note that in three children who developed a recurrent cholesteatoma (bars 13, 14 and 15 from above), a residual cholesteatoma was found at the second stage operation, these three children are not included in Fig. 2. This also applies to the child who had residual disease after three recurrences (bar 12 from above).

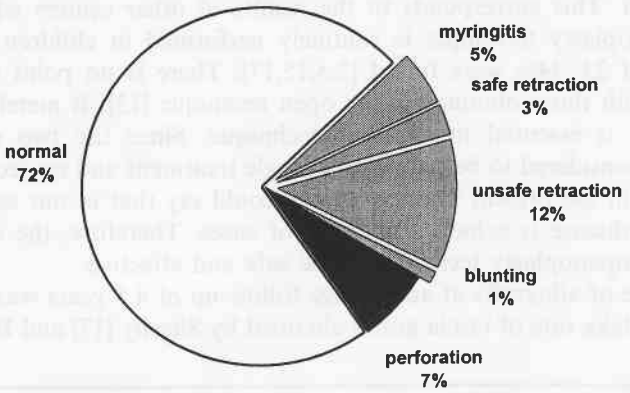

Fig. 4. State of the tympanic membrane graft at latest follow-up ($n = 103$).

in their population of children with cholesteatoma. Only 5% of children showed signs of myringitis, requiring some form of local treatment. It proves that after allograft tympanoplasty a large majority of children had a natural anatomy of the ear canal tolerating water or, if necessary, a hearing aid without the risk of recurrent otorrhea.

The hearing results of the present population are good, though not excellent. The PTA improved by more than 15 dB in 25% of children, hearing improved by 5–15 dB in 25%, preoperative hearing levels were maintained in 25%, and in 25% of children hearing deteriorated. In this series one child developed a severe sensorineu-

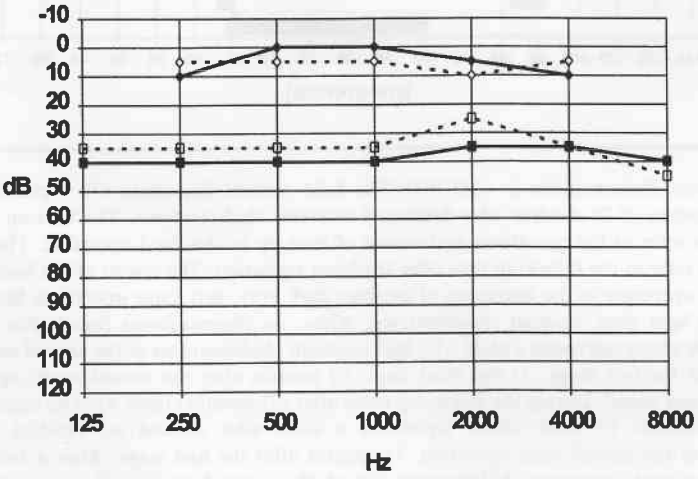

Fig. 5. Pre- and 1-year-postoperative median hearing levels in 90 children for whom full audiometric data were available. ◆, median preoperative bone-conduction threshold; ◇, median bone-conduction threshold at 1-year-postoperative evaluation; ■, median preoperative air-conduction threshold; □, median air-conduction threshold a 1-year-postoperative evaluation.

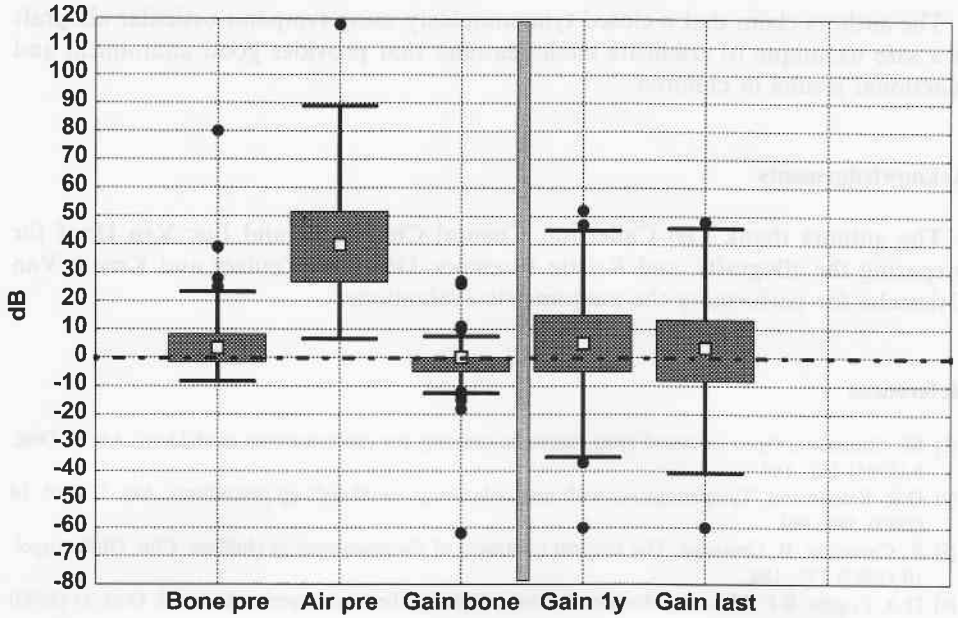

Fig. 6. Pre- and postoperative hearing levels in 90 children for whom full audiometric data were available. Box and Whisker plots [7]. The pre- and postoperative hearing thresholds (PTA) are shown ($N = 90$). Bone pre, preoperative bone conduction threshold; air pre, preoperative air conduction threshold; gain bone, difference between pre and post operative bone conduction threshold; gain 1 y, difference between pre and 1 year post operative air conduction threshold; gain last, difference between pre and latest post operative air conduction threshold; bars, minimum to maximum values; large rectangles, 25th–75th percentile; small squares, median value; dots, outlying values.

