Aggressiveness of Pediatric Cholesteatoma. Do We Have an Evidence?

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Abstract To compare pediatric and adult patients, affected by cholesteatoma for the clinical presentation, disease extent and final outcome. This is a prospective study in which 60 cases of unsafe chronic suppurative otitis media with cholesteatoma were included. These cases were divided into two groups of 30 cases each on the basis of age. All patients were subjected to detailed clinical examination. A high resolution computed tomography of the temporal bone was done in all the cases. An innovative grading system was devised to grade the disease. The patients were subjected to canal wall down mastoidectomy and were evaluated postoperatively for resolution of the symptoms, hearing and cavity problems. Granulation tissue along with cholesteatoma was more common among children ($p < 0.01$), whereas cholesteatoma alone was more common among adults. There was excellent correlation between the radiological and surgical findings. It was seen that higher the grade of the disease, the greater was the risk of complications and recidivism ($p < 0.05$). Recidivism was significantly higher in children ($p < 0.05$) whereas complications were equally common in both the groups. Hearing improvement was equivocal in both groups. Presence of granulation tissue along with cholesteatoma in children causes more aggressive disease with higher recidivism rate.

Keywords Cholesteatoma · Pediatric cholesteatoma · High resolution computed tomography · Temporal bone

Introduction

The goal of treatment in cholesteatoma is total eradication of the disease to obtain a safe and dry ear. If left untreated, this entity can give rise to various intra- or extra cranial complications. The success of any given approach may be measured in terms of the rate of recidivism. Cholesteatoma in children is widely considered to be a more aggressive disease than in the adult population for two major reasons. First, very extensive disease is found more frequently in children compared to adults [1] and secondly, higher rates of residual and recurrent disease have been documented in the pediatric population [1–6]. The literature is replete with the studies on the comparative evaluation of the pediatric and the adult cholesteatoma [2–5, 7–9]. Most of these studies have been performed in a retrospective manner. This study is an attempt at prospective analysis of pediatric and adult cholesteatoma.

Materials and Methods

This prospective study was conducted in the Department of Otorhinolaryngology, Head and Neck Surgery, Nehru Hospital, Postgraduate Institute of Medical Education and Research, Chandigarh, India, during the period from January 2007 to June 2008. The study was approved by the Institute Ethics committee. All the consecutive patients ($n = 60$) attending the Otolaryngology out patients with a diagnosis of atticofurrow chronic suppurative otitis media with cholesteatoma were included in the study. Patients

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with past history of otological surgery in the same ear, or with coexistent canal atresia and/or microtia and cases not willing for surgery or follow up were excluded from the study.

These cases were divided into two groups; group A and B of 30 cases each on the basis of age with group A having cases less than 14 years and group B more than 14 years of age. All the patients were subjected to a detailed clinical and neuro-otological examination. Audiological evaluation included pure tone audiometry (PTA) in all the cases. They were subjected to radiological evaluation with a high resolution computed tomography (HRCT) of the temporal bone with 1.25 mm sections in both axial and coronal planes. The patients were subjected to canal wall down mastoidectomy and tympanoplasty whenever suitable under General Anesthesia. They were evaluated postoperatively for resolution of the symptoms, cavity healing and hearing improvement using PTA at the end of 3 months.

All the patients were instructed to come for follow up after 1 week and then every month for a minimum period of 3 months. The minimum follow up was 3 months and maximum was one and half year postoperatively. During these visits, patients were assessed for subjective improvement in the hearing and condition of the mastoid cavity.

Statistical Analysis

Statistical analysis was done to analyze the data using χ^2 test, Fisher's exact test, Mc Nemar test and Kendall's tau-c test.

Results

The male to female ratio in the pediatric age group was 6:1 while that in adults was 5:1. The age ranged from 5 to 14 years in group A with an average age of 10.97 ± 2.428 years while that in group B it ranged from 18 to 59 years with a mean age of 25.3 ± 7.008 years. The disease was bilateral in 7 of the 30 cases (23.3 %) in children while in adults it affected both ears in 9 cases (30 %).

The severity of hearing loss was categorized according to the WHO Classification. Moderate loss was most common in both groups with 11 (39.28 %) and 12 (41.37 %) patients in group A and B respectively. This was comparable in both groups (insignificant with $p > 0.01$ as per χ^2 test).