ral hearing loss. It showed major involvement of the labyrinth by cholesteatoma at surgery. The authors are not in favor of reporting hearing results in terms of air-bone gap closure [7]. For the purpose of comparison to other studies, postoperative air-bone gaps (postoperative air-conduction threshold minus preoperative bone-conduction threshold) were calculated. Closure of the air-bone gap to within 20 dB was obtained in 30%, and to within 30 dB in 50% of children. These results seem poorer than those reported by some others in surgery for childhood cholesteatoma [2,3,18], but are comparable to those reported by Parisier [13]. Some of the difference may be explained by the fact that all cholesteatomas, irrespective of their extension, and thus all ossicular reconstructions (in 47 ears the stapes crura were destroyed), were included in this analysis.

Finally, cholesteatoma may recur. In the present series this was found in 19% of children. With longer follow-up this figure is likely to increase [13]. Fig. 3 shows that recurrent cholesteatoma may develop years after the first intervention. This calls for regular and prolonged follow-up. If a child presents itself with a recurrence, the authors believe that a closed technique may be used again and may be advocated with the same arguments as those for primary surgery. In a small minority of children (4% in this study) the extent or aggressive behavior of the recurrent pathology urged the surgeon to perform an open technique.

The authors claim that a closed tympanoplasty using tympano-ossicular allograft is a safe technique to eradicate cholesteatoma that provides good anatomical and functional results in children.

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References

[1] M. Abramson, Open or closed tympanomastoidectomy for cholesteatoma in children, Am. J. Otol. 6 (1985) 167–169.
[2] D.E. Brackmann, Tympanoplasty with mastoidectomy: canal wall up procedures, Am. J. Otol. 14 (1993) 380–382.
[3] R. Carachon, B. Gratacap, The surgical treatment of cholesteatoma in children, Clin. Otolaryngol. 10 (1985) 177–184.
[4] D.A. Foggia, B.F. McCabe, Homograft tympanoplasty: the Iowa experience, Am. J. Otol. 11 (1990) 307–309.
[5] M.E. Glasscock, J.R.E. Dickins, R. Wiet, Cholesteatoma in children, Laryngoscope 91 (1981) 1743–1753.
[6] M. Glasscock, C. Jackson, G. Knox, Can acquired immunodeficiency syndrome and Creutzfeldt-Jacob disease be transmitted via otologic homografts?, Arch. Otolaryngol. Head Neck Surg. 114 (1988) 1252–1255.
[7] P.J. Govaerts, T. Somers, F.E. Offeciers, The use of Box and Whisker plots for the graphical presentation of the audiometrical results of treatment of conductive hearing loss, J. Otolaryngol. Head Neck Surg. 1997, in press.
[8] E.P.P.M. Hamans, P.J. Govaerts, T. Somers, F.E. Offeciers, Allograft tympanoplasty type 1 in the childhood population, Ann. Otol. Rhinol. Laryngol. 105 (1996) 871–876.
[9] S. Janssens de Varebeke, P.J. Govaerts, T. Somers, F.E. Offeciers, The 'two-hole' ossiculoplasty technique, Laryngoscope 106 (1996) 507–510.
[10] J. Marquet, Twelve years experience with homograft tympanoplasty, Otolaryngol. Clin. North Am. 10 (1977) 581–593.
[11] J. Martinez-Lage, M. Poza, J. Sola, J. Tortosa, P. Brown, L. Cervenakova, J. Esteban, A. Mendoza, Accidental transmission of Creutzfeldt-Jacob disease by dural cadaveric grafts, J. Neurol. Neurosurg. Psychiatry 57 (1994) 1091–1094.
[12] R.P. Mills, N.D. Padgham, Management of childhood cholesteatoma, J. Laryngol. Otol. 105 (1991) 343–345.
[13] S.C. Parisier, M.B. Hanson, J.C. Han, A.J. Cohen, B.A. Selkin, Pediatric cholesteatoma: an individualized, single-stage approach, Otolaryngol. Head Neck Surg. 115 (1996) 107–114.
[14] A. Perrin, R. Mendoza, Ch. Baril, P. Receveur, P. Roulleau, Résultats anatomiques a long terme des homogreffes tympaniques. A propos de 170 cas, Ann. Oto-Laryngol. (Paris) 104 (1987) 535–539.
[15] M. Sanna, C. Zini, R. Gamoletti, P. Delogu, A. Russo, R. Scandellari, A. Taibah, The surgical management of childhood cholesteatoma, J. Laryngol. Otol. 101 (1987) 1221–1226.
[16] H. Schmid, J.C. Dort, U. Fisch, Long-term results of treatment for children's cholesteatoma, Am. J. Otol. 12 (1991) 83–87.
[17] J.L. Sheehy, Cholesteatoma surgery in children, Am. J. Otol. 6 (1985) 170–172.
[18] E. Vartiainen, J. Nuutinen, Long-term results of surgery for childhood cholesteatoma, Int. J. Ped. Otorhinolaryngol. 24 (1992) 201–208.