The most common type of disease was primary acquired cholesteatoma with 20 (66.67 %) in group A and 22 (73.33 %) patients in group B presenting with it. One each had tertiary type and congenital type in group A while the rest had secondary acquired type (26.67 %). This was comparable in both groups (insignificant with $p > 0.01$ as per χ^2 test).

Intra operatively the pneumatization of the mastoid air cells was different in both the groups. Cellularity was

found to be more extensive in the pediatric group (66.67 %) while in the adults it was mostly sclerotic (83.33 %). This was statistically significant ($p < 0.01$) as per χ^2 test (Fig. 1).

Presence of granulations and cholesteatoma together was commonly observed in children i.e. 18 (60 %) cases whereas in adults both cholesteatoma and granulations were present in 16 (53.33 %) cases. with 13 cases (43.33 %) presenting with cholesteatoma alone (Table 1).

Presence of granulation tissue along with cholesteatoma was more common in children whereas cholesteatoma alone was more common in adults which was statistically significant ($p < 0.005$ as per χ^2 test).

We graded the radiological and surgical findings according to the criteria shown in Table 2.

Radiological Findings

A HRCT scan of the temporal bone was performed in 23 (76.67 %) cases in both groups. It was observed that in both the groups grade II disease was more common with 13 (56.52 %) cases in group A and 11 (47.82 %) in group B having grade II disease.

Surgical Findings

Using the same grading system for surgical findings, it was again found that grade II disease was more common with 20 (66.67 %) cases in both groups having these findings. There was excellent correlation between radiological and

Fig. 1 The distribution as per pneumatization of the mastoid

Table 1 Showing intraoperative findings

	Cholesteatoma alone	Both cholesteatoma and granulations
Group A	6	18
Group B	14	16

Table 2 The radiological grading

Grade	Findings
I	Disease in attic, aditus, antrum
II	Erosion of one bony wall of antrum or disruption of the ossicular chain
III	Erosion of two of the bony walls of antrum with ossicular chain disruption
IV	Intracranial extension, dehiscence of facial canal or the LSCC

surgical findings which was seen in 18 (82.60 %) and 17 (73.91 %) of the cases in group A and B respectively and this was statistically significant ($p < 0.05$ as per χ^2 test) (Fig. 2) with the correlation co-efficient using Kendall's tau-c test value being 0.510 in group A and 0.618 in group B. Using our grading system, we found that recidivism and the risk of complications was more with the higher grade of the disease radiologically ($p < 0.05$) (Fig. 3).

The distribution of patients with both intra cranial and extra cranial complications in both the groups is shown in Table 3. The type of complications present were comparable in both groups (insignificant with $p > 0.01$ as per χ^2 test).

At 6 weeks post operatively the cavity status was comparable in both groups (insignificant with $p > 0.01$ as per χ^2 test). However as the duration progressed i.e. at 3 and 6 months post operatively it was observed that the healing was better in the adults (significant with $p < 0.05$ as per χ^2 test) (Fig. 4).

Fig. 2 Correlation between surgical and radiological findings **Fig. 3** The correlation between radiological grading and recidivism

Recidivism was found only in group A with 3 children having residual disease and 2 having recurrent disease and this was found to be statistically significant ($p < 0.05$).

Operative Complications

We saw one major complication in one patient who had facial paresis which recovered completely by 3 weeks post operatively. Two children developed post aural wound dehiscence in children which required secondary suturing.

Discussion

Cholesteatoma is considered to be more aggressive in children by many [1–6] but there is still a lot of debate on this

Table 3 The distribution of complications in both groups

	Group A	Group B
Facial palsy	3	3
Labyrinthine fistula	4	3
Extracranial abscess	2	2
Intracranial complications	4	3
Temporal lobe abscess	0	1
Meningitis	1	–
Cerebellar abscess	3	2
Total	13	11

Fig. 4 Comparison of mastoid cavities at 3 and 6 months post operatively

topic. This present study analyzed the various aspects of pediatric cholesteatoma compared to adult cholesteatoma.

We found that males were more commonly affected in both the pediatric and adult group, ratio being 6:1 and 5:1 respectively. This is in agreement with studies by Glasscock et al. [1], Olszewska et al. [10], Dornelles et al. [11] and Panda et al. [12] which also showed a male predominance. On the contrary, Edelstein et al. [8] reported more females than males.

We did not find any difference in the duration of the disease in both pediatric and adult cholesteatoma. This is in contrast to findings of other authors like Glasscock et al. [1] who found that duration was longer in adults than in children. Palva et al. [5] reported that average duration of the disease in children was 5.8 years whereas in adults it was 21.8 years which is considerably longer.

The incidence of neuro-otological complaints such as vertigo, tinnitus and earache was comparable in both groups. This is contradictory to the study by Edelstein et al. [8] who found a higher incidence of neuro-otological symptoms in adults. Disagreeing with this author, Darrouzet et al. [13] suggested that fewer complications in the pediatric group were due to the fact that in this group the duration of the disease is on an average below what is found in adults.

We used HRCT of the temporal bone to evaluate the extent of the disease pre operatively using an innovative

grading system designed by us. An excellent correlation between the radiological and surgical findings was observed.

We also found that the higher the grade of the disease the higher were the chances of complications and recidivism. This grading system can thus help us in the prognostication of the disease and deciding our treatment. We should be more aggressive in treating the disease of a higher grade. The HRCT was also very sensitive in detecting residual disease. It detected recidivism in five out of six cases which was later confirmed intra operatively during revision surgery. This is in agreement with other studies in literature which found CT to be the imaging technique of choice for evaluation of residual disease [14, 15].

Intra operatively we observed that the mastoid of children was more cellular when compared with that of adults which was often found to be sclerotic. This is in agreement with other studies by Palva et al. [5] and Glasscock et al. [1]. Presence of granulations points to increased osteitis with more propensity to destroy the bone especially the more cellular mastoid air cell system leading to aggressive and extensive disease in children. We found granulations along with cholesteatoma to be more common in children than adults. However this difference was not significant. Panda et al. [12] also reported in which they also found a higher incidence of granulations alone (53.3 %) in the pediatric age group. Mallet et al. [16] reported more aggressiveness of pediatric cholesteatoma based on the immunohistochemical use of M1B1 antibody in pediatric cholesteatoma. Dornelles et al. [17] compared various markers like CD 31, MMP2, and MMP9 in pediatric and adult cholesteatoma patients. They reported more exacerbated inflammatory response by production of metalloproteinases which could be the cause for aggression of pediatric cholesteatoma.

Presence of complications has no bearing on the age of the patient. This is in contrast to other studies who found a higher rate of complication amongst adults [1–3, 8, 5]. Panda et al. [12] found a higher incidence of complications in the younger age group. On the contrary Glasscock et al. [1] found the incidence in adults to be more than double with 16 % as compared to the 7 % in children. The latest study by Moody and Lambert [18] also showed the incidence to be higher among adults.

We found an increased incidence of fistula in children (16.7 %) than in adults (6.7 %) with the lateral semicircular canal being most commonly involved. This is contrary to other studies in the literature showing a higher incidence of labyrinthine fistulae in adults [1–3, 8, 6]. The most commonly involved canal was the lateral semicircular canal as was the case in other studies in literature [19, 20]. However in the study by Glasscock et al. [1] they found superior canal to be the most commonly affected.

Among intracranial complications, our series showed maximum incidence of brain abscess which is in agreement

with the study by Panda et al. [12]. On the contrary, Samuel et al. [20] and Gower and McGuirt [21] reported meningitis to be a more common complication. This could be attributed to the inclusion of cases of acute suppurative otitis media in their series, which, when aided by aggressive pathogens like *Haemophilus influenzae*, have a high propensity for development of meningitis.

The rate of recidivism was seen to be 16.6 % in our series. These results are comparable to those of Edelstein and Parisier [2] but much lower than those reported by Glasscock et al. [1] and Sheehy et al. [3]. The rate of residual cholesteatoma after canal wall down mastoidectomies ranges from 6 to 72 % [14]. Darrouzet et al. [13] found that it was more with canal wall down procedures. Ahn et al. [22] also reported increased recidivism in patients less than 8 years old. The authors advised a long term close follow up in these children.

The inefficient cavity care due to poor cooperation and difficulty in keeping the cavity clean in children maybe a cause for higher incidence of recidivism. Another reason for the low incidence of recidivism in adults could be due to the relatively shorter period of follow up (maximum 1 year). A study by Glasscock et al. [1] had noted that in adults the majority of recurrence makes itself known after 12 months, while in children, it will manifest within 6–12 months.

Conclusion

To conclude, we found a higher incidence of granulations along with cholesteatoma among children which may be the reason for the more aggressive disease in this group with higher rate of recidivism.

Conflict of Interest There are no conflicts of interest.

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